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The complexity of psychiatric care, from pregnancy to adolescence: Beyond the endogenous-exogenous dichotomy

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The complexity of psychiatric care, from pregnancy to adolescence: Beyond the endogenous-exogenous dichotomy

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Babies in therapy, psychoanalytic interventions for infants and their parents

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Babies in therapy: The study "Psychoanalytic interventions for infants and their parents" conducted on infants through the lens of psychoanalysis and clinical work with both parents and infants all contribute to our knowledge of the nature of early relationship disorders. Psychoanalytic theory's concepts of the depressive position, early defense mechanisms, transference, and psychosomatic reactions to depressive emotions are shown to be crucial in clinical cases, giving therapists new tools for intervention and increasing efficiency. Psychoanalysts have researched the long-lasting effects of early disappointments and the sense of being helplessly abandoned; they emphasize that a disruption in the relationship with the caregiver can produce a psychic economy oriented on the avoidance of anxiety, leaving less energy for development. New parents and their sick infants can benefit from early therapies with therapeutic potential and the possibility of preventing future narcissistic pain issues if they are based on psychoanalytic thinking and knowledge of early symptoms.

KEYWORDS

psychoanalysis, depression, primitive anxieties, dyadic therapy, playfulness

Introduction

Clinical work with parents and babies, psychoanalytic observation of infants, and studies of infancy all contribute to our understanding of how early relationship disorders develop. This paper focuses on the infant's inner world and how some classical and modern Freudian concepts can help the analyst working with dyads. These ideas range from the presumption of primary narcissism to the drive theory, which links libido development to the need for self-preservation and the resulting tensions, conflicts, anxieties, and defense mechanisms. The baby's primitive internal development is understood to be a complex unconscious dynamic process.

The profound effect of early disappointments and the feeling of being helplessly abandoned has been studied by many psychoanalysts, who emphasize that a disruption in the relationship with the caregiver can cause a psychic economy oriented on the avoidance of anxiety, leaving less energy for development.

Joint therapies provide both the suffering mother and child with support in dealing with each other's narcissism; depressive meltdowns in toddlers are always attacks on the integrity of the self that can have serious developmental consequences. For this reason, early and rapid intervention is essential when signs of concern are observed in toddlers, but it is still difficult to know the long-term influence of such interventions. Primary breakdowns have far-reaching effects on people's lives, from early childhood disorders, such as developmental delays or attention difficulties, to serious obstacles to accessing the oedipal conflict, damage in adolescence to traumatic recurrences in adulthood. Even if they fully recover after receiving therapy, some people will still have a problem with the motor and behavioral discharge, cutting off the transitional stakes necessary to sustain the preconscious functioning that would allow for a cognitive activity to form. From the first few months of life, there are disturbing behavioral patterns. I will talk about situations in which libidinal movements are not invested and the risks they pose to the development of the subject.

The narcissistic wound the child feels poses a threat of primary depression, whether due to the mother's postpartum depression, difficulties with the child's attachment, or the baby's difficult temperament.

Lack of vitality and depressive reactions in infancy

At the beginning of life, these wounds leave indelible marks: distress, collapse, the feeling of being let down, of falling vertiginously without hope, the feeling of spreading, of liquefying without contact. These feelings of despair, inscribed in the body as much as in the psyche, are sources of narcissistic damage that can sometimes be repaired.

According to the axis of depressed posture, infant depression can be classified into two clinical entities.

The depressive position described by Melanie Klein (1) is a normal process in the infant's development: it is a stage from which the child enters a relationship with an object and considers himself independent and distinct from his mother. If the threat is the loss of this separate object, which is accompanied by feelings of ambivalence and guilt, it is also the first step to creating a stable and permanent internal object, even in its absence.

During the establishment of this depressive position, the pain of the loss may be too acute and trigger the manic defense. Afterward, the problem of loss and separation, with sadness and mourning, is in the foreground. This is a time when we can share with the child an intermediate space where we are two distinct and empathetic individuals. The therapist will act as an impartial third party by allowing the mobility of the fantasmatic representations through plays, interpretations, and comedy, which encourage libido circulation.

Because of the narcissistic nature of this investment, such as the nipple in the mouth is a part of the self, the loss is first experienced as a loss of a partial object, such as the breast or the oral sensory experience of being with the mother, which is felt as catastrophic and irretrievable. Before experiencing depressive effects, infants experience terror, the anguish of annihilation, the suffering of being able to only survive without being lively, and psychosomatic disorganization. The stake is then the constitution of an identity, a continuous, stable, and well-organized Ego, which can accommodate various perceptions.

As seen in his "Still Face" experiment from 1975, in which he had mothers of typically developing 3-month-olds maintain a rigid and expressionless expression, Tronick was able to observe the baby's expression of helplessness, a depression in the making (2):

The baby looks at his mother and smiles. The mother remains impassive. The baby intensifies his look, stretches his arms, and frowns. Then, the baby turns away for a few seconds and returns toward the still-frozen mother. Faced with the failure of its attempts at interaction, the baby tires, yawns, looks at its mother, and "ends up curling up in an attitude of powerlessness, face turned away, body motionless." This interaction does not last more than 3 min and shows the infant's vulnerability. Mothers interviewed after this experience reported how much they experienced their child's agitation and anxiety with a feeling of sadness, anger, and despair.

I quote Winnicott (3): "A slight defect in holding...gives the child a sense of endless falling."

"The depressive position, classically, opens on the mourning of the omnipotence to have its needs satisfied, on the renunciation of the desire without limits... Partial renunciation, we all know it well! But before being dispossessed of the fantasy of omnipotence, it is still necessary to have experienced it!" As demonstrated by Winnicott, object relations are established in the first few months of life. These ties are founded on the child's awareness of a world outside themselves in which they can take action and engage in reciprocal trade. The mother maintains the illusion that the child has unlimited power over the world. The child's belief in their own agency and cognitive capacity is undermined by this narcissistic involvement in the world as a source of gratification. This belief is essential to developing thought in real life. Therefore, at the start of life, building a strong connection to the real, physical world is essential.

Maternal care's inevitable failures and discontinuities are always felt as narcissistic losses. The child learns that the maternal container has its faults and limits, which are real threats to an infant easily deprived of its state of original distress. Observations of babies with their mothers show how much most of the defects of mutual adjustment can be corrected and how much the dyads know how to reconcile after a misunderstanding. The adjustment between needs and satisfactions, desires and gratifications is never perfect. The mother fails because of too much anticipation or too much delay. She can only be "good enough." Disappointments and unmet expectations, as well as tensions and conflicts stemming from the child's inherent tendency toward individuation, are woven into the fabric of the child's daily life. The maternal excitability barrier is always partial. These recurrences can have a cumulative effect and take on significant traumatic value.

Depressive anxieties are frequent and transient in babies, as observations of infants at home clearly show. They are generally repaired by the quality of the psychic and physical holding. However, unpleasant reactions to smiling, making eye contact, psychosomatic disorders, and withdrawal symptoms might develop if this bonding experience is lacking, whether from the mother or from the child.

In the very first months of life, the child does not suffer from losing an object. The relationship is still too fragile. An infant is in the slow process of differentiating between self and object, and the quality of the object's investment, which is typically still precarious, will be affected.

To establish a solid connection, it is crucial to have frequent sessions. The so-called anaclitic depression prevents children from engaging in sufficient cerebral stimulation. These solemn infants, who never smile and lack any vital tonus, are typically stuck in the infernal repetition of self-calming actions, signaling a transition from autoerotic games to functioning. As Bowlby (4) and Winnicott (3) pointed out, each in their own way, it is not the separation that leads to the baby's depression; it is the loss of hope. An object that has disappeared and been rendered inaccessible during rapid weaning, for example, an interaction with an inanimate mother without vocalizations or looks, or an illness or a surgical operation, can suppress the relationships with the object and leave the body and the psyche destitute. The infant is atonic, withdrawn, slowed down, and rapidly disorganized on the psychosomatic level and can present the pathological defenses described by Selma Fraiberg (5): the avoidance of eye contact, the paralysis of behavior or freezing, and the transformation of effects when, instead of manifesting distress or anxiety, an abused baby manifests a noisy joy and a disorganized excitement, which is called self-aggression.

Between birth and 6 months of age, a baby's "depressive" behaviors are more a response to the collapse of the tonus of life, to the loss of the possibility of investing, than to the loss of an object that is not yet solidly established. As we hold him in our arms, we see a newborn who has lost his skills, isolated from others and himself, with a frozen face, without laughing or smiling, without joyful expression in the eyes; he is rather indifferent, rarely surprised, who turns away and vocalizes very little (6). Illness, pain, and fatigue create these reactions and the loss of relationships between internal objects. At the beginning of life, the body envelope is still fragile, and to maintain the sensations and the experience of being alive through movements, the body's action is essential. The contacts with others help to constitute the physical unity of the body and the being. The feeling of being held, the touch, and the glances all leave physical and emotional imprints. A failure of the object's constitution is a failure of the subjects' constitution. Reestablishing a corporal dialogue is an emergency (7).

It is the economic aspect of the maternal psychic functioning that is communicated to the child in the dance of the preverbal games. The prosody, the rhythm of the enunciation as well as of the carrying, the intensity of the glances as well as the voice are essential signals to which the baby answers with his own register of primitive impulsiveness (8).

We cannot ignore, however, that depression does not exist without hatred; it is an early rage rather than an organized emotion, as Winnicott (3) shows. Let us not forget that Freud classified indifference as a special case of hatred. This intense and early development of negative movements requires an active mobilization of life movements on the part of the entourage. Early therapies are often emergencies.

The impact of intersubjectivity on the psychic development of the child and on the genesis of the sense of self, the first foundation of the Ego, is essential and can provide reference points to the analyst who works with infants. Intervening directly with a baby when the mother allows access to her child is immediately effective. If we think that the emergence of the first representations and the establishment of the processes of symbolization take place in interactive play, we can help a mother give the baby bodily anchoring of the experiences they share and pay attention to the sensations that the baby experiences without always being able to integrate them. The extraordinary appetite of babies for contact with others always facilitates the therapist's work. At the beginning of life, play is gestural. As such, it lays the groundwork for the child's future communication options and is crucial for developing emotional and affective sharing.

Primary depression is always associated with early trauma. When one's own survival is at risk, Childhood sexuality and its auto and hetero erotic investments are no longer appropriate. The risk is the libidinal loss, the loss of the quality of the libido connection, hence a desexualization of the psychic functioning. The loss of pleasure from autoerotic investment and the loss of the joy of a successful action are all possibilities if one's libido declines or if the quality of one's libido connection deteriorates.

The role of the capability for representation

If a nursing infant does not recognize himself in her mother's eyes, they will not be able to share in any joy together and will break the bond that has kept them connected to their mother's pleasure all along. There is a heterogeneity in the depression of infants and older children. In the beginning, the atony of investments dominates the reactions of withdrawal as an ultimate defense of self-preservation. It is in the depression of death that Palacio Espasa (9) observed how the relationships and the binders would disappear when the loss of a representation of the object is experienced as a catastrophe: losing the sense of oneself. A failure in the meeting with the mother is accompanied by a lack of memory trace of her, an absence of representation; hence, the ability to invest in an object is gravely compromised.

Winnicott (3) was the first to draw attention to the depressive suffering of what he called "psychotic depression," which occurs when the loss of an object is accompanied by the loss of a part of the self in a bodily experience: loss of the breast, loss of the mouth, a loss that cannot be elaborated into a psychic experience, leaving the subject destitute and in a state of helplessness.

In psychoanalytic therapies, the goal is to restore the dyadic creative drive through which the infant receives from the mother's skin the senses of smell, taste, warmth, color, security, and maintenance between loving and erotic attachments. A mother with the genius to collect her child's experiences into a cohesive whole will provide the impression of one's own emotional experience. Let's look at the first smiles of babies when they address others: how much the baby seeks to make contact and to seduce.

The feeling of continuity and permanence of the self, what Balint (10) called "fundamental confidence," is a framework that supports narcissism, giving it its qualities of confidence and quietude or distrust and greed. This feeling of internal security depends on the primary relationship to the object, the quality of the pleasure taken in the exchange, and what is appropriate in autoeroticism. Sucking in anticipation of breastfeeding is proof of this confidence in oneself and the environment. Active motions at taking in are essential to feeding and tolerating helplessness and passivity. As soon as the environment lacks that support, powerlessness is felt, and the pleasure of desire disappears. A defensive system is then set up, where the threatened ego immediately solicits aggressiveness in an attempt to safeguard the limits of the ego actively. The therapeutic response is to make a narcissistic alliance that counterbalances the insecurity to make the relationship viable and tolerable: it is necessary to reanimate, to reactivate the movements of life and linkages in the transference, and to restore the ego and its capacity for confidence.

In the danger of de-objectalization, offering a psychic and physical envelope, firm and flexible, is essential: it would be an introjectable container for the baby, provided that the rhythmicity of the tuning at the time of the care allows a true gathering of the senses, a transforming action of the impulsive flow as Bullinger (11) proposes. This shows the importance of direct interventions with infants whose mothers interact inadequately.

The therapeutic process modifies the psychic economy. The therapist's game engages and mobilizes the infant's interest, keeps his body active, and stimulates the development of the related fantasy.

The triangulation associated with the presence of the therapist in front of the dyad, or the mobilization of the paternal presence at the insistent invitation of the analyst, modifies the libidinal investment of the baby for a "non-mother" character who has an essential anti-depressive function by modifying the constellation of anxieties and defenses that characterizes the 6-month-old child at the moment when he reaches the depressive position. The father builds a bridge between the mother and the kid; The father, not as another maternal object, but as different, and containing his wife, creates a bridge between her and the child; it is he who allows the establishment of links.

Using an outside authority figure, such as a therapist who acts as a father figure, is cathartic because it releases pent-up libidinal energy, makes room for fresh investments, and kick-starts stagnant development stifled by the narcissistic system (12).

The role of play

The great play models are organized the absence: the fort da the hide-and-seek are games of controlled loss. The discovery and creation of games, pleasures of all kinds, and the sexualization of life always show an improvement in babies; the appearance of a smile in impassive infants and laughter in little ones who have finally become enthusiastic are reliable signs of change (13).

Stimulated by the environment, babies begin to laugh and become happy and excited at the age of 4 months. By the end of the first year, children begin to laugh on their own *via* their own actions and decisions, and it is from 18 months on that a real sense of humor (non-verbal) will emerge in incongruous and surprising situations.

I am not talking here about the laughter that relieves tension and has poorly organized effects or the big laughs of manic defense; I am referring to the association of pleasure and control in the sense of humor.

A 3-month-old baby

Anna is an almost 3-month-old girl, sullen and silent. Her mother is convinced that she will become autistic because she makes no sounds and smiles little. I know this mother well. She has had disability anxiety for each of her previous children and had to undergo a therapeutic abortion before becoming pregnant with Anna. The mother-daughter encounters are quite gentle around breastfeeding, but the exchanges of glances are poor and lost in anguish. I address the baby directly, telling her how her mother wanted a wonderful little girl but had lost confidence since the tragedy of the abortion, and I add, in a deliberately theatrical tone, that her mother is a woman of inordinate demands who wants perfect children; I surprise the mother, who starts laughing when I add that she has all the possible faults of a mother who idealizes the maternal function. I know that Anna is smiling with her father, and I tell her what a conflict it must be for the little girl to have a handsome father and an angry mother. Anna's mother, of course, reacts strongly and addresses her daughter in turn, saying how much she loves and values her baby, and her voice changes, becoming softer and more melodious. Anna turns to me and coos, then make eye contact with her mother and gets lost in her eyes, smiling. She then not stops babbling and becomes a very happy baby in a few weeks.

It is more difficult when the baby must face the negative effects of a rejecting mother. The threats of annihilation of the internal world are then intense. The persecutory experience is constant. These babies present obvious mood disorders. They are apathetic, inhibited, and always serious. Their development is restricted; attachment disorders take the lead very quickly, with a deficiency in bonds; these children can be entrusted to any stranger. They are neither fierce nor wild. The mother has not been able to be a mirror image of the child's emotions and has not been able to invest in the child as an object of love (14). The narcissistic damage is inevitable, as is the accompanying sense of guilt for having caused the argument with one's mother and a general inability to find peace in the world.

For a child's mind to develop, there must be something around that can hold it, comfort it through its pains, and challenge it just enough. Like a broken container, an uninteresting cosmos might trigger primal fears of abandonment and isolation. The excess of depressive suffering does not allow for the continuity and stability of the psychic structure; the introjective capacities of the baby are diminished; the developmental potentials cannot be realized and remain frozen in pathological identifications (15). The primary identification is with an object that ensures vital functions in a harmonious relationship, and this relationship allows psychic growth. Primary depression can be the equivalent of psychic death with proper defenses in place.

The baby has psychically withdrawn from the relationship. He has disengaged himself.

This failure in constructing the subject as an object of desire jeopardizes the construction of the fantasy. These sick children do not play or laugh.

The feeling of "existing" remains extremely fragile in children with early narcissistic disorders. The feeling of security is sometimes regained by looking at them and through rhythmic exchanges. Eye-to-eye contact is a tactile experience for infants. When a mother can, during therapy with her baby, rediscover the pleasure of contact with a baby she is rocking and singing a nursery rhyme to, a common skin is put back in place (8).

An inconsolable 3-year-old boy

A 3-year-old boy I had known as a baby, subjected to the disorganizing anxieties of his borderline mother, was subject to inconsolable terrors. During the session, he drew elephants covered with three layers of skin. I pointed out to him that they must have felt well wrapped up, unlike him. He described his anxiety about dissolving into the water like sugar on the way to the pool or spilling like an inconsistent liquid on the way to bed at night. This child had never laughed, and it took several years of therapy for him to find a sense of humor that makes distress funny.

In early therapy, play fulfills several functions, from putting the mother's relational capacities back into play to engaging in eye-to-eye communication with a baby who is just waiting for it to the slow resuscitation of a withdrawn infant, hypotonic and almost desireless. A game of imitations, tuning, sound and visual mirroring, offering empathy, sympathy, and the ability to read effects (16).

The emotional exchange is the relationship between the internal world and the outside, the material that allows the relationship to be created and to continue. The danger is not so much hatred as withdrawal, non-emotion, as Bion (17) calls "anti-emotion."

It is essential to allow a baby to act in the world and feel capable of acting on his own body and others. The game unfolds with a shared pleasure, the amused complicity of the therapist. It is important to remain naïve and enthusiastic enough to be surprised by the baby and its formidable capacities for relationship and recovery.

As Lore Schacht (18) has shown, wonder and being surprised are part of therapy with children. It is a capacity that is quickly lost in depression. Emotional sharing is the benchmark for the therapist, sometimes mired in discouraging affect, sometimes excited by the baby's rapid engagement.

Play allows the object's response to be represented: what has been refused or has been impossible, the prohibitions and the losses; play also allows the child to satisfy its need for action and transformation. The therapist must try to remain creative to keep a sense of humor, which allows the differentiation between internal and external reality to be maintained while playing for real or fictitiously. We stay in the realm of illusion to give the baby ways to represent things and to give him a chance to understand how symbolization works.

How fix the damage to the object? It is all the work of the depressive position. Moreover, in the playground that the therapist offers to the child by addressing him directly, a whole chain of representations can develop that manifest the dynamics and the imbalance linked to the depressive symptoms.

I will present my first encounter with a depressed family whose first child compulsively pulls out his hair. This is an example of a way of intervening with analytical thinking based on Freudian metapsychology, the internal economic aspect of the psychic apparatus, and the freedom to play that Winnicott gives.

A toddler suffering from a compulsion to pull out her hair

Paul walks embarrassed, badly balanced on his small, spindly legs. He is alone in his discomfort. His parents consider him an independent person who does not need support.

His father bursts out laughing loudly at seeing him so awkward and frightened when he enters my office. The whole family seems caught up in the fear of the analyst in a stranger's anxiety, leaving the tiny boy in hyper-control of his wobbly body, looking hard. His mother is more worried about my gaze than her son, who will reject her brutally when she finally extends her hand. Paul falls, gets up quickly, and rushes to the toys on my desk without exchanging a glance.

He is a little boy who does not smile and has a stern look and a bitter mouth. He has almost no hair, his skin is damaged, and his appearance is strange and disharmonious. Paul has been compulsively pulling out his hair for several months.

According to his parents, this is a reaction to frustration, but when I try to connect the dots with events, they are surprised to discover that this symptom began when the baby moved to a daycare center.

The father reports that he is furious at his son's reaction and tends to scream and sometimes grab his arm violently, as his own father did when he was holding himself badly. He can talk about this abuse with legitimacy, thus leaning on his father's model, even if this admission is a request for help on his part.

Paul's mother cannot tolerate dirt, and the falling hair drives her crazy and makes her abusive, forcing her to deny the child's suffering.

Paul walks over to my desk, stubbornly avoiding his parents and clinging to a piece of furniture. A few stuffed animals attract his attention. He is more interested in the concrete aspect of the object than in its symbolic value.

His body is solidified around a clumsy muscular carapace, a defensive second skin, as Esther Bick (19) has shown. He is not harmonious because of this muscular contracture. This little boy is not attractive.

One scenario that comes to mind is that of immature, emotionally stunted parents who ignore or abuse their infant. According to Ferenczi et al. (20), when a child is not welcomed he is subject to depression and destructiveness.

Paul touches the toys without looking at them while his parents repeat: "Say thank you to the doctor, do not touch, apologize."

Conversely, the inability to assign this little child a generational position is a manifestation of the restriction against touching, which is one of the organizers of oedipal prohibitions. A child is not touched by his parents if he is not in pain. He is repressed in his attempt at action and out of curiosity. No creativity.

When Paul is left alone with a stranger, he tries to take his anger out on his toys, but his parents, who have a very strong superego, stop him.

I think the symptom of pulling out hair is associated with a lack of sensation between mother and child, with the absence of a container and solid holding that can be internalized. This brings Didier Anzieu's Moi Peau to mind (21).

Their fear of judgment is extreme. A terrifying superego dominates the atmosphere, inhibiting everyone (22). As I point out, Paul's mother recognizes how impossible it is for her to play with her son. When they are together, she cannot stand the needs of the child who touches everything.

I say how distressed the presence of her baby makes her feel. This intervention leads to a respite from parental prohibitions, and Paul grabs a wooden duck. He places it on my desk, and the toy falls on the floor. She holds her breath, visibly distressed.

I then take the duck back and put it on the table, which I tap with the flat of my hand, saying, "Bad duck!" Paul opens his eyes, looks at me at last and waits for my reaction. I repeat the gesture of tapping the table while talking to the duck. Paul opens his mouth, drops his arms on his legs, and looks at me with a burst of surprise. The muscular relaxation is impressive. His mechanical robot look is gone. He approaches the table, throws the duck on the floor and solicits my intervention with his eyes. I repeat, "naughty duck!" We start a repetitive game, where he throws the duck more and more rigorously.

The persecutory atmosphere of the session has dissipated. Paul slams the table as he throws the duck on the floor and screams with his mouth open and head tilted back; it is more of a scream than an expression of joy, but his mother immediately interprets it as a laugh. She exclaims, "My son has never laughed!" This very brief outburst is followed by an excited lively look and a silent request to play again, which we do, with all the toys in his hand.

Paul's father is shocked that I would allow this transgression, but since the change in their son moves his wife, he agrees that I should say, "It's a game!"

Paul, however, does not smile and does not babble. But his look has become alive, and he solicits me. At the end of our first meeting, he points out a small plush ball, and I let him take this toy, which he will take home, while I ask the parents to think about bringing back the toy at the time of our second appointment, which they did, explaining their surprise as Paul replayed the fallen animal and the tapped table at every opportunity. They accepted that it was a game and were impressed by their son's ability to remember the session. This was their first positive comment about Paul. But the child pulls his hair out every night at home, and the parents' screaming does not help.

As I picked up this family in the waiting room for the second appointment, I was amazed to see this lone toddler sitting lopsidedly on the couch, holding a bottle in his mouth, with a blank stare, while both parents were sitting farther back in armchairs, busy with their own reading.

After a reunion with Paul, who took a while to exchange a glance, he got excited and repeated the game of the bad duck. I say my question is about the child's solitude with his bottle. The parents are proud to have an independent baby because, since the age of 6 months, Paul has held his bottle alone; they thought he no longer needed any contact with them.

I think of the relationship between the child's muscular contraction, aggressive tension, and maternal abandonment situations. Paul's mother understands my problem and asks for an interview with me alone. Working with the parents will allow for changes in their closeness to their baby's body and primary needs.

The discharges of tension and aggression will playfully continue in Paul. We play with balls that we exchange while I evoke the movements of proximity and distance from the maternal object.

After several weeks of guidance from the parents and playing with Paul, the symptom of hair-pulling changed. As soon as he pulls out his hair, her mother now sits next to him and gradually offers him tender contact. A doll was found in my office, whose large soft hair has become a subject of pleasure to caress and is on the way to becoming a fetish object.

But Paul needs to find self-calming actions and move from discharge to bonding. The child's therapy will last for a few years. Paul was an infant suffering from emotional neglect and a lack of auto-erotic capacity, and when he became a toddler addicted to a painful, compulsive discharge of despair and aggression. He then developed an ability to play when his mother found pleasure in interacting with him in a containing and daydreaming way (23).

Paul was a child "beyond the pleasure principle" (24), fed with bottles without the presence of the maternal object, and raised in silence when speech only served to emit prohibitions. The impulsive needs were ignored or restrained. Only the discharge remained as a solution to the tension. The experience of pleasure requires the presence of the object and the possibility of satisfaction Connivance around pleasure is sharing in the mirror that a mother may provide by acting as an accomplice and acknowledging the child's desires.

The complicity developed with the analyst allowed for exchanges of glances, emotions, then laughter. It is one of the essential roles of the third party to provide the necessary elements for satisfaction (13).

By identifying his son as being capable of joy, Paul's mother began to find pleasure in playing with him. As a mother abandoned by her own mother, she expected her son to be the one who supported and loved her.

Paul's feelings have been given substance by my interpretations of them. The ability to welcome satisfaction, enjoyment, surprise, and discoveries are balanced—or not—with the capacity for adaptation and control, which has a value of economic balance. The analyst's efforts help the kid develop the skills she needs to overcome the protective mechanisms that hold her back, such as the non-mentalized discharge processes and short circuits that deplete the richness of the child's imaginative play and her ability to represent the world around her.

Paul's first laugh in the session was a mixture of brutality and fragility. Subjected to the calming and destructive process of hair pulling, in an undifferentiated sensuality, he remained attached to the perceptive elements of the relationship. It is indeed an index of the self's premature reaction to the early trauma. By channeling their excitement into positive physiological ego sensations and the excitatory function of the analyst, who speaks and encourages the regression as access to passivity, the young child was able to feel a positive effect. Anguish is sometimes soluble in laughter.

The effect of surprise plays an essential role here. As Winnicott (25) shows in the use of the squiggle, it is when the patient surprises himself that a persistent therapeutic action can take place: the discovery of the unknown of the unconscious never before experienced. "The ability to surprise oneself presupposes the security of being carried by the framework, by the transferential relationship; it presupposes experiencing oneself as being in the attention and solicitude of the person in front of the other" [(26), p. 211].

The social laughter

I observed Paul at the nursery when he was two and a half years old: he ran straight ahead without looking at his mother, who was leaving, made a tour of the friends who had already arrived, settled in the middle, and burst out laughing while shaking his head. A wild, forced laugh was the signal for the group: all laugh and approach him. They shook their heads in rhythm and laughed out loud. Paul forced himself to laugh again to maintain the general atmosphere and the feeling of being together. His mother was gone, and the group was here. With each difficult transition, he shook his head and forced a not-so-funny but convincing throat laugh to trigger a manic defense signal. Merging with the group of restless laughers allowed him to get through the transition without being alone.

The predictable emergence of a discharge of both anxious tension and excitement, the laughter of the small child is contagious. The narcissistic contribution is immediate: "I exist. The others imitate me and surround me." The vitality of the ego-body associated with the laughing gesticulation gives the pleasure of being in the center of the group explosion.

For Paul's laughter to take on an auto-erotic value, it took many sessions of repeating the actions of throwing and tapping and then putting the toys in a container before continuing with games that put in place the symbolic value of the alternation between presence and absence (27).

Paul's compulsion to pull out his hair and to feel pain subsided and eventually disappeared when his parents started touching their child.

In denial of depressive effects, triggering the group's excitement at the nursery allowed aggressive desires to manifest. It was a noisy laugh without humor, sometimes even without comedy. The elaboration of the separation from the mother remained limited, the maternal object being little represented as a source of satisfaction.

Despite hypomanic attempts at collective laughter and the lack of resources in this small boy's symbolic activities, the transformation task of negative affect, rage, despair, and anger was not completed. Let's think of a neurosis of character in the making.

We finally quit the purely economic register of discharge to advance toward representations and a sense of humor when common mother-son laughter became possible. From motor pleasure to emotional pleasure, Paul's laughter finally led to that of his sad mother. Paul never became a clown.

I still think that even if his development went back to a more regular line. He was able to develop good language and cognitive abilities. Paul was at risk of staying in a state of narcissistic vulnerability and a tendency to use compulsive actions to manage his anxieties, as we observe in adults whose infantile trauma leaves an imprint on their capacities (28).

When he was four and was telling me a story about an abandoned child left alone in a dark forest, Paul said, "Do you remember? When I was a baby, I felt like a robot. I was like a mechanical puppet in a state of terror."

Conclusion

The essential contribution of the depression, of the loss of the object of satisfaction, to the construction of the psychic apparatus, is the birth of the capacity for representation, language, and symbolization.

The development of the internalized objects is precocious, and the quantity of investment attached to it depends on the pleasure of functioning in the Ego of the new activities offered by the environment, which plays a fundamental role. A child's joy in taking action and the amazement of their parents are both excellent indicators of progress after a period of initial collapse. What Geneviève Haag (29) described under the term "intracorporeal identification" as an essential sign to identify the positive evolution of a baby: when the child re-enacts with his body interactions with the mother, between integration, autoeroticism, and introjection. The adult must then interpret these experiences anchored in the body.

It seems to me that early joint therapies make it possible to reduce the defensive cleavage in the infant and to give back place and energy to the processes of identification by offering possibilities to act in the world and to experience the body in action. It is as much work with the parents to help them be more empathetic and less projective while tolerating what Winnicott calls the "ruthless cruelty" of their child as it is direct work with the baby through the corporal and interactive games.

Parent-baby psychotherapy means giving libidinal plasticity a chance.

The primary depressive collapse that I am talking about here leaves traces in the body, which keeps the trace of primary disorganizations, and in the psyche by modifying the possibilities of identification. I think that we can see here a determining influence on sensory integration disorders, now diagnosed as a nosography entity, cause serious developmental disorders.

A toddler's ability to laugh out loud or finally crack a smile is always a sign of positive development. Enthusiasm and the capacity for wonder, which are easily shared with the therapist, give the impression that treatment is going well. Serious or frightened infants and young children who are constantly worried or sad, when they cheer up, give the impression that the libidinal quality of existence has returned.

An indicator of development in toddlers is the ability to experience grief over loss, guilt for acting on aggressive impulses, and the ability to regulate these emotions. It is always important to observe the quality of the gaze, the harmony of the communication, the solidity of the motor tone, and the curiosity of a baby for the outside world. It is also necessary to evaluate the evolutionary possibilities of the family environment: can the parents have access to empathy, or do they remain in denial?

Recovery of libido begins with a renewed appreciation for life, but we must also lay the groundwork for the possibility of symbolizations.

It is time to develop research protocols highlighting the therapeutic impact of psychoanalysts' interventions, which helps us know the specific means of action for early disorders.

The contemporary world demands that analytical intervention quality be recognized by offering demonstrations that respect the scientific tradition.

Early therapies with therapeutic value and preventative potential for future narcissistic unpleasant difficulties can be provided to new parents and suffering newborns through the use of psychoanalytic thinking and an understanding of early relationship disorders.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

Ethical review and approval was not required for the study on human participants in accordance with the local legislation and institutional requirements. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

Author contributions

The author confirms being the sole contributor of this work and has approved it for publication.

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The author declares that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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The critical periods of cerebral plasticity: A key aspect in a dialog between psychoanalysis and neuroscience centered on the psychopathology of schizophrenia

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Through research into the molecular and cellular mechanisms that occur during critical periods, recent experimental neurobiological data have brought to light the importance of early childhood. These have demonstrated that childhood and early environmental stimuli play a part not only in our subjective construction, but also in brain development; thus, confirming Freud's intuition regarding the central role of childhood and early experiences of the environment in our psychological development and our subjective outcomes. "Critical periods" of cerebral development represent temporal windows that mark favorable, but also circumscribed, moments in developmental cerebral plasticity. They also vary between different cortical areas. There are, therefore, strictly defined temporal periods for learning language, music, etc., after which this learning becomes more difficult, or even impossible, to acquire. Now, research into these critical periods can be seen as having a significant part to play in the interdisciplinary dialog between psychoanalysis and neurosciences with regard to the role of early experiences in the etiology of some psychopathological conditions. Research into the cellular and molecular mechanisms controlling the onset and end of these critical periods, notably controlled by the maturation of parvalbumin-expressing basket cells, have brought to light the presence of anomalies in the maturation of these neurons in patients with schizophrenia. Starting from these findings we propose revisiting the psychoanalytic theories on the etiology of psychosis from an interdisciplinary perspective. Our study works from the observation, common to both psychoanalysis and neurosciences, that experience leaves a trace; be it a "psychic" or a "synaptic" trace. Thus, we develop a hypothesis for an "absence of trace" in psychosis; reexamining psychosis through the prism of the biological theory of critical periods in plasticity.

KEYWORDS

schizophrenia, psychoanalysis, cerebral plasticity, environment, psychosis

Introduction

Cerebral plasticity is our brain"s capacity to modify itself under the influence of our experiences that are influenced the environment. As early as 1909, the histologist Santiago Ramon y Cajal, who was opposed to the view of the nervous system as being static, had an intuition of this hypothesis “The nerve connections are thus not definitive and immutable, since associations are formed which are destined to subsist or be destroyed in accordance with undetermined circumstances.”(Ramón and Cajal, 1909). Since the 2000s, experimental data have brought to light molecular and cellular synaptic mechanisms of plasticity. These are mechanisms by means of which experience is inscribed in, and leaves a trace within, the neural networks. This modification of the nervous system due to the impact of interaction with the environment, is part of our organism’s process of adaptation and individuation. It is what makes the structure and functioning of our brain unique to each of us.

Synaptic plasticity of the brain remains possible throughout life, and into adulthood. It can also continue in physiological and pathological conditions, such as ageing. Until the 1990s it was deemed that adult neurogenesis (defined as the birth and development of new neurons in adulthood) did not take place after birth, but it has now been demonstrated that specific zones of the brain retain a neurogenic potential throughout life (Just et al., 2022).

However, there exists in the course of the individual’s development, precise temporal windows during which plasticity is at its peak. These temporal windows of plasticity, called *critical periods*, correspond to optimum moments when certain regions of the cerebral cortex can modify and remodel, both morphologically and physiologically, under the influence of environmental stimulations. When the critical period closes, this entails a loss of plasticity that makes the acquisition of new skills more difficult. This explains why, for example, it is easier to learn a second language or master a musical instrument during childhood rather that at adulthood.

The closing of critical periods, and the resultant partial loss of plasticity, can make the repair of damaged cortical pathways difficult. However, far from being only an inconvenience this closure is in fact essential for our development: once adulthood is reached, once the critical periods are closed, the cortex is less plastic, but it is also more stable. This make it possible to memorize and fix what has been learnt, to consolidate the traces resulting from our interactions with the environment that are the source of our autobiographical memories; something that contributes to anchoring our sense of identity. If our brain were to be too plastic, we would not be able to retain information, to consolidate the mnemic traces resulting from our past experiences. This situation could produce symptoms similar to those observed, for instance, in Alzheimer’s disease (Hensch, 2005; Testa et al., 2019).

The cellular and molecular mechanisms that are at the origin of the onset and closure of critical periods are beginning to be better understood. This promising new field of research is one that neurobiology has had a growing interest in for the last 10 years. A focus of this research is ultimately to shed new light on the role of critical periods when considering the impact of environment in some psychiatric disorders (Di Nardo et al., 2020; Vincent et al., 2021). This research, could amount to a significant point of convergence in a dialog between psychoanalysis and neuroscience in the field of mental disorders.

Freud had from the very beginnings of his work, emphasized the importance of interaction with the environment during childhood for psychological development, and the formation of personality (Freud, 1895, 1905). Similarly, he insisted on the role played by certain early experiences in the etiology of nervous disorders, observing that these might have “…more severe and lasting effects than they could do in mature years” (Freud, 1897). According to this view, the early part of an individual’s development could represent an important moment for understanding the impact of certain interaction with the environment on the individual’s mental and emotional development; including understanding the etiology of some mental illnesses. Freud also underlined the role of certain innate predispositions in the etiology of these pathologies (Freud, 1896). To understand the twofold influence of these innate factors, alongside factors acquired during childhood that can act in a complementary way, he put forward the idea of a series of associated factors that complement each other. According to this view of a multifactorial etiology of mental illnesses, pathologies might result either from: the combination of a strong innate disposition and some lesser acquired factors; of the presence of a minor innate disposition mixed with a strong incidence of events linked to interactions with the environment, during the critical period that stretches from the first months of life up to puberty; or, in some rare cases, a pathology could be caused by the influence of only one of these determining factors (Freud, 1917). Freud maintained this concept of a twofold etiology, looking beyond the alternative which would involve making a choice between exogenous and endogenous factors when determining the etiology of mental illnesses.

If we focus particularly on the etiology of schizophrenia, recent data from research in psychiatry seem to support, in a remarkable way, this hypothesis for a possible interaction between genetic and environmental factors. Initially studies into the origin of this pathology concentrated on searching for a genetic mutation (Riley et al., 2005). However, the hypothesis that schizophrenia is a single gene disorder has been abandoned for a number of years now (Brown, 2011; Ripke, 2014). This is because the development of genome-wide association studies (GWAS) has shown that vulnerability to schizophrenia is dependent on a large number of alleles with very small effects, and so can only partially explain the appearance of the schizophrenia phenotype (Sanders et al., 2008; Psychiatric GWAS Consortium Bipolar Disorder Working Group, 2011; Ripke, 2014; Misiak et al., 2018). Furthermore, these genetic factors are not specific to schizophrenia but can be found with other psychiatric pathologies (Van Snellenberg and de Candia, 2009; Hill et al., 2013; Skudlarski et al., 2013; Cardno and Owen, 2014; Wang et al., 2015; Misiak et al., 2018).

Studies of monozygotic twins have brought to light that, when one twin presents a phenotype for schizophrenia, there is a measure of agreement that goes from 40 to 60% of cases where the second twin presents the same phenotype (against a 10 to 15% for dizygotic twins; Fischer, 1971; Kringlen and Cramer, 1989; Cardno et al., 1999). This genetic matching is therefore relative, and the existence of a very high number of twins who share the same genetic heritage but where only one twin develops a phenotype for schizophrenia could be explained by environmental factors. Environmental factors can act *in utero*, and the fetal experiences of the monozygotic twins may prove to be different (Piontelli et al., 1999). They can also act during the perinatal period, during childhood, or even at adolescence (Brown, 2011).

Studies into the role played by the environment in the etiology of schizophrenia developed later than the initial research into the genetic etiology. This was due, among other things, to the methodological problems linked to these studies, as well as the difficulty in scientifically measuring the impact of environmental factors on the development of a phenotype for schizophrenia. Today however, we know that besides the genetic risks a number of environmental exposures can interfere with brain development, and originate some of the brain anomalies observed with schizophrenia. Among these factors, we find events that occur at different stages of development – and thus at critical periods involving distinct areas of the brain. In particular, we find: prenatal infections, obstetric complications, nutritional deficiency and *in utero* maternal stress, postnatal infections, trauma during childhood, consumption of toxic substances such as cannabis during adolescence, but also socio-economic factors such as growing up in an urban environment (Andreasson et al., 1987; Ames, 2001; Arseneault et al., 2002; Aleman et al., 2003; Allardyce et al., 2005; Dean and Murray, 2005; Amminger et al., 2007; Arion et al., 2007; Amminger et al., 2010; Brown, 2011; Marangoni et al., 2016).

Yet, when they are studied in isolation, these environmental factors, just as the genetic factors, present very low levels of effect and explain only partially the development of a vulnerability to schizophrenia. A 2017 study proposed a method (the polyenviromic risk score) for calculating the combined score of various environmental risk factors that seemed to corelate the most with a vulnerability for schizophrenia (Padmanabhan et al., 2017). However, insofar as genetic and environmental risk factors identified in schizophrenia remain of little significance taken in isolation, for a number of years now scientists have increasingly been focusing on the possible interactions between these two kinds of factors (Misiak et al., 2018) – in effect revisiting the Freudian hypothesis of a series of factors that complement each other, involving both types of determinants. Studies focusing on the interaction between these two factors aim to bring to light cases where the effects of certain environmental exposures are predominant in subjects who present certain underlying genotypes; or, alternatively, where the expression of a particular genotype would be dependent on the presence of given environmental factors (Ottman, 1996; van Os et al., 2008; Ayhan et al., 2016). The majority of studies focusing on gene/environment interactions have concentrated on the effect in variation of COMT, BDNF and FKBP5, which may interact with certain environmental exposures such as the presence of trauma or abuse in childhood, or the use of cannabis during adolescence (Brown, 2011; Misiak et al., 2018).

The research protocols for the link between genetic vulnerability and environmental exposure study three kinds of interaction (Clarke et al., 2009). One area of research is the increased effects of environmental exposure, owing to the presence of a certain susceptible genes (Brown, 2011). For example, the risk of schizophrenia developing in offspring of mothers who were exposed to pyelonephritis during pregnancy, is higher when antecedents for psychosis exist in the family and two genes (*DISC1* and *MIA*) may be involved. Another focus is the possibility that genetic vulnerabilities might constitute a risk factor for certain detrimental environmental exposures. This would be similar to the genetic predisposition to compulsive behaviors that can make some individual more susceptible to environmental incidents such as head trauma (Kendler and Eaves, 1986). Finally, there is research into the epigenetic factors involved in the etiology of schizophrenia. These studies look at functional changes that do not involve alterations in nucleotide sequence. They indicate that some environmental exposures that occur during critical periods in postnatal development can alter the epigenome, leading to changes in gene expression throughout the individual's life (Zhang and Meaney, 2010). These alterations can be at the origin of an increased vulnerability for schizophrenia and, in some cases, are susceptible to transmission across generations (Brown, 2011).

Thus, some surprising similarities between recent data on the etiology of schizophrenia and some of Freud's intuitions, have come to light. These similarities concern both the interaction between different factors for vulnerability, as well as the importance of critical periods in development during which the effects of certain environmental exposures are increased. If the idea of critical periods can represent a starting point for a promising dialog between psychoanalysis and the neurosciences, as regards research into psychiatric pathologies, some of the recent data in particular, encourages us to initiate that dialog. Indeed, as research in neurobiology has begun to better understand the molecular and cellular mechanisms that determine the onset and closure of critical periods, some studies have evidenced the existence of anomalies in the physiological mechanisms linked to critical periods of brain plasticity in schizophrenia patients.

The aim of our work is therefore, to initiate that dialog between psychoanalysis and the neurosciences around the role of critical periods in an understanding of schizophrenia, while at the same time, indicating the possible therapeutic horizons this interdisciplinary approach presents. To that end, we propose first to define the biological concept of *critical periods*, using the model of a developmental pathology linked to the acquisition of binocular vision, namely amblyopia. This pathology has allowed scientists to better understand the molecular and cellular mechanisms that determine the onset and closure of critical periods. We propose describing some of the mechanisms linked to the maturation of inhibitory parvalbumin-expressing interneurons, and we will discuss the anomalies related to the functioning of these inhibitory cells that have been observed in schizophrenia patients.

We will then revisit the research around the cellular mechanisms of critical periods and their anomalies observed in schizophrenia, through the prism of a dialog between psychoanalysis and neuroscience. This dialog is based on the intersection of these two disciplines around the concept of “trace” (Ansermet et al., 2007). This model draws on the fact that psychoanalysis and neuroscience come together around the shared observation that experience is inscribed leaving an organic trace. On the basis of this model, we will endeavor to reframe the Freudian and Lacanian psychoanalytic theories on the etiology of schizophrenia – thus putting them into perspective with the research done on critical periods and the amblyopia model. We propose putting forward an interdisciplinary view of schizophrenia as resulting from an “absence of trace,” that is to say, a defect in inscription of certain experiences linked to interactions with the environment during critical periods of brain plasticity. We will then propose some hypotheses relating to the nature of these traces – traces that may be at the root of our sense of self, and of identity –, as well as the effects resulting from the absence of their inscription (or pathological inscription, as is the case in amblyopia) for schizophrenia. Finally, we will consider, from an interdisciplinary perspective, the therapeutic possibilities offered by the work being done around the reopening of critical periods for plasticity.

Critical periods as temporal limits to plasticity: The amblyopia model

The research done by David Hubel and Torsten Wiesel on the visual system of cats – for which they received the 1981 Nobel Prize in Physiology or Medicine – was the first to experimentally demonstrate the temporally circumscribed character of cerebral plasticity. Their questions stemmed initially from a disease named amblyopia. In humans, amblyopia is a pathology characterized by a unilateral visual impairment in children who had been operated too late for cataracts, or who presented a significant strabismus that was not treated before the ages of four or five. The absence of stimulation in one eye during a certain length of time, led to a definitive weakening of cortical representation in the visual cortex for that eye in favor of the visual field of the other eye (Prochiantz, 2012). Although the term commonly used for this disorder is “lazy eye,” it is however not a lesion or a functional disorder of the eye that is the root cause. The sensory organ itself is completely functional, it is at the cortical level that the representation is absent (the representation of both eyes is modified). Wiesel describes having initially tried to build an animal model that could demonstrate this particularly of the visual system"s development, based on experimentation using kittens. The results brought to light that, depending on the timing and the duration of the suture of one eye, the development of the visual system in the visual cortex showed noticeable differences. If the sutures were performed immediately after birth and left for 3 months, the kitten were definitively blind in the eye that had been closed (Wiesel, 1982). Even once the eye was allowed to open, the majority of neurons situated in the visual cortex only respond to stimulations from the eye that had remained open. The work of Hubel and Wiesel, thus made it possible to demonstrate experimentally that the development of binocular vision requires the stimulation of both eyes, during a very specific timeframe in postnatal development. Their research showed that the development of the visual cortex is dependent on the environment. The absence of stimulation in either of the two eyes, can have irreversible consequences for the acquisition of binocular vision.

If a kitten’s eye is closed 2 weeks after birth, at the end of 18 months only the eye that was left open will give rise to cortical activity. However, if the eye is closed 10 weeks after birth, the impact is lessened, and significant activity can be observed in the cortical region when stimuli is applied to that eye. Closing of the eye that occurs after 1 year still shows some impact, but far less than the two previous interventions; and if the closing takes place at 6 years of age, no modification or reorganization of the cortical circuits are observed (Prochiantz, 2014; Di Nardo et al., 2018; Di Nardo et al., 2020). There exists therefore a precise temporal limit for the establishment of the phenomena of brain plasticity, insofar as that it is significantly reduced after a certain “critical” period for the development of any given brain function (in this instance binocular vision). After the closure of the critical period within the primary visual area, treatment of amblyopia becomes more difficult, even impossible (Scheiman et al., 2005). Sensory experience therefore, plays a significant role in the building of the central nervous system. The work of Hubel and Wiesel contributed to showing that, although brain development is in part determined by innate genetic factors, that development can be very labile within any given genetic makeup. The environment, especially through certain early experiences, modifies in a durable or even irreversible way the structure of the nervous system. These modifications can, however, only happen if the stimuli occur within a precise temporal window. Brain plasticity is therefore not limitless throughout development, and interactions with the environment will not have the same effects on the organism depending on when they take place (Hensch, 2005). Consequently, there exist strict temporal windows that limit the impact of experiences on the organism and on the nervous system.

If the work done on critical periods within the nervous system has played an important role in the experimental demonstration of this phenomenon, the existence of such temporal windows for the development of the central nervous system affects many other areas of the brain, and the critical periods for these different areas of the cortex are not synchronous. Depending on the regions of the brain and the circuits involved, the critical periods can occur at very different stages of overall development. In the case of some functions, plasticity persists up to quite a late age such as adolescence. While other faculties have a plasticity that is limited to the first years of infancy.

There are critical periods for various processes of increasing complexity, and they generally happen in a cascade through different neuronal circuits (Di Nardo et al., 2020). Initially they occur within the primary sensory areas such as sight or hearing; then they spread to reach the parts of the brain that handle motricity and language. The physiologist Jeannet Werker evidenced a hierarchy of these critical periods within the different cortical areas that are involved in language, and the acquisition of the ability to recognize the maternal language and its pronunciation. Each area has its own window of plasticity (Werker and Hensch, 2015). Then, much later, a critical period takes place in the areas linked to the control of emotions and cognitive functions; these remain able to modify up to the end of adolescence (Takesian and Hensch, 2013; Werker and Hensch, 2015; Nelson III and Gabard-Durnam, 2020). Additionally, depending to the areas that underpin these different skill acquisitions, the duration of the critical periods appears to be vary widely: from a few months for the ability to discriminate phonemes for example (and other sensorial areas), up to several years for higher cognitive functions. As an example, the prefrontal cortex has a neuronal circuit that has a particularly lengthy period of plasticity that continues up until puberty and also in young adults.

The cellular mechanisms behind critical periods: The role of parvalbumin-expressing inhibitory neurons

Despite the diversity of cerebral areas that are subject to critical periods, studies on mice have brought to light that the physiological mechanisms that control the onset and closure of these periods are the same (Di Nardo et al., 2020). Furthermore, these mechanisms are closely linked to the maturation of inhibitory neuronal circuits. The balance between excitation and inhibition within the cerebral cortex is key to understanding the physiological mechanisms that regulate critical periods (Hensch, 2005; Bernard and Prochiantz, 2016). Inside the nervous system, it is the levels of inhibitory activity of particular GABAergic neurons that plays a determining role in the balance between excitation/inhibition. Their inhibitory neurotransmitter is Gamma-aminobutyric acid (GABA), which when liberated inhibits the firing of the postsynaptic excitatory neurons. The onset of critical periods occurs at the start of the maturation of a particular group of inhibitory GABAergic neurons called *basket cells*, which synthesis a specific protein called parvalbumin (PV). Experiments performed on the visual cortex of mice show that the critical period closes when these neurons reach complete maturity (Hensch, 2005; Werker and Hensch, 2015). Thus, the onset of a critical period is triggered by the maturation of the PV expressing inhibitory interneurons, and when maturation reaches its peak the period closes, resulting in a considerable decrease in the plasticity of the circuit. Consequently, when inhibition reaches a certain level the circuit becomes closed, losing its plasticity. It can be observed that, at the moment of their maturation, the basket neurons physically become “fixed” by surrounding themselves with perineuronal nets (PNNs). These PNNs are an extracellular matrix structure, in the form of a proteinaceous net enriched with complex sugars from the glycosaminoglycan family (GAG). These structures contribute to stabilizing the system and considerably reducing its plasticity (Hensch, 2005; Bernard and Prochiantz, 2016; Testa et al., 2019; Di Nardo et al., 2020). Additionally, the balance between excitation and inhibition that is reached at the end of the critical period, through the maturation of the PV inhibitory neurons, is necessary to maintain the non-plastic state of the adult brain. That inhibition reduces the plasticity in the adult brain, is demonstrated by the increase in plasticity that follows the reduction in GABAergic transmission in adult rodents, caused either by a GABA antagonist or by the inhibition of GABA synthesis (Harauzov et al., 2010).

Scientists have noted that, from the point of view of energy, the metabolism of PV producing cells involves a significant and sustained physiological activity (Alitto and Dan, 2010). Indeed the GABA released by the PV cells binds to the alpha-1 type GABAergic receptors present on the cell body of the pyramidal neurons. Owing to the rapid desensitization of the receptors and the excitatory feedback loop coming from the postsynaptic cells, a high-frequency (between 40 Hz and 80 Hz) oscillation occurs during the critical period when inhibition is at its peak. The release and capture of ions as the depolarization and repolarization of the membrane of the PV cells takes place requires the production of high levels of ATP. The metabolic rate of the PV cells is thus very high, and a deficient metabolism can therefore have serious physiological consequences on the functioning of these inhibitory circuits (Prochiantz, 2014).

A study on primates undertaken in 1996 showed that the maturation of PV cells took place at an earlier stage in the sensory areas A1 and V1, and progressively later, depending on its progress, through the hierarchy of the cortical areas (Condé et al., 1996). In humans, the critical periods for discriminating phenomena or for the acquisition of binocular sight also end early on, since the sensorial neuronal networks become fixed relatively early on in the course of development. However, it should be borne in mind that it is the photons hitting the retina or the waves hitting the internal ear cilia that allow for the initial maturation of PV cells. There is no clock for this maturation, and sensorial deprivation, as keeping both eyes closed in animals, delays PV cell maturation: plasticity does not open until the eyes are open. This also explains why the critical periods are not synchronous (Werker and Hensch, 2015).

In contrast to the sensory areas, the neuronal networks of the prefrontal cortex, associated with higher cognitive functions and mood regulation, have a particularly long period of plasticity: the maturation of PV cells is observed in these areas only at the onset of puberty, with a critical period that ends around the age of 20 years. The exceptionally slow nature of the maturation of the PV cells within these cortical areas is especially noticeable in humans (Prochiantz, 2019).

The role of OTX2 in critical periods and their environmental modulation

Among the factors identified that regulate the maturation of the PV neurons, the homeoprotein OTX2 plays a key role. It is a transcription factor, that is, a protein which is necessary for regulating the transcription of certain genes, while also interacting with the DNA and RNA. The homeoprotein OTX2 is transferred from an extra-cortical source (the choroid plexus) to the cortex (Spatazza et al., 2013; Di Nardo et al., 2020). An initial activity-dependent condensation of the extracellular matrix of PV cells into perineuronal nets (PNNs) allows OTX2 internalization, initiates the opening of critical periods at a first concentration threshold and closes it at a second one. OTX2, once internalized, maintains PNN assembly, initiating a positive loop of interaction between PNNs and OTX2 (Sugiyama et al., 2008). This mechanism first demonstrated in the mouse visual cortex (V1) was generalized to the primary auditory cortex (A1), and medial prefrontal cortex (mPFC). Lee et al., 2017). Since, choroid plexus-derived OTX2 accumulates in PV cells throughout the cerebral cortex, it can be proposed that it is a general critical period regulator.

As mentioned above, the regulation of the onset and closure of critical periods through the accumulation of OTX2 takes place within a process that involves a two-thresholds model (Di Nardo et al., 2020). At the first OTX2 accumulation threshold, the window of plasticity opens. Then, the crossing of a second threshold of accumulation triggers a closure of the plasticity and maintains the non-plastic state of the adult brain. Prochiantz and Di Nardo, 2015). In the experimental protocols put in place using mice, a cortical infusion of the OTX2 protein accelerates the appearance and the closure of the critical period. While if OTX2 internalization by PV cells is blocked in a region of the cortex, the onset of the critical period is delayed in that region. Most importantly, permanent OTX2 import is necessary to maintain the adult non-plastic state and, as shown in V1, decreasing OTX2 in adult PV cells temporarily reopens plasticity.

Thus, if the influence of OTX2 on the maturation of PV cells has a direct incidence on the onset and closure of the critical periods, genetic or environmental factors that act upon the expression of OTX2 or its assimilation by the PV cells can result in a poor synchronization of the critical periods for plasticity within the cortex, possibly resulting in subsequent disorders. Genetic modelling in mice has shown that a genetic point mutation in the recognition motif of the GAG PNN by OTX2 delays the maturation of the PV interneurons not only in the primary visual area, but also in the primary auditory area, and the mPFC. This delayed plasticity can be correlated with a reduction of anxious-like behavior in mice (Lee et al., 2017). And more recently Vincent et al. have demonstrated that anxiety-like behaviors in the adult can me modulated by changing OTX2 levels in the choroid plexus or blocking its transfer into mPFC PV cells (Vincent et al., 2021). OTX2 signaling, and the synchronization of critical periods, can also orchestrate complex behaviors, reflecting the interaction of several sequential critical periods, as with language (Werker and Hensch, 2015). In consequence, a disrupted OTX2 signaling may be the cause of the onset of certain psychiatric and cognitive disorders (Gogolla et al., 2009; Le Magueresse and Monyer, 2013; Maeda, 2015).

Besides these genetic factors, environmental factors can also act upon the OTX2 signals, and influence the onset and closure of critical periods. In dark-reared mice, the retina is not stimulated and the activity signal is not sent to the cortex (Sugiyama et al., 2008). Thus, PV cells are not informed that the eyes are opened (because of the dark) and do not start to assemble the PNNs. As a consequence, OTX2 is not captured by the PV cells. In this mice, a 70% decrease in the quantity of OTX2 present in the PV cells was observed (Sugiyama et al., 2008). This absence of OTX2 capture does allow a further reinforcement of PNN assembly. The initial PNN assembly is activity dependent - if not all critical periods would be synchronous. It appears then, that the onset of critical periods requires adequate environmental stimuli to regulate the OTX2 signals (Di Nardo et al., 2020). Another study on mice has demonstrated that depriving the newborn of the maternal presence during a critical periods stretching from the 10th to the 20th day postpartum can result in a higher percentage of the offspring being subject to permanent anxiety and/or depression – even if some individual appear to present a degree of resilience (Peña et al., 2017). In the non-resilient progeny, the expression of OTX2 is transiently weaker in the ventral tegmental area (VTA) during the period of maternal separation. We should recall that the VTA is a dopaminergic hub in the mesencephalon, whose neurons project into the cortex and the sub-cortical structures, with a greater number of projections in the limbic area. The role of OTX2 in complex traits such as depression or anxiety has been evidenced not only in murine models of early stress, but also in humans, where studies have been done with children victims of maltreatment (Peña et al., 2017; Murthy et al., 2019; Vincent et al., 2021). Samples of DNA from children who had suffered maltreatment showed a correlation between the presence of depression and the state of OTX2 gene methylation, and of genes downregulated by OTX2 (Kaufman et al., 2018). This would seem to strengthen the hypothesis that OTX2 is an important modulator of mental health (Di Nardo et al., 2020).

Cellular anomalies linked to critical periods in schizophrenia

Anomalies in the maturation of PV cells correlate with a number of psychiatric disorders (Gogolla et al., 2009; Lee et al., 2017); (Maeda, 2015). To explain these anomalies, an OTX2 hypothesis can be envisaged but is not the only one, and other models are not incompatibles with a regulation by OTX2, like the oxidative stress model which also explains PV cell maturation and excitation/inhibition balance modifications in schizophrenia. For example, the dysfunction of PV cells can be explained by the high metabolic requirements of these cells, which may render them susceptible to redox dysregulation and oxidative stress. A protocols using mice carrying a genetic redox imbalance has demonstrate that PNN play a critical role in the protection against oxidative stress. Although the perineuronal nets act as a protective shield, they are also themselves sensitive to excess oxidative stress. The protection might therefore reflect a balance between the oxidative burden on perineuronal net degradation and the capacity of the system to maintain the nets. Abnormal perineuronal nets, as observed in the postmortem patient brain, may thus underlie the vulnerability and functional impairment of pivotal inhibitory circuits in schizophrenia (Berretta et al., 2015).

More generally, it has been observed that patients with schizophrenia present a lower density of PNNs than control groups, within the amygdala, the entorhinal cortex, and the dorsolateral prefrontal cortex (Pantazopoulos et al., 2010; Mauney et al., 2013; Berretta et al., 2015). This translates into an anomalous maturation of the PV cells in this region. The prefrontal cortex is one of the areas of the cortex whose disfunctions have been particularly associated with schizophrenia (Callicott et al., 2000). In this region of the brain, in healthy subjects, the maturation of the PV neurons at puberty usually triggers an increase in the quantity of PNNs, which translates into an increase in inhibition. In schizophrenia patients, although the density of PV neurons is not abnormal, it is a fault in their maturation that creates a disruption in inhibition within the prefrontal cortex (Enwright III et al., 2018). Hence the reduction in high-frequency gamma oscillations, and a lower PNN density observed in some patients (Uhlhaas and Singer, 2015; Di Nardo et al., 2020). Lacunae in the number of inhibitory synapses can also be observed in schizophrenia patients, along with a reduction in the pruning of excitatory pyramidal neuron dendrites (Insel, 2010).

It is therefore an increase in excitation, to the detriment of inhibition (a modification in E/I balance), that takes place within the neuronal circuits. Following this idea, scientists have noted an immaturity of the PV cells within the prefrontal cortex of schizophrenia patients (Mauney et al., 2013). These results have been confirmed by Daniel W. Chung's team, who have also brought to light a significant reduction in the expression of PV within the dorsolateral prefrontal cortex of schizophrenia patients (Chung et al., 2018). There could therefore be a link between this kind of psychiatric pathology and an anomaly in the timing of the periods of plasticity. Such periods of plasticity would extend over an abnormal length of time, owing to a lack of inhibition, subsequent to a late maturation of the inhibitory circuits (Mauney et al., 2013). Still, it is difficult to determine if these alterations in the balance between excitation-inhibition and the gamma oscillations in schizophrenia, linked with the faulty maturation of the PV neurons that do not sufficiently surround themselves with PNNs, could be a cause or a consequence of the pathology (Testa et al., 2019).

To bring some answer to this question, it is appropriate to note that certain genetic etiological factors and environmental factor associated with schizophrenia, can have consequences for the maturation of the PV cells and their PNNs. For example, several studies highlight a link between schizophrenia and anomalies in the expression of certain genes that are necessary for the formation of the PNNs (such as NEUROCAN, HAPLN4, or PTPR; Schizophrenia Working Group of the Psychiatric Genomics Consortium, 2014; Testa et al., 2019). Similarly, murine models that use environmental exposure to induce a phenotype that mimics aspects of mood disorders in mice, show anomalies in the maturation of the PV cells. For example, a study in mice using maternal immune activation during pregnancy to induce anxiety-related phenotypes among the offspring, has shown that these mice present a reduction of PNNs in the mPFC and the amygdala (Paylor et al., 2016). Similarly, as we have already observed, in mice the exposure to early stress in the first day of life induces changes in the expression of OTX2 in the VTA or the choroid plexus, and it is known that the expression of OTX2 in the choroid plexus regulates the maturation of PV neurons (Peña et al., 2017; Di Nardo et al., 2020; Vincent et al., 2021).

The alterations in the PNNs observed in schizophrenia patients could therefore appear as a consequence, linked with the genetic and/or environmental factors associated with schizophrenia. However, it should also be pointed out that the removal of the PNNs in the hippocampus of mice can also directly bring about behaviors similar to the positive symptoms of schizophrenia (Shah et al., 2013). Likewise, experimentally inducing a lowering of the transfer of OTX2 from the choroid plexus to the PV neurons in rodents, results in anxiety-related phenotypes (Vincent et al., 2021). The lack of inhibition linked to the abnormalities in the maturation of the PV neurons that regulate critical periods could therefore, in the case of schizophrenia, be associated with an excess of plasticity. Such an excess of plasticity, is at the root of certain symptoms observed in this pathology.

But if we concentrate on a defective maturation of PV cells on the basis of markers, maturation could be so low that the first threshold (plasticity opening) is not even passed. Then the schizophrenia phenotype would be a consequence of an absence of plasticity when needed to repose to an environmental change.

Yet, if this faulty maturation of the PV interneurons has been recognized as associated with the disorders in cerebral development present with schizophrenia, this observation is compatible with an interaction of genetics and environmental factors in schizophrenia etiological models (Prochiantz, 2019).

A dialog between psychoanalysis and neuroscience around the anomalies in critical periods observable with schizophrenia: The hypothesis of the absence of trace

Even if much remains to be done, and caution should be exercised, recent discoveries about the cellular and molecular mechanisms determining the onset and closure of the critical periods of plasticity have opened a new field of research in the understanding of certain psychiatric disorders such as schizophrenia. If the anomalies in the maturation of PV cells within the prefrontal cortex observed in this pathology can be both genetic and environmental in origin, their primary consequence is a disorder in the timeframe of critical periods for plasticity. This disorder can impact on the manner in which the environment models and structures the development of the cerebral cortex. Indeed, if the onset of a critical period does not occur at the right time, or if stimulations from the environment do not take place in a suitable way at the time when the critical period is open (as was exemplified with the case of amblyopia), the connections of the neuronal networks can fail to be put in place, or be inadequately put in place, within the cortex. In other words, for the stimuli linked to our interaction with the environment to be inscribed within our nervous system and guarantee a good development of our neuronal connections, these stimulations need to take place at the moment when the plasticity is at its peak, during the critical period for the region of brain concerned, and then be stabilized and consolidated through the closure of the window of plasticity. When it has not been possible for the environmental experiences to be inscribed in the form of neuronal traces during the critical period of plasticity for a given region of the cortex, one of the consequences is that we observe anomalies in the structure of the cerebral cortex in adulthood. This can be seen in the data from brain imaging of patients with amblyopia. Now, if anomalies linked to the maturation of the PV cells in schizophrenia cause either a delay in the onset of a critical period, or a failure in its closure, the consequence may be that certain traces resulting from interaction with the environment might not be inscribed, or would not be adequately consolidated in the cortical regions involved, including the dorsolateral prefrontal cortex. This hypothesis coincides with the data coming from brain imagery of patients with schizophrenia, where numerous anomalies in the structure of the cortex can be observed (Arnone et al., 2009; Gur and Gur, 2010).

Structural imaging data have shown that schizophrenic patients tend to have a thinner cortex in parts of the brain such as the insula, have enlarged lateral ventricles, and have smaller hippocampal volumes (Haukvik et al., 2013). With regard to functional imaging data, many brain regions and networks show abnormal connectivity in schizophrenia. The principal results observed concern a hyperactivity of the default mode network (Hu et al., 2017) and functional connectivity anomalies in the insula (Wylie and Tregellas, 2010), hippocampus and prefrontal cortex (Lalousis et al., 2022). Aberrations observed on functional MRI scans in these regions are particularly interesting to relate to the PV cells' abnormalities. The significant loss of PV cells in the hippocampus can lead to hyperactivity of this region that then induces a hyperdopaminergic disorder. This is associated with psychotic symptomatology. Data on the hyperactivity of the dopaminergic system in schizophrenia could thus appear to be strongly correlated with the loss of PV cells in the ventral hippocampus (Grace and Gomes, 2019). A recent study has proposed an animal model of stress during critical periods, to explain abnormalities of the hippocampus and prefrontal cortex in schizophrenia. Exposure to a prolonged level of stress impacts the ventral hippocampus, inducing an abnormality in PV cell function and cell loss. This contributes to the hyperactivity of glutamatergic pyramidal neurons. Therefore, a hyperdopaminergic state appears – which is strongly associated by many studies with psychotic symptomatology. Furthermore, the medial prefrontal cortex, and particularly the prelimbic portion, has been shown to be involved in the control of stress response in basolateral amygdala. The implication is that the prefrontal cortex would no longer be able to regulate the amygdala's reactivity to stress. This would then lead to glutamatergic hyperactivity, which in turn could induce damage to parvalbumin cells and then generate a hyperdopaminergic state (Gomes et al., 2019).

If the latest advances in biology research on critical periods show promise for the understanding of the role of early interactions with the environment in the etiology of schizophrenia symptoms, these discoveries also open the way for an innovative dialog with psychoanalysis. Freud also had very early on emphasized the important place held by our childhood experiences, and early stimuli linked to the environment, in our subjective construction, as well as in the appearance of certain psychopathological disorders (Freud and Breuer, 1895; Freud, 1905). The work of several post-Freudian psychoanalysts, such as Jacques Lacan, also sought to apply this perspective to the understanding of the psychopathology of psychosis (Lacan, 1997). To draw a comparison between these psychoanalytic hypotheses and research in neurobiology on the critical periods, we are now going to propose integrating this research into a previously formulated model of interdisciplinary dialog (Ansermet et al., 2007; Ansermet and Magistretti, 2010; Tran The, 2022).

This model of dialog between psychoanalysis and neuroscience is rooted in the fact that both these disciplines, despite their radically different epistemological foundations, nevertheless meet around the idea of "trace." Indeed, if the neurosciences have demonstrated that experiences stemming from our interactions with the environment are inscribed in the form of traces (such as synaptic traces) within the nervous system – traces that integrate structural and functional modifications in the organization of neuronal networks –, the concept of "trace" is also at the root of the whole Freudian theory of the functioning of the psychic apparatus. In this respect, psychoanalysis and neuroscience come together over the shared observation that experiences linked to the environment are inscribed and leave a trace, whether we think of this trace as "physical" in the neurosciences, or as a "psychic" trace in the psychoanalytic perspective (Freud, 1887-1904).

This model is also based on the fact that, if experiences are inscribed and leave a trace (both psychic and synaptic), these traces are associated with somatic states. Representations (*R*) resulting from exteroceptive perceptions are linked, owing to an association through simultaneity, with representations resulting from interoceptive perceptions of somatic states (*S*) that have been the object of a concomitant and synchronic perception. Experimental discoveries around synaptic plasticity and the neurobiological mechanisms of memory, as well as the Freudian theory of the formation of the first mnesic traces in the experience of satisfaction, put the emphasis not only on the perceptions stemming from the external environment, but also on the paramount importance of the interoceptive perception of the organism's state (be it a corporal perception of pleasure or displeasure) that accompanies the external perception (Freud, 1900). The action of the perception of the internal states of the body are therefore critical to the inscription of the experience; and the simultaneity of the perceptions *R* and *S* appear to be an essential condition for the inscription of the trace in the form of an association *R-S*.

Furthermore, the Freudian definition of thought as being in the service of the pleasure principle, Freud (1900) as well as the biological definitions of thought proposed by neuroscientist such as Alain Prochiantz – who sees thought as "an adaptive relationship that binds the individual and the species to their environment" [our translation](Prochiantz, 2001) –, or Antonio Damasio, according to whom the human faculty of reasoning appeared in evolution primarily to serve homeostasis, Damasio (2017) all these theories imply seeing cognitive function withing a homeostatic framework where the representations produced by our thoughts fulfil the purpose of regulating our somatic states. On the basis of this coming together of psychoanalysis and neuroscience around a homeostatic understanding of the psychic functioning (and in particular thought processes), we can underline the importance of this homeostatic functioning of the trace. This function appears to be a point of intersection between these two field, whose epistemological foundations are yet incommensurable (Ansermet et al., 2007).

In his model of the newborn's experience of satisfaction, Freud described how the first experiences linked to interactions with the environment – most notable during the fulfilment of the newborn's basic needs by the adults who care for it – were inscribed within the psychic apparatus (Freud, 1900). The exteroceptive perceptions – that accompanied the pleasure that results from the satisfaction of basic needs such as hunger – are inscribed in the infant's memory, and these mnesic traces that are the origin of the first representations, play the role of regulators in the infant's first somatic experiences. These first experiences are marked by a state of complete helplessness, which Freud termed *Hilflosigkeit*. This is a state in which "unpleasure" dominates the first corporal experiences of the newborn. According to the psychoanalytic perspective, in the child's subsequent development, different traces (notably the Oedipus complex) that stem from interactions with the environment, as well as those produced by the subject themselves, perform this homeostatic function.

It is possible to integrate the Freudian model of the experience of satisfaction, and neuroscientific research on the role of the perception of somatic states in the inscription of experiences (Tran The, 2022). For example this has been done in research on interoception,(Craig, 2009) or on somatic markers (Damasio, 1996). In this interdisciplinary perspective, the representations (R), which come from the environment, from language and from those who take care of the newborn, play a regulatory role on the body states and on the interoceptive perceptions (S). This perspective also accords with Wilfred Bion's description of the alpha function of the psychical apparatus. The child is invaded by "beta" content, such as somatic perceptions that are sources of anxiety, in a raw relationship to the reality of its body and the world. The alpha function corresponds to the subject's capacity, through the mediation enabled by the adult, to metabolize beta elements. According to Bion, the prototype of the alpha function is the parent's capacity for dreaming. The parent takes into himself the contents deposited by the infant and restores elaborated alpha elements to the infant. In turn, these elements then allow the infant to distance himself from experiences, and from the intense and painful sensations resulting from its state of *Hilflosigkeit*.

Another prolongation of the Freudian understanding of trace inscription resulting from the newborn's experience of satisfaction, is the perspective put forward by Lacan. Based on Saussurean linguistics and Lévi-Strauss' structural anthropology, the psychoanalyst Lacan proposed rereading the Freudian concept of "*trace*" in term of "*signifier*." Lacan's argument was that "The memory phenomena that Freud is interested in are always language phenomena" (Lacan, 1997), and language is the result of the incidence of intersubjective relationships with others – including the adults caring for the child during its development. Thus, Lacan draws the field of the signifier into that of the "Other," which is to say alterity or otherness in its language dimension. From there he will postulate the existence of a field of the "primordial signifier," of traces whose function is homeostatic and structuring for psychic development (Lacan, 1997). Based on this idea, the Lacanian psychoanalytic perspective understands psychosis as the result of an absence of inscription in childhood of some of those traces essential to our subjective construction.

Thus, by rereading Feud's theory on the organizational role of the Oedipus complex in psychic development, Lacan proposed the hypothesis of an etiology of psychosis based on the signifier (Lacan, 2002a). Lacan develops his theory of "*foreclosure*," that is to say the rejection of the primordial signifier, as a precondition for psychosis (Lacan, 1997). In line with this position, at the origin of psychosis one would find the absence of inscription of what is, for the psychic apparatus, a primordial and structuring representation – be it the unifying of the image of the body or the organizing role of the Oedipus complex (Lacan, 2002a).

The Lacanian psychoanalytic hypothesis, reformulated within the framework of the model of an "*R-S*" interdisciplinary dialog, is in essence to see psychosis as being characterized by the absence of, or defective inscription of, certain traces whose function is homeostatic and structuring for our psychic development. This hypothesis of an absence of trace, which stems from a psychoanalytic theory of psychosis, offers an ideal formalization for an interdisciplinary dialog, since it draws on the paradigm of the trace that is common to neuroscience and psychoanalysis. Thus it forms a possible intersection between these two disciplines.

Research in neurobiology on the cellular and molecular mechanism of critical periods for cerebral plasticity, which shed light on the role of early interactions with the environment in certain psychiatric pathologies like schizophrenia, resonates with this psychoanalytic idea of an absence of traces in psychosis. Research on the critical periods for plasticity have demonstrated that the inscription of traces stemming from experiences with the environment is conditioned by various factors. Obviously, it is necessary that the interaction does take place – for example, in the case of the visual cortex, it is necessary that an effective environmental stimulation of the eyes is produced for the neural connections to be put in place. However, it is also essential that this interaction with the environment takes place during a precise temporal period – a timeframe situated between the moment of the onset of the critical period of plasticity for the region of the brain in question, and its closure.

The discovery in schizophrenia patients of certain anomalies at the level of the molecular and cellular mechanisms that determine the onset and closure of critical periods, represents a major area for discussion in the dialog with psychoanalysis regarding the absence of traces as the origin of psychosis; since critical periods play such a determining role in the inscription of early experiences through the mechanisms of plasticity. On that basis, we can envisage that the inscription of the first representations *R* that stem from exteroceptive perceptions (associated with the interoceptive perceptions of corporal states *S*) whose function is homeostasis, could only take place within a limited timeframe defined by a critical period. Thus, we can postulate that in psychosis the inscription of traces whose functions are structuring for psychic development, may not have taken place within that limited timeframe – either because the critical period was not open at the time of the interactions with the environment, or because the environmental stimulations did not occur at the appropriate time (as in the amblyopia example).

Different clinical models can be used to illustrate how breakdowns in the relationship with the environment can result in an absence in the inscription of traces that structure psychic development. Levine's study demonstrated that, confronted with a prolonged period of maternal separation in their early development, young rodents initially presented anxious and agitated behavior, but that subsequently significant manifestations of behavior similar to despair appeared. These were accompanied by important physiological changes. The body temperature of the young rats dropped significantly, as did their heart rate. This resulted in a degree of withdrawal and a lessening of motricity amounting to a kind of catatonia (Levine, 1957). A parallel can be drawn between these observations and research done in humans on the effects of family separation during the Second World War in the U.S. – notably the syndrome of hospitalism described by Spitz. This study highlights the psychic, cognitive and physiological consequences of emotional deprivation in children who were separated from family and raised as orphans (Spitz, 1946). A parallel can be drawn with the study done by Nelson that appeared in *Nature* in 2007 following the "Bucharest Early Intervention Project (Nelson III et al., 2007). The latter study concerned children raised as orphans during the Ceausescu regime in Romania, under extremely precarious circumstances from a emotional and material perspective. It showed that children who were place in foster families sufficiently early, demonstrated significant advantages in terms of emotional, cognitive and social development in comparison to those children who were placed only after 20 months. This occurred despite the two groups of children (raised in institutions or raised in foster families) both presented anomalies in the volume of gray matter when compared with children in a control group who had not experienced family separation.

These works are interesting for showing the psychic, cognitive and cerebral consequences of the absence of certain stimulating environmental interactions in the first months of infant development. The hypothesis for anomalies in critical periods in schizophrenia allows for a shift in relation to a direct linear causal view between certain environmental experiences and the psychopathological consequences. It should be underlined that an understanding of the molecular and cellular mechanisms for the onset of critical periods demonstrates that, even in the event of an appropriate and stimulating environment, neurobiological anomalies could have the same consequences for the brain and psyche as the absence of stimulation. It is from this pluri-factorial perspective, and not from a linear one, that the interdisciplinary hypothesis for the absence of trace in schizophrenia stems.

The onset of schizophrenia in adolescence as a consequence of the absence of trace

The hypothesis of a link between the physiology of critical periods and certain psychiatric pathologies has become a new field of research for neurobiology, notably in view of the delayed maturation of PV cells in the prefrontal cortex that has been observed. This late maturation, occurring at puberty, could be correlated with the generally accepted fact that the onset of schizophrenia typically occurs during puberty (Prochiantz, 2012).

From the psychoanalytic perspective, the triggering of psychosis is understood as a two-phase process. This process involves firstly that, during childhood, certain traces whose function is homeostatic and structuring for psychic development have not been inscribed in the subject. However, for the symptomatology to appear and for psychosis to be triggered in a manifest way, certain life events in the subject's trajectory need to solicit those missing traces (Lacan, 2002a). In as much as those traces concern, among other things, the Oedipal question – that is to say a relational complex with the parental figures, involving strong affective elements, as well as questions of origin, of reproduction, sexual identity and the difference of the sexes – all event that solicit these themes can lead to the triggering of psychosis (Lacan, 2002). Puberty, which raises the question of sexual identity and identifications inherited from the Oedipus complex, is a key moment for the revival of the Oedipal problematic. Therefore, this juncture can turn out to be conducive to soliciting those missing traces (or what Lacan terms "foreclosed signifier"), leaving the subject facing emptiness and an absence of meaning that prevents them from responding to the reconfigurations that take place at puberty (Lacan, 2002a).

From a biological point of view the physiological changes, notably hormonal changes, during puberty, also demand a process of adaptation and a significant reconfiguration in the relationship to the environment. As Prochiantz points out, in this regard schizophrenia could be linked to "an inability on the part of the brain, be it for genetic or developmental and epigenetic reasons – both are possible and can combine –, to mobilize the necessary plasticity for that adaptation" [our translation] (Prochiantz, 2012).

Although the disorder may already have an underlying presence before puberty – be it genetically determined, or epigenetically determined by early environmental interactions – puberty and the transition between the end of adolescence and the beginning of adulthood would seem to be a particularly favorable moment for the appearance of the symptomatology of schizophrenia; that is to say, for the expression of the phenotype that characterizes this pathology. One of the neurobiological hypotheses is that there is a link between the decompensations frequently observed at adolescence with schizophrenia patients, and the maturation of the PV cells in the dorsolateral prefrontal cortex that takes place only at the end of adolescence (Prochiantz, 2014). The hypothesis is that PV cells do not respond appropriately to the trigger (for example a hormonal trigger), just as if PV cells in V1 did not respond appropriately to eye opening. It could be advanced that it is only as the critical period comes to its end that the defects in inscription of certain traces, and thus the absence of structural modification to the neural pathways dependent on the environment, become fixed owing to the closure of the period of plasticity. Work on the cellular mechanisms of critical periods, undertaken by Prochiantz's laboratory at the Collège de France in collaboration with Takao Hensch at Harvard University, are specifically motivated by the hypothesis that schizophrenia finds its origin in a defect in development:

> *Following what is only a hypothesis, this defect would materialize around adolescence during a critical period of the prefrontal cortex's development […]. [E]ven if the phenotype appears at a late period of development, this does not entail that the "primary causes," for example one or more epigenetic mutations or modifications, did not take place at an earlier stage, or are hereditary […]. As regards schizophrenia, the hypothesis is that the prefrontal cortex does not adapt to the socialization that follows the hormonal maturation which precedes and accompanies adolescence.* [our translation] (Prochiantz, 2014)

This perspective echoes the structural psychoanalytic view according to which, in the decompensation of a psychotic episode, an event precipitates the onset of psychosis, thus making manifest an underlying psychopathological structure that predates the advent of that event. Freud illustrated this idea of a prior structure, with his crystallographic metaphor:

> *If we throw a crystal on the floor, it breaks; but not into haphazard pieces, it comes apart along its lines of cleavage into fragments whose boundaries, though they were invisible, were predetermined by the crystal's structure. Mental patients are split and broken structures of this same kind.* (Freud, 1933)

The psychoanalytic hypothesis of structures thus requires that with psychosis, the psychotic structure, although it is present before the manifest onset of positive symptomatology, will only become apparent ulteriorly following an event that precipitates the onset; just as a crystal that falls and shatters, reveals the lines of its structure in doing so.

From an excess of plasticity to a disorder in the stability of a sense of self in schizophrenia

In line with an interdisciplinary dialog between psychoanalysis and neuroscience on critical periods and schizophrenia, it seems fruitful to consider the hypothesis of a critical period for the inscription of fundamental traces that have a structuring function in psychic development. Indeed, a disturbance in the usual duration of critical periods for plasticity within the higher cortical pathways, may result in the non-inscription of certain traces linked to interactions with the environment, and thus to the absence of certain representations that could perform an organizing role for the whole of the psychic functioning. If we follow this hypothesis, according to Prochiantz there would be "no difference in nature between the mechanisms that lead to a benign condition like amblyopia – and a devastating illness such as schizophrenia" [our translation] (Prochiantz, 2012). In effect, just as amblyopia is the result of a failure in environmental stimulation within the visual cortex during a critical period for the acquisition of binocular vision (prevented by the reorganization of neuronal networks in that cortex owing to the absence of stimulation), schizophrenia may also be the result of an absence of the inscription of environmental stimuli within the neuronal networks (notably with the prefrontal cortex).

If there is a low PV cells maturation (as in the OTX2-AA mouse), then plasticity opens later and amblyopia can be induced at P100 instead of P30 (Lee et al., 2017).For a psychiatry disease, PV maturation could be too low to permit plasticity and thus adaptation to the post-pubertal adult environment is too difficult. It might also be that the hormonal adult environment is not accompanied by a proper social affective environment (e.g., maternal separation model in mice). The first situation (plasticity is not ready when needed by the environment) is more genetic, and the second one (environment is not appropriate when plasticity is ready) is more environmental. Indeed, any combinations between genetic and environmental factors are possible. So, in schizophrenia, the absence of trace inscription may be linked either to the absence of stimuli, or to the fact that the onset of the critical period had not correctly taken place at the time when the stimuli occurred.

As we have seen, different data support the presence of anomalies in the timing of critical periods in schizophrenia patients. Usually, in the prefrontal cortex, the maturation of the PV cells at adolescence induces a balance between excitation and inhibition circuits. In turn, this terminates the period of plasticity by fixing and rigidifying the neuronal circuits and consolidating E/I balance. In schizophrenia patients however, lacunae in the number of inhibitory synapses, as well as a reduction in the pruning of excitatory pyramidal neuron dendrites, can be observed (Insel, 2010). It is therefore a rise in excitation, to the detriment of inhibition, that takes place within the neuronal circuits of these patients. This in turn prevents the closure of the critical period (or its non-opening or late opening). As we have noted, these data should be considered alongside the immaturity of the PV expressing cells within the prefrontal cortex detected in schizophrenia, since a significant reduction in the expression of parvalbumin within the dorsolateral prefrontal cortex has been observed with this pathology (Chung et al., 2018). A link may therefore exist between this psychiatric illness and anomalies in the mechanisms of onset and closure of critical periods for plasticity. These would be critical periods that were abnormally prolonged, because of a defect of inhibition owing to the belated maturation of the inhibitory circuits (Mauney et al., 2013). Surprisingly then, with schizophrenia patients it would appear that from a physiological perspective it is not a defect, but on the contrary an excess of plasticity – linked to these anomalies in the duration of the critical periods during which cortical plasticity is at its peak – that is observed.

Brain plasticity is certainly an advantage from a biological point of view, since it offers a significant advantage in terms of adaptation. However, stability also plays a major role in the homeostasis of the organism. A tension, inherent to the living organism, seems to emerge between these two poles: the need to adapt to the environment through the neurobiological mechanisms of neuronal plasticity on the one hand; and the necessity for a form of rigidity the makes it possible to ensure the stability over time, of some neuronal pathways. The maturation of the PV cells, that triggers the end of a period of plasticity, thus constitutes an essential stabilizing factor that limits plasticity; a stabilizing factor that, by rigidifying the neuronal circuits, durably fixes certain traces, which then become less subject to remodeling and change. The fact that the critical periods for brain plasticity are framed by precise temporal limits can thus be the precondition for a certain permanence, a stability necessary to the construction of a sense of self that is durable despite the permanent changes to which the organism is prey.

Memory in particular, appears to be one of the factors that enables the introduction of the permanence necessary to the formation of our stability and our sense of identity:

> *Indeed, memory requires a minimum of structural permanency, cerebral in particular […]. [D]espite the reality of the morphological and physiological changes, which even in the brains of sapiens continues up to the end, there exists a curb on plasticity that, if left free rein, would erase the history as it was being inscribes in the structure of the brain.* [our translation] (Prochiantz, 2012)

Along similar lines, the neuroscientist, Damasio, has also proposed understanding memory as the root of our "autobiographical self." This is an autobiographical self that relies on our capacity to remember significant events in our personal history, and thus participates in constructing our identity (Damasio, 2000). This understanding can be put in parallel with the words of Prochiantz for whom our capacity to produce narratives around our history – be they close or distant to the reality, "on occasion mythical like the fantasies that surround roots" – are one of the fundamental pillars of the "construction of the individual, of the illusory consciousness that he has of being himself beyond the biological changes, that inscribe themselves, sometimes cruelly, in his flesh, brain included" [our translation] (Prochiantz, 2012). At a cellular level, the critical periods, and notably the maturation of the PV cells that induce their closure, could appear to be one of the neurobiological mechanisms that guarantees a certain fixedness of the neuronal networks, thus ensuring the permanence of those representations or mnesic traces at the root of our identity and our autobiographical selves.

Research by the laboratory of Pico Caroni has demonstrated the importance for long-term memory, as well as for memory consolidation, of a certain population of PV cells within the hippocampus. These neurons, whose early neurogenesis when compared to other PV cells that appear later on in development, are characterized by a strong expression of PV (Donato et al., 2015). The inscription of the traces that determine what Damasio terms autobiographical self (that is, memories of the past, the remembrance of which underpins our sense of identity) therefore involves older PV cells, since these memories necessitate the establishment of a consolidation and a firm stability to become permanently inscribed. Animal models would suggest that in schizophrenia patients a reduced population of these PV cells with a strong expression of PV can be observed – younger cells with a weaker expression of PV being in the majority (Carvalho, 2017). The consequence of this may be disorders in the process of consolidation within the hippocampus, something which has also been observed in other studies of these patients (Genzel et al., 2015).

If we consider the hypothesis of an absence of traces being at the root of psychosis in the light of the neurobiological data on defects in the inhibition of cortical plasticity – defects owing to anomalies in the timeframe of the critical periods in schizophrenia – it is possible that it is particularly the traces and representations at the origin of an autobiographical sense of self that have not been fixed and consolidated in these patients. These traces usually constitute the stable reference point in time that allows for the continuity in our sense of existence. The absence of these traces may thus be at the root of the impairments to the sense of self manifest in the psychopathology of psychosis.

Following the idea of "embodied cognition" developed by Damasio, our feeling of autobiographical self and of self-identity might specifically develop based on certain representations linked to the cartography of our organism, to the mental picture of our own body, within certain areas of the brain. The neuronal mechanisms that generate this representation may appear as the basis for a certain stability of the organism. A stability that Damasio calls "proto-self," which is at the root of the sense of self. It is from this stable representation of the body, cornerstone of a stable point of reference in time, that the sense of autobiographical self will subsequently develop. This is a sense of autobiographical self from which our sense of identity originates, beyond the permanent changes that model us:

> *what might give the brain a natural means to generate the singular and stable reference we call self" […] the possibility that the part of the mind we call self was, biologically speaking, grounded on a collection of non-conscious neural patterns standing for the part of the organism we call the body proper.* (Damasio, 2000)

The stable representation of the body, that comprises "a remarkable degree of structural invariance" (Damasio, 2000), offers a point of reference over long periods of time. Unlike another kind of quasi-instantaneous cartography of the states of the body, which is the object of continuous updating, and on which the mechanisms of homeostatic regulation rely, the stable representation of the body is ensured by other neuronal circuits. These are neuronal circuits with more stable neuronal maps that offer the fixed support of a perennial representation of the general structure of the body. This neuroscientific perspective – in which the neuronal circuits generate a stable representation of the body – represents precisely that reference point – guarantor of the stability and continuity of our conscience and our feeling of existence – that coincides with a psychoanalytic approach.

Following the psychoanalytic understanding, the primordial traces at the basis of the stability of our psychic functioning are the result of the representations and affects that are mobilized during interactions with the parental figures in the Oedipal stage. While, according to the Lacanian theory of the "mirror stage," it is at an even earlier stage of childhood development that the acquisition of a unified representation of our own body serves as basis for our "self" (Lacan, 2002b). This unified representation of the body does not stem from a representation that is directly linked to the state of the organism, but rather from a stable representation, acquired at an early age, through interactions with the environment and with the adults who themselves mirror back to the child this image of its unity. The image that the child gains of its own body as belonging to a unified whole, is therefore a cornerstone of subjective development, since it is a stable representation that is inscribed as a lasting psychic trace in the subject. It is a representation that will subsequently be the basis for a whole series of other traces or identification, on which the construction of a sense of identity will be formed.

In the Lacanian theory of the mirror stage, it is precisely this representation of a unified body that is absent in psychosis (Lacan, 2002b). This absence of a fundamental trace, which is the source of stability for the development of subjectivity and awareness of self, is made apparent with the onset of psychosis by the appearance of symptoms connected with an altered perception of the body. The experience of fragmentation, the strong presence of hypochondriac thematics, and coenesthetic hallucinations that are characteristic of the clinical picture of schizophrenia, may be understood as a consequence of this absence of a unifying and organizing representation (Lacan, 1997).

Freud had brought to light the existence of this altered perception in the representation of the body with his study of the Schreber case. In the initial phases the clinical picture was dominated by coenesthetic hallucinations and ideas of a hypochondriac nature (such as the feeling of no longer having a stomach, or of having certain organs such as the lungs damaged or even destroyed; Freud, 1911c). The introduction of narcissism would then enable Freud to disentangle the two phases of the psychotic process that consisted of a twofold libidinal movement. The first phase of psychosis consisted in a massive withdrawal of libidinal cathexis, and of all the psychic energy, from external objects. The libido flows back exclusively on the ego and the subject's own body. This first phase was characterized by the fact that the patient (notably those with schizophrenia) "…withdraws his interest from the external world completely…" (Freud, 1911c). The negative symptomatology of schizophrenia, such as being withdrawn, blunted affect, apragmatism, and deteriorations in representations of the body (coenesthetic hallucinations, hypochondria, and dysmorphophobia), are characteristic of this initial phase.

Freud postulated that this first phase is common to all forms of psychosis. However, in order to fight against this movement of libidinal withdrawal, some patients put in place a second process that would consist in an attempt to reinvest external objects with the libido. This second phase finds its expression in productive symptomatology such as auditory hallucinations and delusional ideas. In this perspective, Freud describes delusion as an "attempt at recovery" process (Freud, 1914c).

In the interdisciplinary model of schizophrenia as an "absence of trace," the delusional ideas can be equated to an attempt by the subject to produce new traces, new representations (R) by way of the delusion. This is in order to pacify the altered experience of the somatic states and the fear of fragmentation. It is an attempt, in the aftermath, to make up for the traces that had not been inscribed during development.

Psyochanalysis and neuroscience come together around the shared observation that certain traces and representations, lastingly and durably inscribed, constitute a reference point for our development and ensure a stability on which the sense of self, identity, and autobiographical consciousness can be built despite the continuous changes to which the organism is subject. The existence of critical periods of cerebral plasticity that are strictly delimited in time, constitutes an indispensable condition for the acquisition of a sense of self.

The anomalies observed in the molecular and cellular mechanisms that regulate the timeframes of the critical periods seen in schizophrenia patients, can therefore, owing to an excess of plasticity, have as consequence an absence of inscription or of consolidation of those traces that result from interactions with the environment. These are traces that offer a reference point, in terms of stability, for psychic development and the acquiring of a sense of autobiographical self. As we saw, this was the case also for the acquisition of a stable representation of a picture of the body, and according to Freudian theory for the psychic traces resulting from the resolving of the Oedipus complex. The absence of these traces, that usually form a reference point for everything that concerns procreation, access to parentality and maternity, and sexual identity in adult life, may in this way lead to the symptomatology that is characteristic of the clinical picture for schizophrenia; in particular symptoms connected with disorders in the perception and representation of the body, as well as an impairment of sense and awareness of self.

The reopening and reclosing of critical periods in schizophrenia: A therapeutic horizon

The dialog between psychoanalysis and neuroscience around the hypothesis of an absence of trace and anomalies in critical periods in schizophrenia may also open new horizons for treatment. Based on research into the importance of critical periods in the process of neurodevelopment at work in schizophrenia, a better understanding of the molecular mechanisms connected with the maturation of PV cells that conditions the onset and closure of critical periods, points to significant new therapeutic possibilities. Recent research on the molecular mechanisms of critical periods have brought to light the possibility of acting upon these mechanisms through pharmacological treatments, perhaps even reopening and reclosing certain periods of plasticity in adults. This research has raised the hope of being able to repeat certain stages of brain development, through the reopening and then closing of windows of plasticity. In particular, experiments have succeeded in reversing experimentally induced amblyopia in mice, enabling individual with amblyopia to regain normal binocular vision after the closure of the critical period for acquiring binocular vision. This was done by reopening a window of plasticity through the administering of a pharmacological drug within the visual cortex of the mice.

Several neuroscientific experiments have proposed different strategies to enable the manipulation and reopening of periods for developmental plasticity (Bavelier et al., 2010). The preferred approaches focus generally on neuromodulators such as dopamine, norepinephrine, or acetylcholine, as there are advantages to using drugs that are already approved for distribution (Bavelier et al., 2010). Experiments undertaken in the laboratory of Hensch have identified, in mice, the role played by a protein called Lynx1, which contributes to the plasticity inhibiting mechanism, and induces the closure of critical periods. The expression of Lynx1 contributes to maintaining the stability of mature cortical networks, as well as the suppression of molecular breaks that allow a modulation of the balance between the excitatory and inhibitory circuits that reactivate visual plasticity. For this reason, it represents an important avenue for the treatment of amblyopia, and opens up further interesting therapeutic prospects (Morishita et al., 2010).

Other similar studies have focused on the role of serotonin and norepinephrine in adult plasticity. These studies have successfully induced the reopening of critical periods by modulating the concentration of these neurotransmitters within the cortex (Spolidoro et al., 2009). Other scientists have for their part focused on another route towards the reopening of critical periods, notably through research into the role played by PNNs in the regulation of cortical periods of plasticity. Based on a study of the makeup of these extracellular matrices, which are organized in networks around neurons, Tommaso Pizzorusso's team have shown that their deterioration induced by hydrolysis with chondroitinase-ABC (chABC) leads to a reactivation of cortical plasticity in rats, thus enabling the reopening of the critical period for binocular vision (Pizzorusso et al., 2002; Harauzov et al., 2010).

The research done on the homeoprotein OTX2 that is internalized by the PV expressing basket neurons, by Prochiantz's team in collaboration with Hensch's laboratory, offers another interpretation of these results. The internalization of OTX2 is necessary and sufficient for opening then closing the critical period of plasticity in the visual cortex of mice. The teams have demonstrated that it is the perineuronal networks that surround the PV cells which capture the OTX2, by way of binding sites specific to the GAG present in that homeoprotein (Beurdeley et al., 2012). By identifying the molecules involved in these binding sites, the scientists have managed to demonstrate that the hydrolysis of the perineuronal networks by the chABC reduces the quantity of OTX2 present in the PV cells. This study has also shown that the direct infusion of a peptide, RK-pep, disturbed in a similar way the binding of OTX2 to the PV cells, reducing the expression of PV as well as the presence of PNN, thus reopening plasticity in the visual cortex of mice (Beurdeley et al., 2012). In the adult animals with amblyopia this momentary reopening of plasticity allowed a recovery of binocular vision.

There exists, therefore, different ways of reopening the critical periods for plasticity, using pharmacological means to act upon quantities of OTX2 (Despras et al., 2013; Testa et al., 2019). Modifying the binding of the homeoprotein OTX2 to the PNNs through pharmacological means, constitutes a potential new therapeutic tool. Such a tool might make it possible to restore the cortical plasticity not only in the visual cortex of humans, but also in other regions of the cerebral cortex. Based on the results obtained with amblyopia, scientists to come will be able to envisage new therapeutic horizons for other neurodevelopmental pathologies, and in particular for psychiatric pathologies such as schizophrenia.

Some studies have already sought to apply these ideas to animal models of schizophrenia, and have obtained significant results. Based on the hypothesis that the critical periods within different cortical regions are regulated by common mechanisms involving the maturation of PV basket cells, scientists have attempted to induce experimentally a structural, functional, or molecular lifting of the breaks that prevent plasticity in the areas implicated in the physiopathology of schizophrenia. In particular, Felipe Gomes and his team have looked at the critical period for the development of the ventral hippocampus that takes place in adolescence. This constitutes a period that is particularly sensitive to exposure to environmental stress factors. These can ultimately lead to a reduction in the physiological regulation of stress, and become a risk factor for schizophrenia (Gomes et al., 2019). The same team has also used sodium valproate, which has a strong inhibiting action, and which has been recognized for inducing a reopening of critical periods. Results have shown that administering valproate in adult mice enables a reopening of the critical period for vulnerability to stress, inducing a state of physiological maturation similar to that of adolescent mice. These data evidence the fact that adolescence is, from a physiological point of view, a critical period for acquiring a vulnerability to stress; a period that can be restored in adults through pharmacological means. These results are particularly important for our understanding of the physiopathology of schizophrenia, where this critical period could constitute a period of particular sensitivity to the impact of environmental factors that can contribute to the expression of a schizophrenia phenotype.

In so far as critical periods can be pharmacologically modulated, the experimental lifting of the various breaks on brain plasticity can be seen as a significant therapeutic step forward for schizophrenia symptoms. In the wake of the interdisciplinary hypothesis of an absence of trace at the root of this pathology, it is possible to envisage that a reopening of the critical periods for plasticity in certain regions of the brain could enable the replaying of some developmental processes; processes that had been disrupted or hindered the first time around. Thus, ultimately, it may be possible to achieve a re-inscription or a consolidation of certain traces that had not been stabilized during development.

If we follow the hypothesis that the lack of inhibition linked to the abnormalities in the maturation of the PV neurons that regulate critical periods could therefore, in the case of schizophrenia, be associated with an excess of plasticity, a therapeutic strategy could be the closure of the plasticity, with the aim of consolidating the traces that could not be stabilized during development.

But if we concentrate on a defective maturation of PV cells, following the hypothesis that maturation could be so low that the first threshold (plasticity opening) is not even passed (which means that the schizophrenia phenotype would be a consequence of an absence of plasticity when needed to repose to an environmental change), the therapeutic strategy will be different. The objective will then be to open critical period of plasticity, provide appropriate environment stimulation, and then close the critical period. This process would allow a long-lasting traces inscription.

If we consider schizophrenia as a neurodevelopmental pathology, linked in particular to anomalies in the duration of critical periods (anomalies that prevents the consolidation and the stabilization of certain representations which have a regulating and homeostatic function in psychic development) we could suppose that the reintroduction of plasticity within certain regions of the adult brain could allow, when associated with certain environmental stimuli such as psychotherapy, for a consolidation of the association between certain traces with a homeostatic function, thus making them more stable and perennial. This could be a way of attempting to replay the fundamental *R-S* tie that had not been the object of a lasting inscription during the subject's development. The potential for a psychological treatment would thus be increased, insofar as the return to a certain developmental plasticity would make it possible to inscribe and anchor in a more durable way the representations or signifiers evoked by the patient during sessions. Neurobiology's search for a chemical means of reopening critical periods would thus constitute a new therapeutic perspective for psychosis. It may even be an important trump card that would increase the effects of a talking cure, becoming a significant pharmacological aid in the psychoanalytic treatment of psychosis.

Conclusion

The interdisciplinary hypothesis of an absence of trace in psychosis, guided by research on anomalies in critical periods observed with schizophrenia, makes it possible to reconsider this pathology through an analogy with a developmental pathology such as amblyopia. Thus, seeing schizophrenia as consequent to the absence of inscription of certain fundamental traces linked to early interactions with the environment. Even when these early interactions, and the social and affective stimuli they imply, have taken place, an anomaly in the timeframe of the critical periods can prevent the inscription and the fixing of the neuronal traces stemming from these interactions. Cerebral anomalies identified by brain imaging in schizophrenia patients, may therefore be (as is the case with amblyopia) the result of this absence of reorganization in the neuronal networks that takes place through the influence of experience and environmental stimuli.

Research into the molecular mechanism that regulate the onset and closure of critical periods for brain plasticity have brought to light the regulation of these mechanisms – for example through the expression of the transcription factor OTX2, and by the PNN that surround the PV neurons. This demonstrates that genetic factors, as well as environmental ones, can act upon the timing of critical periods. The dialog between psychoanalysis and neuroscience that has developed around the cellular anomalies observed in the regulation of critical periods in schizophrenia patients, fits into a perspective that goes beyond the narrow opposition between genetic and environmental factors that has been linked to the etiology of this pathology.

Finally, neurobiological research into the pharmacological reopening of critical periods for plasticity represent a promising new therapeutic horizon, in that it may open the way to a reintroduction of developmental plasticity within certain cortical regions, in adult schizophrenia patients. Combined with environmental stimuli, this reopening could make it possible to consolidate or reinscribe certain associations of traces that had initially not been inscribed in a stable and perennial way during the subject's development. If these possibilities remain to date only in the realms of fiction, they could, ultimately, lead to new interdisciplinary therapeutic strategies between psychoanalysis and neurobiology, in the treatment of schizophrenia. The possibilities for a psychological treatment, could be vastly increased using chemical methods aimed at reintroducing a degree of developmental plasticity. The dialog between psychoanalysis and neuroscience centered on the role of critical periods in schizophrenia, shows itself to be rich in promise as regards new approaches in the treatment of this pathology.

Data availability statement

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author.

Author contributions

JT is the main contributor of this manuscript as part of her postdoctoral research. FA and PM supervised the research, contributed to the conception and development of the research, and revised critically the manuscript for intellectual content. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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Parents' dilemma: A therapeutic decision for children with spinal muscular atrophy (SMA) type 1

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Background: SMA type 1 is a severe neurodegenerative disorder that, in the absence of curative treatment, leads to death before 1 year of age without ventilatory support. Three innovative therapies are available to increase life expectancy.

Purpose: (i) To increase knowledge about parents' experiences with their decision to have opted for an innovative therapy; (ii) to assess the middle-term psychological consequences in the parents' lives.

Methods: We used an in-depth interview; a self-administrated questionnaire and self-report scales (BDI-II, STAI-Y, PSI-SF, SOC-13, PBA, DAS 16 and FICD). We compared parents hesitant before the decision to parents who were not-hesitant and the group of parents whose child was treated with gene therapy (GT) to parents whose child received another innovative therapy.

Main results: We included $n = 18$ parents of 13 children. Parent's mean age was 34.7 (±5.2), child's average age was 44.3 months (±38.0). Retrospectively, most parents felt involved by doctors in decision-making on treatment, they felt their point of view was considered and were satisfied with the effects of the treatment. The group of parents "non-hesitant" was more depressed ($p < 0.001$), more anxious ($p = 0.022$) and had higher parental stress ($p = 0.026$) than the group of "hesitant" parents; the group of "GT-treated" parents was more depressed ($p = 0.036$) than the group of parents with "other therapy". Qualitative data highlights revealed: the need to save the child's life at all costs; the fear of coping with end of life and palliative care, the high value of perceived physician confidence in the treatment, the hope that the child will acquire autonomy or be cured. At the time of the decision, no parents felt they fully understood all of the issues regarding therapy and the disease.

Conclusion: Hesitating before making a decision did not predispose parents to depression and anxiety. The narratives suggest that the parents faced a dilemma regarding their child's health in an urgent context. The decision was not final, and parents will continue to think about it throughout the care process.

KEYWORDS

dilemmatic decisions, palliative care, parents, caregivers, spinal amyotrophy type 1, SMAPAR Study, burden, innovative therapies

Introduction

Spinal muscular atrophy (SMA) is a degenerative disease of the motor neurons that, emerge from the spinal cord and brainstem. It is due to an alteration of the survival motor neuron 1 (*SMN1*) gene on the two alleles of maternal and paternal origin in the genome of the affected child. Parents transmitting the anomaly never develop the disease, as it is an autosomal recessive disease. It is the leading cause of pediatric neurodegenerative genetic disease, with an incidence in France of 120–150 births per year (15–19 new cases/ 100.000 births) and the second most common genetic pediatric disease after cystic fibrosis (1–3). These numbers are likely overestimated as only 35–40 new cases per year have been evaluated during national multidisciplinary consensus meetings (MCM) of reference centers for rare neuromuscular diseases (FILNEMUS) as of June 2018. SMA type 1 is the most severe and common form (50%–60%) of SMA. It appears within the first 6 months of life and is characterized by significant muscle weakness and very little motor acquisition (the independent sitting position is not acquired). Bulbar functions (swallowing disorders) and respiratory (ventilatory disorders) functions are also impaired, the latter being a key factor in the severity of the disease (1, 2, 4). In France, the various natural histories come from the follow-up of nearly 200 patients over 20 years (1999–2016). The patients received active palliative care but no long-term ventilation (non-invasive ventilation or tracheotomy). They showed a rapid evolution with a median age of diagnosis at 3 months (range 0.6–10.4) and a median age of death at 6 months (range 1–27) (5). A subgroup of children (type 1c) was defined by onset between three and 6 months (acquired head posture) usually with longer survival but a very severe disability (2). In other hand, *SMN2* gene has been found to modulate the age of onset and severity of loss of function of the *SMN1* gene (6). The first clinical trials began in 2008 and led to the approval for the first innovative therapy in the USA in 2016 (7), then in Europe in 2017 (8).

Currently, three innovative therapies[1] can increase life expectancy and improve motor and respiratory functions. Clinical studies show greater and faster efficacy when treatments are initiated early and ideally at a pre-symptomatic stage (9). Unlike in other countries, systematic neonatal screening is not yet available in France, which limits the possibility of treatment immediately from birth (6). A pilot project, DEPISMA, is being tested in two French regions. While these therapies change the natural course of the disease and bring new perspectives for patients and their families, many uncertainties remain (10) and families and medical teams navigate between "isolated islands of certainty in an ocean of uncertainty" (11).

In France, the therapeutic management of children with SMA needs to be ratified during MCM of reference centers for rare neuromuscular diseases (FILNEMUS) (12). Doctors discuss treatment indications based on the clinical (motor, bulbar and respiratory functions), the child's genetic profile and the available scientific data. They are thus faced with the ethical dilemma of choosing between palliative support through the end of life, or implementing an innovative therapy. The decision is most often broached with parents during the diagnostic confirmation visit and the final decision is an emergency. When the medical team advises palliative care, but the parents want an innovative therapy, they may experience the decision as an imposed sentence and often

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[1] Spinraza® (INN: Nusinersen) is an antisense oligonucleotide that increases the production of functional *SMN* protein by acting on the splicing of the *SMN2* gene. This is a very well tolerated treatment but requires an injection every 4 months into the cerebrospinal fluid via a lumbar puncture (intrathecal route) for life after a loading dose of 4 injections; Zolgensma® (INN: onasemnogene abeparvovec) is a gene therapy treatment designed to deliver the normal gene into defective motor neurons via a viral vector (AAV9). This is a one-time IV treatment that requires the injection of high doses of viral capsids that can cause serious side effects; Evrysdi® (INN: risdiplam) which is a motor neuron survival protein 2 pre-mRNA splicing modifier that increases the production of functional *SMN* protein. It is a very well tolerated oral treatment (syrup) requiring daily intake for life.

seek a second opinion. When the MCM proposes innovative therapy, the final decision about whether to treat the child is always up to the parents. They may sometimes interpret this proposal as a solution repairing the shock of the announcement of the child's illness, particularly when the SMA is severe (13). After the final decision, therapies are most of the time initiated 2 weeks after the genetic diagnosis has been confirmed.

In this urgent context, parents of children with SMA type 1 are faced with a dilemma: the decision to either consent to innovative therapy or to pursue palliative care and accompany their child through the end of life.

The aim of this study is to describe how parents experience their prior decision regarding an innovative therapy for their child with SMA type 1. This is a psychoanalytically oriented clinical psychology research based on a triangulation of quantitative and qualitative data.

This article presents the results of the subgroup of parents of children with SMA type 1 of the French cohort, of the SMAPAR study: a large Franco-Chilean study looking at the experience of parents of children (aged 1–18) with SMA type 1, 2 or 3 and the feeling of parental overload.

Objectives: (i) to increase knowledge about parents' experiences on their prior decision for an innovative therapy for their child with SMA type 1; (ii) to assess the middle-term psychological consequences in their lives.

Patients and methods

The inclusion of parents in the SMAPAR study took place in France between January 1, 2021 and September 30, 2021, within specialized neuropediatric departments (FILNEMUS) who chose to participate and through patient associations: French Association against Myopathies (AFM-Telethon) and Together Against SMA type 1 association (ECLAS). The parents who participated did so voluntarily and both parents within a couple could participate independently.

We used a mixed methodology with both qualitative tools (in-depth semi-structured interviews) and quantitative tools (self-questionnaire developed by members of the Steering Comitee of the SMAPAR study and validated in conjunction with members of patient associations and 7 self-questionnaires evaluating the medium-term psychological consequences on the parents). Complete methodology of the SMAPAR study is available in the **Supplementary Material**. Mixed methods propose to combine, in a more or less intimate way, quantitative and qualitative methods in order to produce results that combine credibility and meaning (14).

The study complies with MR-004 reference methodology that provides a framework for the processing of personal data for the purpose of studies, evaluations or research not involving the human person as studies that do not meet the definition of research involving the human person. It received approval from the Ethics Committee (No: #00011928, December 15, 2020) including the use of reported speech verbatim in publications. All participants signed written statements of non-opposition.

To meet the objectives of this study: we identified the variables related to therapeutic decision-making, mental health, and the impact of the disease on daily life and family in the catalog of data from the SMAPAR study:

(1) Variables from the in-house self-questionnaire: the self-questionnaire consisted of open and closed multiple-choice questions and visual analog scales (VAS) that ranged from 0 (lowest agreement or satisfaction score) to 100 (highest agreement or satisfaction score).

We isolated the following variables:

- *Parents*: sex, age, marital status (multiple choice), parental status (primiparous Y/N), economic status (multiple choice), professional status (multiple choice), social assistance and coverage status (multiple choice), perception of overall health (VAS) and impact of the disease on the mental and physical health (Y/N) of the parents, parental satisfaction with comprehensive care (VAS)
- *Children*: age, type of SMA (multiple choice), age at onset of first symptoms, age at diagnosis, type of therapy, time between treatment start and research participation (calculated variable),
- *Therapeutic decision*: parental participation in therapeutic decisions (Y/N) and satisfaction that their opinion has been taken into account by the medical team (VAS)
- *Treatment effects*: effects observed by the parents following treatment (Y/N), effects on motor function (multiple choice) and autonomy (Y/N), satisfaction with the observed changes (VAS), evolution of parental concerns regarding the health and future of the child (multiple choice) and evolution of the feeling of parental overload related to the disease and its management (multiple choice)

(2) Variables from the self-questionnaires evaluating the medium-term psychological consequences on parents: depression—Beck Depression Inventory, BDI-II (15), state-trait anxiety—State-Trait Anxiety Inventory, STAI-Y (16, 17), parental stress—Parental stress Index short form, PSI SF (18), sense of coherence—Self-questionnaire, SOC-13 (19), parental burnout—Parental burnout questionnaire, PBA (20), marital satisfaction—Dyadic Adjustment Scale, DAS 16 (21), impact of the child's disability on the family—Family impact of Childhood Disease, FICD (22, 23).

(3) Variables from semi-structured interviews transcribed and analyzed with N'Vivo: "Therapeutic decision-making" sub-node.

Data analysis

Quantitative data from the in-house self-questionnaire and self-assessment scales were analyzed using SAS software (version 9.4; SAS Institute Inc., Cary, NC, USA). Data were reported as counts and frequencies (%) for qualitative variables and mean ± standard deviation for quantitative variables.

The thematic analysis allowed us to identify two parental attitudes towards decision-making: "hesitant parent" and "non-hesitant parent". We also identified a difference in parental ideas about gene therapy (GT) vs. other innovative therapies. This allowed us to perform a descriptive statistical analysis by subgroup using "hesitant" parents ($n = 7$) vs. "non-hesitant" ($n = 11$) parents and parents whose child received GT "GT-treated" ($n = 6$) vs. those with a child treated with "other therapy" ($n = 12$). The homogeneity between the subgroups was tested using the appropriate tests: the χ^2 test (or an exact Fisher test) for qualitative variables, and a Student test (or non-parametric Mann-Whitney test according to the distribution variable) for quantitative variables. The results were considered statistically significant when $p \leq 0.05$.

The interviews were transcribed and imported into qualitative data analysis software NVivo 10 (version 1.6.1). The encoding map was produced jointly by three experienced psychologists using deductive methodology: two main thematic nodes were defined based on the literature and the objectives of the SMAPAR study: "Parenting" and "Parental overload".

We then moved on to an inductive phase: three psychologists (SB, MA, and MG) analyzed the first three interviews together in order to identify the emerging themes, define the sub-nodes to organize the data hierarchically, and check internal validity. A research psychologist (SB) then coded the other 15 interviews from the coding map, which was gradually improved by the iterative interpretative approach. Rereading the interviews allowed us to verify and specify the sub-nodes. We present an analysis of the "therapeutic decision-making" sub-node, created within the parent node here: Parenting → Discovery of SMA → Therapeutic decision-making and it's 5 themes, some of which are broken down into sub-themes.

The interview guide and research methodology were pilot tested on one mother and one father prior to the study and adjusted according to their feedback. Excerpts of the interviews were translated to English to be used in this paper.

Results

Participants

The sample (**Table 1**) consists of parents of a child with SMA1 treated with an innovative therapy ($n = 18$), including 12 mothers (66.7%) and 6 fathers (33.3%). The average age of the parents was 34.7 years (±5.2); 15/18 parents (83.3%) were married; 3/18 parents (16.7%) were separated from the other parent of the affected child at the moment of assessment (2/3 had another partner and 1/3 were single); 10/18 parents (55.6%) participated in the research jointly as a couple, 10/18 parents (55.6%) were first-time parents. The mean age of the child as reported by the parents was 44.3 months (±38.0); 10/18 parents (55.6%) reported that their child was classified at type 1 SMA and 8/18 (44.4%) of type 1c. They evaluated the average age of onset of symptoms at 3.5 months (±2.3) and diagnosis at 5.4 months (±3.2). The average time between the introduction of innovative therapy and research participation was 813.6 days (±455.2). The different therapies and management options proposed to the 18 parents are divided as follows: 11 parents (61.1%) innovative therapy at the time of diagnosis, 2 (11.1%) participation in a clinical trial, 2 (11.1%) a choice between a clinical trial or an innovative therapy, 2 (11.1%) a palliative (end of life) care, and 1 parent (5.5%, child 1c) was offered pro-active symptomatic care, as therapies were not yet available at the time of diagnosis. Finally, all children of participating parents had either received or were receiving innovative therapy at the time of the study. Note that 2/18 (11.1%) parents reported that their child was not treated, although the two children received a gene therapy injection. Regarding to the parents' socio-economic status, 2/18 parents (11.1%) were considered "low-income", 9/18 (50.0%) "working class", 5/18 (27.8%) "middle class" and 2/18 (11.1%) "high-income". Among the parents, 13/18 (72.2%) were working and 5/18 (27.8%) were unemployed; 15/18 parents (83.3%) received social assistance related to the child's education and or child's disability.

Data from the in-house self-questionnaire

The results of the descriptive analysis are presented in **Table 2**. When asked about their participation in therapeutic decision-making (treatment and care) for their child, only 1/18 parents (5.6%) did not feel involved. The parents felt their opinion was considered in the medical teams' decision with an average score of 80.6/100 (±16.6). All parents who reported that their child had received innovative therapy ($n = 16$) observed changes upon introduction of the treatment, including improvement in motor function (16/16, 100%), acquisition of new motor functions (12/16, 75.0%) and increased autonomy (10/16, 62.5%). Parental satisfaction with the observed improvements is on average 71.7/100 (±17.7). Despite the observed improvements, disease burden decreased for only 3/16 parents (18.8%), remained stable for 10/16 parents (62.5%) and increased for 3/16 parents (18.8%). Concerns related to the child's health decreased for 6/16 parents (37.5%), remained stable for 8/16 parents (50%) and increased for 2/16 parents (12.5%). Concerns for the child's future increased for 5/16 parents (31.3%), remained stable for 8/16 parents (50%) and

TABLE 1 Characteristics of participants (*n* = 18, homemade self-reported questionnaire).

All parents	Total *n* = 18	Non-hesitant *n* = 11	Hesitant *n* = 7	GT-treated *n* = 6	Other therapy *n* = 12
Gender of parent					
Male	6 (33.3%)	3 (27.3%)	3 (42.9%)	1 (16.7%)	5 (41.7%)
Female	12 (66.7%)	8 (72.7%)	4 (57.1%)	5 (83.3%)	7 (58.3%)
Age (years)	34.7 (±5.2)	32.6 (±5.1)	38.0 (±3.3)	32.8 (±6.8)	35.7 (±4.1)
Marital status					
Married	15 (83.3%)	8 (72.7%)	7 (100%)	6 (100%)	9 (75.0%)
Separated and in couple	2 (11.1%)	2 (18.2%)	-	-	2 (16.7%)
Separated and single	1 (5.6%)	1 (9.1%)	-	-	1 (8.3%)
Participation in couple					
No	8 (44.4%)	5 (45.5%)	3 (42.9%)	4 (66.7%)	4 (33.3%)
Yes	10 (55.6%)	6 (54.5%)	4 (57.1%)	2 (33.3%)	8 (66.7%)
Primiparous					
No	8 (44.4%)	2 (18.2%)	6 (85.7%)	1 (16.7%)	7 (53.8%)
Yes	10 (55.6%)	9 (81.8%)	1 (14.3%)	5 (83.3%)	5 (41.7%)
Economic status					
Poor	2 (11.1%)	2 (18.2%)	-	1 (16.7%)	1 (8.3%)
Popular	9 (50.0%)	6 (54.5%)	3 (42.9%)	4 (66.7%)	5 (41.7%)
Medium	5 (27.8%)	3 (27.3%)	2 (28.6%)	1 (16.7%)	4 (33.3%)
Easy	2 (11.1%)	-	2 (28.6%)	-	2 (16.7%)
Parent’s working					
No	5 (27.8%)	2 (18.2%)	3 (42.9%)	2 (33.3%)	3 (25.0%)
Yes	13 (72.2%)	9 (81.8%)	4 (57.1%)	4 (66.7%)	9 (75.0%)
Social benefits					
No	3 (16.7%)	1 (9.1%)	2 (28.6%)	1 (16.7%)	2 (16.7%)
Yes	15 (83.3%)	10 (90.9%)	5 (71.4%)	5 (83.3%)	10 (83.3%)
Age of child (months)	44.3 (±38.0)	46.5 (±48.0)	40.7 (±15.2)	25.2 (±3.4)	53.8 (±44.0)
Type of SMA					
Type 1	10 (55.6%)	7 (63.6%)	3 (42.9%)	3 (50.0%)	7 (58.3%)
Type 1 c	8 (44.4%)	4 (36.4%)	4 (57.1%)	3 (50.0%)	5 (41.7%)
Age of symptoms’ onset (months)	3.5 (±2.3)	3.5 (±2.7)	3.6 (±1.8)	2.8 (±1.5)	3.9 (±2.7)
Age of diagnostic (months)	5.4 (±3.2)	5.5 (±3.7)	5.3 (±2.4)	5.3 (±2.5)	5.5 (±3.6)
Treated with innovative therapy					
Answer no, but it was yes	2 (11.1%)	2 (18.2)	-	2 (33.3%)	-
Yes	16 (88.9%)	9 (81.8%)	7 (100%)	4 (66.7%)	12 (100%)
Type of innovative therapy					
Gene therapy	6 (33.3%)	5 (45.5%)	1 (14.3%)	-	-
Other therapy	12 (66.7%)	6 (54.5%)	6 (85.7%)	-	-
Time from research participation to start of treatment (days)	813.6 (±455.2)	666.8 (±375.9)	1002.3 (±505.7)	586.0 (±118.6)	889.4 (±503.6)

decreased for 3/16 parents (18.8%). The average score of parents, when asked about their overall health satisfaction, was 67.3/100 (±20.8). However, 16/18 of parents (88.9%) believe that SMA has impacted their physical health and 12/18 (66.7%) their mental health. There is no significant difference between the subgroups of “hesitant” parents vs. “non-hesitant” parents and the subgroups of “GT-treated” vs. “other therapy” for all the variables mentioned above.

Evaluation of medium-term psychological effects on parents’ lives

The results of the descriptive analysis are presented in Table 3. Overall, the parents do not have high depression scores (mean BDI score: 9.8 ± 6.1), 5/18 parents (27.8%) presented with mild depression and 1/18 parent (5.5%) with moderate depression. The sub-group analysis highlights that:

TABLE 2 Quantitative data from the in-house self-questionnaire (results are expressed as mean score ± standard deviation).

All parents	**Total** *n* = 18	**Non-hesitant** *n* = 11	**Hesitant** *n* = 7	***p*-value**	**GT-treated** *n* = 6	**Other therapy** *n* = 12	***p*-value**
Involvement in medical and therapeutic decision process				0.412			0.146
No	1 (5.6%)	1 (9.1%)	-		1 (16.7%)	-	
Yes	17 (94.4%)	10 (90.9%)	7 (100%)		5 (83.3%)	12 (100%)	
Satisfaction with the way their views were taken into account	80.6 (±16.6)	77.7 (±9.8)	84.7 (±23.7)	0.137	77.6 (±22.8)	81.8 (±14.4)	1.000
Satisfaction about their overall health	67.3 (±20.8)	63.1 (±14.6)	74.0 (±28.0)	0.291	62.0 (±20.5)	70.0 (±21.3)	0.458
Parents whose physical health is affected by the SMA				0.060			0.289
No	2 (11.1%)	-	2 (28.6%)		-	2 (16.7%)	
Yes	16 (88.9%)	11 (100%)	5 (71.4%)		6 (100%)	10 (83.3%)	
Parents whose psychological health is affected by the SMA				0.087			0.289
No	6 (33.3%)	2 (18.2%)	4 (57.1%)		1 (16.7%)	5 (41.7%)	
Yes	12 (66.7%)	9 (81.8%)	3 (42.9%)		5 (83.3%)	7 (58.3%)	
Parents who declared their child was treated	***n* = 16**	***n* = 9**	***n* = 7**		***n* = 4**	***n* = 12**	
Observed change related to treatment: Yes	16 (100%)	9 (100%)	7 (100%)		4 (100%)	12 (100%)	
Type of motor changes observed				0.771			0.182
Motor function improvments	16 (100%)	9 (100%)	7 (100%)		4 (100%)	12 (100%)	
And/or New motor function acquisition	12 (75.0%)	7 (77.8%)	5 (71.4%)		4 (100%)	8 (66.7%)	
Changes in level of concern about their child's health				0.881			0.641
Increased	2 (12.5%)	1 (11.1%)	1 (14.3%)		-	2 (16.7%)	
Same	8 (50.0%)	5 (55.6%)	3 (42.9%)		2 (50.0%)	6 (50.0%)	
Decreased	6 (37.5%)	3 (33.3%)	3 (42.9%)		2 (50.0%)	4 (33.3%)	
Changes in level of concern about their child's futur				0.866			0.915
Increased	5 (31.3%)	3 (33.3%)	2 (28.6%)		1 (25.0%)	4 (33.3%)	
Same	8 (50.0%)	4 (44.4%)	4 (57.1%)		2 (50.0%)	6 (50.0%)	
Decreased	3 (18.8%)	2 (22.2%)	1 (14.3%)		1 (25.0%)	2 (16.7%)	
Satisfaction with the changes observed following treatment	71.7 (±17.7)	70.3 (±14.8)	73.4 (22.1)	0.741	74.3 (±18.3)	70.8 (±18.3)	0.751
Changes in burden due to the SMA and its management				0.660			0.537
Increased	3 (18.8%)	2 (22.2%)	1 (14.3%)		1 (25.0%)	2 (16.7%)	
Same	10 (62.5%)	6 (66.7%)	4 (57.1%)		3 (75.0%)	7 (58.3%)	
Decreased	3 (18.8%)	1 (11.1%)	2 (28.6%)		-	3 (25.0%)	

the “non-hesitant” group of parents presented mild depression (13.2 ± 4.3) as compared with the “hesitant” group of parents who presented no depression (4.4 ± 4.5) ($p < 0.001$); the group of parents of a “GT-treated” child also presented mild depression (15.0 ± 3.6) as compared with the group of parents of child treated with “other therapy”, which presented no depression (7.2 ± 5.4) ($p = 0.013$).

With regard to anxiety, the parents presented a low level of trait (YB) and state (YA) anxiety on average (mean score Stai YB = 40.7 ± 9.7 and YA = 39.3 ± 8.8); 6/18 parents (33.3%) and 7/18 parents (38.9%) presented average trait and state anxiety scores respectively.

The sub-group analysis highlights that: the group of “non-hesitant” parents presents mild anxiety (average score YB = 45.1 ± 7.1; YA = 43.0 ± 7.6) compared to the group of “hesitant” parents who presents very low anxiety (mean score YB = 33.7 ± 9.5; YA = 33.6 ± 7.9) ($p < 0.05$); the parents in the “GT-treated” group show mild anxiety (mean score YB = 45.5 ± 4.4; YA = 42.7 ± 4.2) as compared with parents in the “other therapy” group who had very low anxiety (mean score YB = 38.3 ± 10.8; YA = 37.7 ± 10.2). There is no significant difference between the 2 groups.

At the time of assessment, the parents did not show significant parental stress (average PSI-SF score = 82.9 ± 22.0). However, according to establish criteria of the scale, 11/18 parents (61.1%) did suffer from clinically significant levels of stress. The analysis by subgroup underscores that the “non-hesitant” parents presents a clinically significant level of parental stress (average PSI-SF score = 91.8 ± 17.9) unlike the “hesitant” group who does not present parental stress (score mean PSI-SF = 69.0 ± 21.7) ($p = 0.027$). The parents in the “GT-treated” group and the parents in the “other therapy”

TABLE 3 Medium-term psychological effects on parents' lives (results are expressed as mean score ± standard deviation).

All parents	Total *n* = 18	Non-hesitant *n* = 11	Hesitant *n* = 7	*p*-value	GT-treated *n* = 6	Other therapy *n* = 12	*p*-value
Depression (BDI-II)	9.8 (±6.1)	13.2 (±4.3)	4.4 (±4.5)	<0.001	15.0 (±3.6)	7.2 (±5.4)	0.013
State anxiety (Stai-YA)	39.3 (±8.8)	43.0 (±7.6)	33.6 (±7.9)	0.022	42.7 (±4.2)	37.7 (±10.2)	0.270
Trait anxiety (Stai-YB)	40.7 (±9.7)	45.1 (±7.1)	33.7 (±9.5)	0.010	45.5 (±4.4)	38.3 (±10.8)	0.139
Parental stress (PSI-SF)	82.9 (±22.0)	91.8 (±17.9)	69.0 (±21.7)	0.027	83.7 (±18.1)	82.6 (±24.5)	0.925
Parental burnout (PBA)	26.7 (±19.0)	30.0 (±18.9)	21.6 (±19.4)	0.374	27.0 (±25.1)	26.6 (±16.4)	0.967
Sense of coherence (SOC-13)	57.8 (±12.5)	53.8 (±10.7)	64.1 (±13.4)	0.088	56.8 (±8.5)	58.3 (±14.5)	0.819
Dyadic adjustment (DAS-16)	114.4 (±21.8)	103.3 (±18.7)	127.2 (±18.5)	0.076	110.2 (±4.2)	117.2 (±28.2)	0.486
Negative impact of the disease on family life (FICD −)	27.4 (±6.3)	29.2 (±6.2)	24.7 (±5.9)	0.290	27.7 (±6.7)	27.3 (±6.4)	0.920
Positive impact of the disease on family life (FICD +)	33.2 (±4.8)	32.5 (±5.1)	34.3 (±4.3)	0.475	33.3 (±4.3)	33.2 (±5.2)	0.947

group did not show significant parental stress (mean PSI-SF score = 83.7 ± 18.1 and 82.6 ± 24.5, respectively). There is no significant difference between the two subgroups.

As a group, the parents presented no particular risk of parental burnout (average PBA score = 26.7 ± 19.0). However, taken individually, 2/18 parents (11.1%) presented a low risk of parental burnout and 5 parents (27.8%) a moderate risk. The mean internal consistency score (SOC-13 score) was 57.8 ± 12.5 and overall, the parents showed a good level of dyadic adjustment within their relationship (mean DAS 16 score = 114.4 ± 21.8). The subgroup analysis for these 3 scales indicates that there are no significant differences between the different subgroups.

Finally, examination of the impact of disability on family life (FICD) reveals both a moderately negative impact (27.4 ± 6.3) and a strong positive impact (33.2 ± 4.8). There was no significant difference between the different subgroups.

Qualitative data from semi-structured interviews

Thematic analysis of the interviews allowed us to define 5 themes within the "Therapeutic decision-making" node, some of which were further broken down into sub-themes.

Theme 1: Experience of the treatment proposal

The doctor's ability to deliver complete and reasoned information

According to the parents, how the neuropediatrician proposed the therapeutic strategy seemed to be based on his experience with clinical trials and new therapies. The parents found that doctors made proposals in a measured way. Their manner of proposing treatment on the one hand hinted at the epistemic uncertainty surrounding the innovative therapies, while also conveying confidence in their acquired experience since the beginning of use of the new therapies: both in terms of effectiveness as well as with regard for the clinical state of the child.

> *"The therapeutic trial was explained to us fairly well; we knew very well that there was no miracle. It was clearly explained to us that it would not cure the disease, but that it would reduce its effects. That being said, we had no idea what we were getting in to and we also knew that there was a part of the trial that used placebo"* (21)

> *"He explained to us that there was a new drug that had arrived which had been through a phase of tests and that with this drug, maybe he would live beyond two years, even up to 50 years, we didn't know. But he explained that there were not enough long-term results"* (17)

> *"We talked about it a bit with the doctor. When he told us about the appointment that had been made for the gene therapy trial, we said to him "please give us your opinion, we'll take it!" He was quite courageous, because he said that if it was for his children, he would do it without hesitation, he would not ask questions. So, it reassured us from a medical point of view, to say to ourselves "We don't know anything about this, but if we look at this person who is very interested and informed, and seems very professional, he would do it!"* (10)

> *"According to him, our son was not particularly ill, and it also did not seem to be a question of him not receiving treatment […] Anyway, I think that if we had absolutely wanted him not to be treated, I don't think the doctor would have necessarily objected. But to him, our son was a really good candidate for innovative therapy"* (13).

Doctors who convey incomplete information or false hopes

Conversely, some parents reported that when treatments were discussed at the time of diagnosis by doctors who they later perceived as less informed, the information was sometimes incomplete, and conveyed a prediction of the future or unreasonable hope.

> *"They were unable to tell us what we could expect with the therapy. At that time, she was already nine months old, so we said to ourselves: "we have three months left""* (11)

> *"The doctor told me about gene therapy and said, "we will be able to cure your son". And I asked him "what do you mean cure him? She replied: "Ma'am, he will be able to walk! ". That's the only thing I retained. But when we went to meet another doctor, at the expert center, it was a rude awakening. She examined him and told us that she did not think he was particularly affected and that she wasn't sure if the therapy would work on him, she was very honest and she gave us a few days to consider our options"* (41)

The sense of urgency in decision-making

Given that the prognosis of the child was at stake, the parents' testimonies highlight the context of emergency in which the decision was made. The parents recounted that the doctor conveyed the need to treat the child as quickly as possible just after making the diagnosis.

> *"She told us that she was giving us a little over 24 h to think things over, because we had to act quickly. At that moment, my husband and I looked at each other and we said to ourselves: "It's not worth it, we are not going to wait 24 h. If there is something to try, we'll do it now…"* (41)

> *"The doctor said: "But we really have to make up our minds because it's a race against time and you have to treat your son now, he can't wait any longer, we don't have time."* (13)

Parental ideas about innovative therapies

At the time when the decision was made, the parents remember having had vague ideas about innovative therapies. Some parents understood that these treatments would not cure their child and that there was uncertainty about the expected effects. Others evoke the idea of a miracle or a cure that had been conveyed by certain professionals, social networks, and/or the media. In addition, the parents who were offered gene therapy mentioned greater efficacy and a less restrictive mode of administration than the other alternatives. They also cite a potential risk of greater side effects, but this risk does not tip the balance on their final decision.

> *"We knew that it was partly random, we didn't know why it worked better for some children than for others"* (13)

> *"The doctors told us: "She will be able to walk". So, they ignited hopes that were unrealistic at the time"* (60)

> *"I asked him, "What's the difference between Spinraza and Gene therapy?" Is there really a noticeable difference? "She said to me: "When it is introduced before symptoms, there is a real difference" And I also said to myself: "He won't have to have epidurals every four months in his spine", I found it less burdensome on a daily basis for our family, for our child."* (44)

> *"At the time, I think we said to ourselves, it's a bit like a magic wand. And they're going to give us back our little girl, the one we had when she was born."* (32)

> *"We had some fears: won't this have other effects that we have no idea about today? Because it goes in the blood, we do not know exactly what other risks might be associated with the treatment"* (10)

Theme 2: Parental attitudes at the time of the decision

Accepting without hesitation to save the child's life at all costs

Among the 16 parents who received approval for an innovative therapy, 9 parents accepted that their child be treated unequivocally regardless of the proposed strategy.

> *"Spinraza had been on the market for about six months. So here it is! We didn't hesitate, we were told about it and we said: "Well, if there's already a treatment, we're not going to take the risk of doing nothing."* (14)

> *"Since it was that or nothing, well we went for it"* (21)

> *"We had understood that without innovative therapy, he would not make it to age 2 and that, maybe with this treatment, he would get better. He might never be able to walk, but maybe he could live. So, if there was something to try, it was logical that we would try it."* (41)

> *"We said to ourselves, we are going to put all the odds in his favor. In any case, we had nothing to lose. We were told he was going to die before the age of 2"* (17)

Hesitating before making a decision

Some parents ($n = 7$) hesitated between pursuing a treatment or pursuing end of life care due to the uncertainty, the seriousness of the disease, and the fear of imposing a life of suffering on their child in the form of severe disability.

Several mention the risks of both options: (i) pursuing palliative care might also mean taking the (low) risk that the child will survive with a severe disability; (ii) accepting innovative therapy means accepting the risk that the treatment will not be effective, particularly if symptoms were already present. The parents highlight the dilemma they had to face between two solutions marked both by uncertainty.

Among the 7 hesitant parents, 2 parents had initially chosen to refuse the treatment proposal, then changed their minds after a second opinion from an expert center where they had been informed that their child suffered of a later and more slowly progressive form of SMA (1c) and that he would most certainly survive for years to come, even without innovative therapy.

> *"I said to myself: "Before doing anything, we will have to have very specific opinions on: What state is he currently in? What can we expect and what should we do? For my part, I wasn't ready to let him live at any cost""* (44)

> *"My husband and I didn't really know what to do because at the time, it was not presented as a treatment. It was more palliative, more as support for a death without suffering, in fact. So, my husband and I took 48 h to respond, we didn't really know what to do."* (17)

> *"We still ask ourselves the question, "is it this the life that I want or not? "We were still given the possibility of doing nothing and accompanying him towards his end…. with a small risk that he will in fact live, with a huge handicap and without being able to do anything. We ask ourselves all of these questions because having a disability is not the life we had wanted for us, nor for him"* (9)

> *"In the beginning we were told that this little girl, she was going to live less than a year that she was surely going to have a tracheostomy, a gastrostomy that her heart was in danger of stopping. And we said to ourselves: "Okay, but we aren't going to impose additional treatment on her" […]. We said to ourselves "this is still a very important question for us, so we need a second opinion." And in fact, when we saw the doctor, he told us: "Of course she needs to be treated [….] in my opinion, without treatment she can live until the age of three". And we said to ourselves "there are actually children like her, adults like her and maybe, even with this disease, she will be able to have her own child. In the end, we suddenly fell in love with the idea of her destiny and that really made the decision for us"* (11)

Theme 3: Whose decision is it?

A shared decision between parents and doctors

The majority of parents feel that they took part in the decision thanks to a trusting relationship with the medical teams.

> *"In general, I think that the, well, I think we can talk a little about collaboration between the medical team and ourselves because, yeah, we are involved, and they are involved as well. She is really very, very good, we are really…Well, we realize how lucky we are to have this doctor and this whole medical team"* (13).

Parental responsibility for the decision

For 5 parents, the decision was a radical choice between the life and death of their child. Faced with such a dilemma, parents reacted in a variety of ways: 1 couple chose to delegate their decision to the doctor, as they found it to be an impossible responsibility, another couple shifted the decision-making to their child, somehow entrusting him with the responsibility to choose whether he wanted to live or not, and finally, 1 parent took on the responsibility to make the decision with his partner, but not without guilt.

> *"It did not seem to us to be our role as parents to have to make a choice, to say he must have this therapy, or he must have this other therapy. So, for us it was clear. And for the doctor it was also clear that ultimately he would be the one to make the decision"* (12)

> *"We agreed that we didn't want to choose between life and death because it's not up to us to choose, it would be up to him. And despite his young age, I think children if they want to live, they live, if they don't want to live, they don't live. We said to ourselves, we're going to put the odds on his side."* (17)

> *"There are very few cases in life where you are responsible for the life or death of your child. We, however, gave birth to a child with a fatal illness. So already, there was an extra weight on us. And afterwards, we said to ourselves: "Are we forcing him to live? And at what cost? "but it's still us who have to decide. So twice in a row, we said to him: "We gave you this life, we will make you survive"* (44)

A feeling of dispossession, an imposed decision

In our study, 3 parents felt that the decision had been imposed upon them either because the choice of therapy was made by the MCM without taking into account the parents' viewpoint, or because they had not received approval for their child to receive an innovative therapy.

When palliative care is proposed but the parents prefer their child to be treated, it feels like a door slammed shut. These parents believe that the decision was imposed upon them, and they experience it as a theft of their parental authority.

> *"She explained to us that in any case, it is not us who decide on the therapy she will be able to have. It is a group of doctors who decide after analyzing […] We understood that it was really the doctor who decides because it was ultimately a medical decision"* (32)

> *"Clearly, we were advised against it, we were not told: "You can try", we were told: "No. Don't do it" right?"* (3)

> *"Uh, he didn't advise us at all. In actual fact, we were not given a choice. We were told, "Gene therapy, don't count on it. And Spinraza, we submitted an application and in committee, it wasn't accepted because he was too ill"* (4)

Theme 4: Seeking information and support from the medical teams

The internet and patient associations

After the diagnosis and treatment proposal, one of the main sources parents report finding information is the Internet. They report feeling a need to learn about the disease and new therapies on French and international websites. They also hope to find other more detailed information or testimonials from other parents. Several parents consulted patient association sites or were referred to them by their doctor to find out about the disease, or to seek support when palliative care was being considered. In this specific case, contact with a patient association allowed a parent to develop his ideas about palliative care thanks to a conversation with a parent who had been through it with his child. It also allowed some parents who had initially been offered palliative care to seek a second opinion at an expert center.

> *"We looked for a lot of information on the internet: what is this treatment? What is gene therapy? There was a study in the United States two or three years ago, on 12 children. So, we were looking for all the documents which we were then trying to translate. Our life revolved entirely around that"* (9)

> *"I had a conversation on the phone with someone who told me about the death of his child… so I asked him a lot of questions about the circumstances of his death, and it did me a lot of good. It was interesting because we needed to hear from other parents who had been through a similar experience"* (11)

> *"We said to ourselves, we are going to meet parents who this has happened to, we'll discuss their experience in order to have some moral support for being with our child in end-of-life care. It was my wife who contacted the association, the woman asked her questions about our son's health and right away, she suggested a second opinion. On my end, I didn't necessarily believe it was worth it and that's when my wife was put in touch with another mother who put us in touch with her neuropediatrician"* (9)

Support from the medical teams

Overall, the majority of parents who were offered innovative therapy said they felt supported by the medical teams throughout the decision-making process. They describe professionals with a sense of humanity who were available and committed to them. Nevertheless, one parent highlighted the difficulty of interpreting the quantity of information given to them at the time of diagnosis. In contrast, the couple who was initially offered palliative care evoked the lack of support from the medical team when they challenged their decision and obtained treatment in another center.

> *"When we made our decision, it was also about the support from the team. We knew we would have the answers to our questions that we were really given all the information we needed. But it was up to us to interpret and understand the information how we wanted to understand it… I'm not saying that the support was the best"* (17)

> *"They never had the same perspective as we did. We were in action mode while they were in end-of-life mode. And when our son was not doing well, even then, they, they would say, "well, we told you so. You shouldn't have expected anything else! Since his pediatrician didn't believe in the treatment, well suddenly everything she said was pushing in that direction."* (3)

Theme 5: The experience of decision-making in the medium term

Satisfaction with the observed effects

The majority of parents describe themselves as satisfied with the progress they have observed in their child following treatment. This satisfaction is essentially linked to their child's motor progress and improvement in respiratory function. Note that their satisfaction is not related to the child's autonomy. The fact that the child was able to survive is deemed a miracle by some parents and a source of hope despite the child's severe disability. On the other hand, five parents describe mixed satisfaction with the treatment: because the child encountered medical problems following its implementation, because the child's progress is not up to the standard of parental expectations, or because signs of the disease appeared unexpectedly to the parents.

> *"It's been three weeks since we started a new treatment and we still have the impression that he talks a bit, well he babbles a lot more than before. He's starting to be able to turn from his back on both sides and come back on his own. He's been in really great shape since December and that's been really good for us."* (13)

> *"Right now I have a four-year-old girl, not one with spinal muscular atrophy, but a four-year-old girl with a motor delay"* (14)

> *"She is 5 and a half years old, it's a miracle. It's awesome. It is indescribable. Finally, I'm actually happy"* (22)

> *"So, he can't hold his head up, he can't stay seated or anything at all really, but hey, well, we can still see that the treatment is working and that something is happening. So, we're being patient and we're telling ourselves that over time it will happen"* (3)

> *"He can sit up, but we can't leave him sitting alone, because we're still afraid that he will fall. So, it's more of a type 1, but we still have the life of parents who have a young type 2. So, we say to ourselves: "At the moment, it is how it is, even if it doesn't evolve positively, it won't evolve negatively."* (44)

> *"There are things that are going well and there are things that can't be controlled. And scoliosis is one of the things that's described, especially when the treatment comes a little later on"* (10)

A decision that doesn't go away

In retrospect, parents perceive the diagnosis as confronting them with the helplessness and uncertainty of a future marked for premature death or severe disability, their discourse evokes decision-making as the only way of taking action. That being said, parents report that when making the decision, they did not understand all of the issues relating to their child's illness and its treatment.

At the time the decision had to be made, it was impossible for the parents to imagine what the future of their child might be or the impact of the disease on their daily and family life in the short and medium term. It was also impossible for them to envisage how end of life care for their child would be.

According to the parents, it is not as if the decision is final, but rather a decision that continues to be relevant and which they will reexamine at different times throughout the course of care or the development of the child. This is particularly true when the child's health is worrying and the risk that he or she suffers from a severe disability is confirmed.

In these moments, the parents wonder if the decision to support their child in palliative care would not have been better and 4 parents mention their fear that their child will 1 day blame them for the choice that they have made on their behalf.

> *"Very quickly we were told: "You have the opportunity to do something!". And then the first day, it's like a huge void. But we reacted fast because we knew that we had to act quickly in order to succeed in getting the most decent life possible for him, so we moved to action quite quickly."*(9)

> *"At the time I had not realized the impact on respiratory function. When I was told muscle, I didn't think of that. And swallowing was ultimately much more significant than the impact on motor muscles. So, it's true that at the time, I didn't understand what it was going to produce in terms of breathing and swallowing and that's our son's main issue."* (13)

> *"I told them, "Listen! I can survive the death of my child". But then, witnessing the end of his life, I really don't know how it will happen, I don't know if I will be able to do it. And I told them, "Listen! If it doesn't go well, I'll smother him under a pillow"* (44)

> *"I hadn't understood what it would entail: this amount of care, of time throughout the day.... or anything really. I didn't realize, what it was going to mean in my life, you know?"* (22)

> *"It's true that seeing your child suffer, being unwell, to have the impression that it's something chronic and that it's improving, and then they relapse. At certain times we ask ourselves: "were all these efforts really necessary and is treatment necessarily the best solution? "Then again, we also don't have the impression that it is relentless or overly aggressive"* (12)

> *"It happens less and less, but there are still times when I say to myself: "Oh my God! I would rather he was dead, you know?" Well, even now […] I think it'll only be when he's an adult and he can say to me: "Wow, Mom! It was so worth it" that I will could think to myself, "We made the right decision."* (44)

Discussion

This is the first study in France to examine how parents of children with SMA type 1 experience the decision-making process around innovative therapy for their child. The participants in our study were mainly mothers, which is consistent with participation rates found in the literature (24).

The average age of parents in our study is slightly higher than the average age reported in the only other study providing information on the age of parents of children with type 1 SMA (25). Most parents in our sample were married and reported a good level of marital satisfaction. We believe that the young age of their children and the hope that a treatment offer provides may have a protective effect on the couple, an effect which remains to be demonstrated over the long term.

Parents describe a high degree of satisfaction with the effects observed in their child following the start of treatment as is consistent with caregivers' evaluations found in the literature. According to Audic et al., parents seem to be overly optimistic; this over-optimism may reflect the considerable hopes invested in these new therapies (8). Despite this, when asked if the burden related to the disease and the care of their child had decreased since starting treatment, results show that it only decreases for a minority of parents. In our study, we did not measure parental responsibility quantitatively as we felt there were no relevant tools to do so. Brand et al. point out that studies based on quantitative methodologies report an average to moderate burden (24, 26–29) in parents of children with SMA. However, these studies used the Zarit scale or the Caregiver Strain index, scales initially developed to assess the burden of caregivers of patients with dementia and/or the elderly and which have not been validated in a population of parents of children with degenerative diseases (30, 31). In addition, we believe that the many cares related to their child's disease carried out by the parents of our study might be perceived as expected for the care of any young child whose autonomy is limited, and therefore not perceived as a burden. In addition, a large proportion of the parents in our study were primiparous parents who could not have representations of what constitutes parenthood of a disease-free child.

The parents' satisfaction with the observed treatment effects also contrasts with their concerns with the future and health of their child. Concerns decreased for only 33.3% and 16.7% of parents after starting treatment. Indeed, despite the availability of innovative therapies, which change the course of the disease, a cure is still not possible (24). Uncertainty regarding the long-term efficacy of treatments raises ethical, medical, and financial questions for families (24). Accordingly, parental concerns are reported in the literature regarding the cost of innovative therapies, side effects and complications that therapies could induce (32, 33). These concerns also relate to the alteration in the quality of life of the child and their psychological distress linked to becoming aware of their difference and disability (32, 33).

The rate of depressed parents (33%) was found to be similar to that reported in the literature (34), but the level of depression in our cohort was mild to moderate, whereas Cremers et al. observed a high level of depression (34). On average, the low trait and state anxiety of our cohort contrasts with the high level of anxiety described in the literature among mothers (34). Note that the tool used by Cremers et al. to measure maternal depression and anxiety (HADS) is not the same as the one we used, limiting its comparability (34). Furthermore, the parents in the "hesitant" group are significantly less depressed ($p < 0.001$) and less anxious ($p = 0.022$) than those who were "not hesitant", independently of their satisfaction with their child's progress after starting treatment. In our study, we attempted to determine the psychological states of the parents quantitatively using the BDI-2 and STAI scales. However, our qualitative approach leads us to believe that these scales cannot fully capture the very complex feelings parents report. Even though the interviews show similarities in themes, the experience reported by each parent was unique. The decision-making process seems to be very intimate and shaped by the personal history of each of the parents, which seems to be brought to the fore when confronted with such an extraordinary defy of parenting imposed on them by the disease. It seems important for us to point out that the majority of parents had never met with a psychologist and that this research was for many the first opportunity to recount their experiences at their own pace to someone with deep respect for their experiences. The psychologist who conducted the interviews (SB) perceived an intensely traumatic sentiment among the parents' accounts, particularly regarding the experience of misdiagnosis and then receiving a diagnosis, illustrating the intensity of what they have experienced on a psychological level. In addition, the psychologist herself experienced intense countertransferential feelings during the interviews, such as shock, blankness of thought, avoidance behaviours such as thinking about something else, and a feeling of helplessness. As the interviews progressed, she also described several somatic manifestations (fatigue, a feeling of physical overload), difficulties in getting back into the analysis of the interviews and a need to take care of herself. We believe that these manifestations are part of vicarious trauma, a term proposed by Laurie Pearlman and Karen Saatvine in the 1990s to describe the traumatic experiences of therapists working with survivors (35). It is now described in the therapeutical relationship, when the stories of patients have a traumatic effect on the therapist due to their repetition, which is associated with significant personal changes as a result of empathic engagement with the patient (35).

Furthermore, we might ask ourselves if the ambivalence at the time of the decision and the subsequent anticipation felt by some parents when presented with a treatment proposal may be protective. This could be pertinent for neuropediatricians who may perceive parental hesitation as an obstacle to decision-making. In reality, this first period of uncertainty could be considered necessary in order to integrate the complexity of their experience and begin the mental work needed throughout the evolution of the disease.

Similarly, in the context of neonatology, it has been shown that parents who could anticipate difficulties during their pregnancy were better prepared to face them when the child had to stay in neonatology (36). It should also be noted that the parents of children who received gene therapy presented a significantly higher level of depression than those who received another therapy. This leads us to wonder if the proposal of gene therapy may induce an idealized vision of the treatment's effects or excessive hope of a cure, leading then to depressed mood when the parents are confronted with the current reality and forced to let go of that hope.

More than half of the parents reported high parental stress and more than a third of parents were at a low to moderate risk of developing parental burnout. The parental stress score is higher than that observed in the literature in parents of children and adolescents suffering from various neuromuscular diseases including SMA (37%, measured with the Parental Stress Scale) (37). According to Von Gontard et al. families of children with SMA, show high levels of stress compared to matched controls. The author points out that these levels of stress seem to be influenced by social support, the child's behavior, and the degree of disability (38). However, the use of different tools to measure parental stress makes the results difficult to compare and to our knowledge, no other studies have looked at parental burnout in parents of children with SMA. Furthermore, the “hesitant” group of parents did not present clinically significant levels of parental stress compared to the “non-hesitant” group. On the other hand, the parents of the “hesitant” group were not first-time parents. It is possible that having other healthy children may play a protective role in dealing with a new child's disease.

In our study, we adopted a salutogenic approach by focusing on the feeling of internal coherence: an important factor in psychological adjustment to stressful situations (39). The literature emphasizes that a high SOC score indicates better resistance to stress (39). The mean SOC score of parents in our study is lower than the mean score reported in a study that compared SOC in $n = 368$ parents of children with intellectual disability and/or autism and $n = 387$ parents of normal children (40). The literature points out that a weak sense of internal coherence is more strongly associated with the risk of parental overload, depression, and anxiety (39, 40), which does not seem to be the case in our research. However, the absence of norms to describe the SOC means we are not able to interpret its value in our population. Additional analysis to investigate the link between SOC scores and those of depression, anxiety, and parenting stress would allow us to see if the parents with the lowest SOC scores are also those with the highest depression, parental anxiety, and stress scores.

The fact that parents report a more positive than negative impact of the disease on family life, leads us to believe that the parents who agreed to participate in our study had a positive and combative attitude towards the disease. These results can therefore not be generalized to all parents, constituting a bias in our study.

The interview analysis reveals 3 determinants in the treatment decision-making process of parents of children with type 1. (i) **Giving your child every chance**: the literature on parents' expectations of the first approved therapy, Nusinersen, emphasizes that the majority of parents hoped that the treatment would modify the course of the disease, prolong the life of the child, improve their quality of life and allow the child to take charge of their illness and live with the most autonomy possible (24, 32, 33, 41); (ii) **Trust in the prescribing physician**. Kiefer et al. emphasize that the relationship with the medical team is the most important factor influencing parents' final decision. The author specifies that to feel supported, parents need to receive sufficient information delivered in a neutral fashion with regard to the parent's decision by a doctor with expertise in the innovative therapies (33). Two parents in our study had initially chosen not to treat their child but changed their minds after seeking a second opinion from an expert center doctor who told them their child would not die. (iii) **Save the child's life and/or avoid being confronted with palliative care**, the notion of saving the child's life is never addressed in studies that have focused on therapeutic decision-making. In contrast, in their study on parental decision-making around the use of invasive ventilation in children with type 1 SMA in a palliative care setting, Pechman et al. point out that ventilation was the only possibility for parents to influence and prolong the life of their child (42, 43).

In our research it appears that retrospectively, parents believe they have been involved in medical decisions about their child. This finding is consistent with the work of Beenaert et al., which emphasizes that health professionals do not make decisions without informing parents (44). However, much like in our study in which several parents report having vague information, the author states that nearly 1/3 of the parents felt insufficiently informed about SMA and its treatments. Pechman et al. also point out that the parents of children with type 1 SMA diagnosed before new therapies, had not received sufficient information to understand all the issues surrounding the disease and palliative care at diagnosis. The authors emphasize that the traumatic context of the diagnosis and its repercussions are not conducive to understanding all the information. In addition, parents reported that doctors provided abstract and prosaic information that contrasted with the emotional intensity of their situation and led parents to seek further information from parent associations, social media, and the Internet (43). In contrast to these observations, a recent study conducted among parents who had received a neonatal diagnosis of SMA for their child reports that the advice given by the doctor had an impact on their decision for 83.3% of the participating parents and that it constituted the main influence over information from other health professionals, social networks, family and friends (45). In 2020, experts came to a consensus concerning the use of gene therapy in severe cases. They agreed that gene therapy could stabilize the disease without necessarily reducing disability or the child's quality of life (46). Following this publication, Gusset et al. were concerned that such a statement would leave little room for joint decision-making with families, would be misinterpreted by less experienced clinicians, would bias families' perspectives, and disrupt their decision-making process (47). The authors insisted on the need for the families concerned to be in contact with patient associations to develop realistic ideas about the life of a person and family with SMA and take part in medical decisions (47). In response to Gusset et al., Kirschner et al. highlight the need for parents not to be influenced in one way or the other. They emphasize the importance of explaining the wide range of outcomes associated with this therapy to parents, mainly because the public image is often idealized and seen as a cure, as in healing completely from SMA (46, 48). It should be noted that in our study, two parents stated that their child had not benefited from an innovative therapy when they had received gene therapy. When asked to elaborate on their response, the parents said that a treatment that did not cure the disease could not be considered as a treatment.

This surprising finding deserves more consideration with regard to families in long-term care as it is likely that the evolution of the disease will impose technical adaptations and a loss of function that the treatment will not be able to curb. Some parents may later feel disappointed and disillusioned with treatment and may consequently be reluctant to accept the prescribed medical recommendations to manage the child's disability. The innovative character of new therapies, in particular that of gene therapy, could be seen as a pharmakon (a philosophical idea denoting remedy, poison, and scapegoat). On the remedy side, it conveys hopes of healing, while on the poison side it induces fears about long-term side effects.

Retrospectively, several parents in our study reported that they had not understood all the issues around treatment when the decision was made and had constructed idealized representations based on what the doctor told them, information found on the Internet and/or transmitted by the associations, as well as their desire to save their child. We also noted that the families seem to have a perception of a hierarchy of innovative therapies leading them to prefer gene therapy as it seems to them more effective and less restrictive than other therapies despite the lack of long-term data on their safety and efficacy. Deng et al. (45) report similar findings about the criteria influencing therapeutic choice for parents of children who have undergone neonatal screening.

Our research has several limitations. First, like with most research on rare diseases, the size of our sample does not allow us to generalize these results to all parents of children with SMA type 1. Second, our study may be subject to selection bias, as the parent volunteers were mainly recruited from specialized consultations by family neuropediatricians and patient associations with close ties with the families.

Conclusion

While the availability of innovative therapies is radically changing the management of SMA and the lives of families, the uncertainty of how well innovative therapies are tolerated over the long-term forces parents and medical teams to face particular dilemmas. In the absence of more reliable data and neonatal screening to initiate pre-symptomatic treatment, parents are faced with an impossible decision between the early death of their child and the risk that treatment will not allow their child to live independently.

The therapeutic decision appears to not remain in the past but instead reappears as the disease and its care progress. When observing their child's progress, the parents feel reassured. Conversely, when disability worsens or no progress is made, parents may wonder if they have made the right decision for their child.

The urgent context in which this decision takes place, just after the announcement of the diagnosis, highlights the importance of frank and open discussions with the doctor to provide comprehensive information and answer the questions that parents may have during the treatment proposal. Moreover, the urgent nature of this decision should spur medical teams to systematically propose a meeting with a psychologist with expertise in SMA whenever possible and at a minimum, after the moment of decision-making. In our opinion, this is an essential part of helping parents prepare for the sequence of decisions that will have to be made as treatment and the child's development progresses as well as for forging an alliance of trust between families and the medical team.

Data availability statement

The raw quantitative data supporting the conclusions of this article will be made available by the authors, without undue reservation. In compliance with the General Data Protection Regulation and to preserve the confidentiality of study participants, the raw qualitative data can't be shared.

Ethics statement

The studies involving human participants were reviewed and approved by Name: Comité d'éthique et de la recherche APHP.5 (CER APHP.5). Affiliation: Assistance Publique Hôpitaux de Paris. The patients/participants provided their written informed consent to participate in this study. Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

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Author note

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Author contributions

MG, SB, MA, ID, OB, MN, AI, SQ, CC, MJ were responsible for the study conceptualization and design. SB, MG, MA, MN, SQ, CC were involved in the specific self-questionnaire conceptualization. LO and NB were involved to validate the research process and the qualitative and quantitative tools. MG and SB were responsible for obtaining funding and ethical committee authorizations. SB was responsible for creating the e-CRF in REDCAP, enrolling parents, collecting and managing data, and ensuring the conduct of the study. She had full access to all the data in the study and took responsibility for the integrity of the data and accuracy of the analysis. SB, MA, and MG were responsible for the analysis and interpretation of data as well as drafting/revising the manuscript and its intellectual content. CS was responsible for performing statistical analysis of quantitative data and revising the manuscript. ID, CB, OT, AI, MN, LO, NB were responsible for the recruitment of parents. ID, OT, CS, SQ, CC were responsible for revising the manuscript. SMAPAR study group took part in the recruitment of parents. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fped.2022.1062390/full#supplementary-material.

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The complexity of the dialogue between psychoanalysis, neurosciences, and genetics

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Introduction

The question of psychogenesis and organogenesis is among the oldest and—according to Ey (1)—the most complex chapters in psychiatry. At the start of the 19th century this debate was expressed through the opposition between functional and organic disorders. Other terms such as endogenous—most often opposed to exogenous or, alternatively, to reactionary, psychogenic, neurotic or even sociogenic—are almost simultaneous variants of the previous tandem (2, 3). In recent years, with the development of genomics and neuroscience, the etiology debate in psychiatry has tended to shift toward the interaction between the genome and environmental factors (the so-called "exposome"), as early on as the gestation period [see (4)]. A much more recent functionalist conception of the body-mind problem sees the brain as the hardware and the mind as the software, but Kendler (5) considers this dualistic conception to be an unfortunate combination of 19th century Cartesian neuropathology and computer functionalism. Nevertheless, the discoveries on neuronal plasticity [see (6)], with the observation that lived experience inscribes traces in the nervous system that can be re-combined with later experiences, provide the possibility to go beyond the opposition between psychic causality and organic causality. Thus, genetic and environmental factors could contribute in their own way to the configuration of these traces but also to their re-combination. Moreover, Crow has proposed (7) that "schizophrenia is the price Homo Sapiens pays for language". Specifically, Crow has hypothesized that, for this very reason, the genes for schizophrenia must be related to the genes that make language possible. Moreover, in his 1946 Bonneval speech, Lacan argued that "Not only can man's being not be understood without madness, but it would not be man's being if it did not bear madness within itself as the limit of his freedom" [(8), p. 144]. Thus, views on psychopathology from fields as removed as genetics and psychoanalysis may unexpectedly echo one another.

The complexity of genome-environment interaction in psychiatry

Kendler and Eaves (9) distinguished three main models (and their possible association) for the interaction between genotype and environment: additive effects of genotype and environment, genetic control of sensitivity to the environment, and genetic control of exposure to the environment. Thus, it would appear that genetic factors may sensitize or predispose individuals to exposure to particular environmental factors or even to experiences with depressogenic potential. For the same authors (9), genetics influences on human personality are probably related to traits such as impulsiveness, personal stability, and frustration tolerance. These traits could predispose to life experiences such as work and relationship changes which, in turn, can induce depression. Furthermore, genetic heritability in psychiatry is governed by complex mechanisms, since the same genes or the same genomic segments in copy number variation (CNVs) do not produce the same phenotypes and phenocopies can occur (these are environmentally-induced phenotypes which are identical to phenotypes that are genotypically determined in other individuals). Moreover, in psychiatry, heritability is not considered to be purely genetic and discoveries made since the 1980s through. Kendler suggests (10) that family twin and adoption studies (FTA) indicate that there is both environmental and genetic inheritance. Thus, "[t]he claim that FTA studies 'prove' that a disorder is'biological' is weak. An astonishing wide array of human behaviors and traits, such as hours spent watching television, sports participation, church attendance [. . .] are heritable" [(10), p. 1058]. He also claims (10) that a substantial proportion of genetic variation results from large numbers of small effect variants, each of which has a limited impact. Genome-wide association studies of schizophrenia (11–13) and manic-depression (13, 14) confirm this claim. Furthermore, with regard to autism, Munnich [(15), p. 89] argues that: "Since 2014, our exploration has included targeted sequencing of 250 genes known and published to cause autistic syndrome [. . .] This approach has shown us the high frequency of accidental, genetic but non hereditary events [. . .] there is no single autism gene, but dozens, even hundreds of distinct mechanisms that contribute to autism. Making the diagnosis of a genetic disease that presents as autism in no way precludes the use of psychotherapeutic approaches". Even where genetic inheritance seems plausible, it is mostly non-specific and, furthermore, we cannot know whether what is inherited relates to specific nosological entities [which are, as Kendler (10) puts it, clinico-historical constructs] or a pathophysiological pathway. Which may predispose either to vulnerability to one or more clinical types or to one or more possible clinical expressions of the same. Moreover, Post and Weis (16) suggest that a particular genotype may predispose also to protective or adaptive mechanisms and other authors argue (as will also be seen below) that genes can predispose to creativity, which can be passed on alongside genes for vulnerability to certain psychopathologies.

From a single cause to a multitude of possible causes

Kendler (17) claim that in the second half of the 20th century, psychiatry sought to distance itself from psychoanalytic domination and to re-establish its medical legitimacy through research into theories positing a single cause, much like Bayle [(18), see also (19)] had succeeded in isolating tertiary syphilis as the sole cause of general paralysis insane. For one thing, this complexity moves us away from the idea of a single causality and, for another, the plurality of causes means that each individual case is unique. As Ansermet and Giacobino [(4), p. 10] put it: "Rather than presiding over the repetition of sameness, genetic determinism poses the question of the production of difference. Inter-individual variations and the defining of singularity also become crucial questions for genetics". What is more, "psychoanalysis involves the subject and its evermore unpredictable genesis, which relates to a gap between cause and effect that cannot be bridged" [(4), p. 74]. In this dialogue between genetics and clinical human sciences, the kind of psychopathology that takes into account the singularity of each case—namely, psychoanalytically-oriented psychopathology—surely has its place. In the light of neuroscience advances in neuroplasticity, the neuropathology of so-called mental illnesses should not abolish psychopathology. On the contrary, as is the case with psychosomatic diseases where pathological modification of tissues is a prerequisite for them to be qualified as psychosomatic, neuropathology does not contradict the "psychosomaticity" of so-called "mental illnesses". Indeed, for Lacan (19), psychosomatic phenomena have to do with the gelification of the signifying chain: signifiers, being sometimes gelified, lose their signifying function and, as a result, become signals from the other and obtain an imperative value for the organism. Peirce's phaneroscopy helps us [see (20–23)] to conceptualize this as a process of semiotic reduction that moves from signifying tierceity to the secondness of the signal (which could condition or provoke reactions) or, even beyond, to the primacy of complete automatism. Even beyond the framework of classical psychosomatic phenomena, this process of semiotic reduction would be conceivable also for other clinical states related to homeostatic circuits of the brain, as I argued elsewhere regarding schizophrenia (20, 21), manic-depressive psychosis (22–24), depression (24), catatonia (25), addictions (26), traumatic neurosis (27), and panic disorder (20, 27).

The Freudian approach has always taken into account organic—genetic and epigenetic—factors in the determinism of psychological disorders. Freud certainly did not reject organicity as a cause. In fact in his concept of the *complementary series* (28), two factors converge in fulfilling an etiological requirement, and in his concept of *somatic complacency* (29), every hysterical symptom involves the participation of both sides, psychical and somatic, the latter offered by some normal or pathological process "in" or "connected" with one of the bodily organs. Hence, these two concepts were meant as a conversation with the psychiatric theories of his time—namely, the theory of the "constitution". Lacan, for his part, far from excluding organic causes in the determination of psychoses and neuroses, referred to what could be a sensitization [(8), p. 182–83] or conditioning [(19), p. 207] of human soma.

Let us recall in this regard the sentence that closes Lacan's 1946 text [(8), p.182–83] on psychic causality: "I do not hesitate to say that it will be possible to demonstrate that the Oedipus crisis has physiological resonances—and that, however purely psychological it may be in its scope, a certain dose of Oedipus may be considered as having a humoral efficacy of the absorption of a desensitizing drug". And the action of psychotropic drugs—which we could describe, at least in several cases, as desensitizing processes [see (22, 30)]—can resonate with the much more complex and most efficient desensitizing processes that psychoanalysis allows us to observe. Indeed, an infant's soma is conditioned by the signals of her primordial Other, that is until she becomes desensitized to these signals, through the triadic—mainly linguistic—processes inherent in ternary semiotics, the properly human semiotics. Nevertheless, sometimes, as a described above, through a "semiotic redaction", produced both by psychic and biological predispositions, the human body may once again become attuned to signals that induce "automatic states". Affects for instance—which rely on the metaphorical and metonymic processes of the unconscious—may be reduced to automatic emotions and/or moods (20–24, 27)—which loop and sustain themselves. Besides, in a letter to Fliess dated 1 January 1896 (31) Freud (K manuscript) recognizes heredity as a further determining factor, in that it promotes and increases pathological affect. Furthermore, Freud (32) in his paper, of the same year, "Heredity and the Aetiology of the Neuroses" gave also an important role in heredity regarding the causality of psychic suffering.

An opportunity of renewed dialogue between psychoanalysis and psychiatry

The dialogue between psychiatry and psychoanalysis is by no means incompatible with neuroscientific and genetic studies. On the contrary, such interaction may reveal points of inertia in subjectification. Indeed, organic factors—amongst others—may interfere with the abovementioned desensitization and, consequently, with the subjectification of a given individual insofar as they interfere with his or her interactions with others, especially in the early years, which we know from psychoanalysis that they are foundational years. This is why intensive psychoanalytically inspired screening and psychotherapeutic work during these early years is crucial [e.g., (33, 34)]. Developmental psychology and neurodevelopmental studies also engage on this subject, with the concept of critical (or sensitive) periods [e.g., (35)] which allows precisely to bring new lights to the interaction genes/environment. Recent studies [for an overview see (36)] claim that there are neurobiological mechanisms (genetically determined) which regulate the impact of the environment during some precise "time windows" and also that these mechanisms can themselves be modified by the impact of certain environmental stimuli [for an discussion on this issue including psychoanalytic theory with a focus on schizophrenia see (36)]. However, the so-called "desabilitys" that can emerge when there are barriers to the knotting of the body with the symbolic, should not be reduced to deficiencies as they appear in the latest psychiatric classifications as psychiatric syndromes of the "neurodevelopmental disorders" category. The scientistic spirit of these taxonomies disregards the subjectivity of the so-called "mentally ill". Indeed, their "desability" is primarily an interpersonal issue which must be addressed by clinicians who understand the meaning of "madness". This should only additionally involve rehabilitative and medicinal treatments.

Biological psychiatry can interact with psychoanalysis either synergistically or antagonistically. In many cases of psychosis and autism neuroleptics can reduce the alienation felt by patients and, thus, facilitate transference and psychotherapeutic work and rehabilitation. This can typically happen in cases of coenesthetic and/or sensory invasion, pervasive feelings of persecution or states of agitation that, in the words of Czermak (37) (for person suffering from psychosis), render transference "irresistible" and "traumatic". On the other hand, the immoderate use of antidepressants (38), neuroleptics (39), and lithium (40, 41) can, in a number of cases, result in losing motivation to address important issues, or even in emotional apathy, anhedonia, decreased libido and attenuation of creative capacities.

On the genetic predisposition that is common to certain psychopathologies and creativity

Jamain [(42), p. 311] claims that epidemiological studies (43) regarding "Bipolar disorders" allow us to estimate that the proportion of "the disease explained by genes is about 60–80%, whereas the effect of the environment would contribute to about 40% of the disease" [see also (11, 14)]. According to Masson and Brun (44), the basis for "manic-depressive illness" is the pathological drop in the threshold of emotional reactivity, making also reference to Ey (45) who wrote about in his *Psychiatric Studies* as early as the 1950s. According to Masson and Brun (44), the emotional hyperreactivity that results from cerebral dysfunctions can be amplified by certain traumatic, behavioral, or toxic phenomena that trigger critical episodes. However, several studies have documented the possibility of a parallel genetic transmission of the propensity for creativity or certain forms of intelligence and vulnerability to certain clinical types. For example, manic-depressive psychosis is reported to be much more common in writers, especially poets (46–49).[1] According to Andreasen (46) creative individuals have a particular cognitive style that both predisposes them to creation and leaves them vulnerable to thymic fluctuations. In this study, the predisposition to creativity also concerned their "healthy" relatives whose creativity was not otherwise limited to writing. Of course, the mechanisms of the correlation between madness and creativity are far from being elucidated. Is there a common mechanism for both? Or is it rather an effect of unusual experiences—as might also happen under the influence of psychedelic drugs—or even an attempt to synthesize divergent states or stimuli[2] that push toward creation. Probably there is more than one cause here, but as we shall see in the conclusions, psychoanalytic theory has something to say about this issue.

Discussion

The fact that all of us harbor something that is "out of the norm", a form of "madness", is by no means something to take offense at, but rather it allows us to relate to what is abnormal and specific about each individual—both the suffering or even deficits, and the originality and inventiveness. It has been established that the same genetic variants can predispose to symptoms but also to original creativity, and even, as Baron-Cohen et al. (53) suggest in the case of autism, to talents in fields that require systematization.[3] As Malafosse also concludes, "it is possible that certain susceptibility alleles of psychiatric disorders constitute an advantage for the people who carry them and for society as a whole" [(54), p. 48]. Creativity and psychopathology are in no way incompatible, and one could even argue that many times is quite the opposite. . . .

Since Freud, there have been discoveries about artistic creations by means of sublimation (55) and about delirium as a healing process (56). More recently, Lacan contributed his concepts of "suppléance" (supplementation) (57) and the "sinthome" (55). According to what Lacan says in 1975 (58) about James Joyce's writing style, language [which meant to have the function "not to inform but to evoke" [(59), p. 247], as he claimed back in 1956] has the main property

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[1] For other authors such as Sclesinger (50) the association between creativity and "mad geniuses" is overstated in the professional and popular press.

[2] For which the terms "janusian process" (51), "overinclusive thinking", and "cognitive inhibition" (52) have been used.

[3] This particular intelligence would be linked to sensory hypersensitivity and hyper-systematization.

to make us enjoy, beyond any sense—even equivocal—at the same point that it remains, according to Soler, “enigmatic” [(60), p. 18]. Scientists (61) have recently found that the language of apes is more “robust” than that of humans, but the latter is more efficient, “enabling better exploitation of information capacity and complex use of the neural vocabulary to adapt and learn new environments [(61), p. 606] [. . .] In turn, it can contribute to fragility underlying human psychopathologies” [(61), p. 596]. In my opinion, human language is apt to “deviate” in order to “treat” the enigma of the social Other’s desire. This again brings us back to the relationship of language to psychosis, at the genetic level. And to conclude, as Lacan [(62), p. 477] noted: “the only organicity that is essentially involved in this process: the organicity that motivates the structure of signification”.

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Becoming a mother when suffering from a chronic illness

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Introduction: The critical period of early motherhood when facing serious health problems constitutes a major public health issue. The disease may interfere with, influence, and compound the difficulties experienced over the course of pregnancy and during the parenthood processes. These processes are to be understood as a passage involving so many changes and fraught with difficulties leading to a series of psychological mobilizations. Illness also needs to be considered as a moment of transition, sometimes of severance, marking the lives of the people concerned in a more or less lasting way. Various developments are possible, some of which can be envisaged as leading to more positive outcomes, while others appear as if blocked or doomed to failure.

Clinical data and method: This clinical study is the result of twelve analytically oriented psychotherapeutic follow-ups. The sessions took place weekly over periods ranging from 1 to 6 years. Some of the patients we met had become pregnant while they had a declared but not yet chronic nephropathy. The other patients were already on dialysis and had become mother before starting dialysis. There is also one instance of a pregnant patient on dialysis. In the background, there are also many women who talked about giving up fertility and motherhood. After an in-depth study of each follow-up, a cross-sectional study was conducted to identify the main themes.

Results and discussion: Main considerations on the psychotherapeutic treatment: We regularly observe people who find it impossible to muster the internal resources that would enable them to deal with the trials they undergo in such situations. For the psychologist, there is a need to listen to archaic remnants. Gradually, in the space of psychotherapeutic work, possibilities of "reanimation" of the psyche emerge, an internal space that renews fantasmatic activity as it begins to be able to project into the external space and into the space of the sessions.

KEYWORDS
pregnancy, perinatal care, links, chronic illness, psychotherapeutic treatment, reverie

1. Introduction

Little has been written about the desire to become a parent and assuming all this entails when suffering from a serious somatic disease. In the medical field and also in psychology, work on women who are seriously ill and pregnant mainly relates to acute illnesses and cancer [for example (1–6)], or AIDS [for example (7–12)]. A growing interest in other severe conditions (13), including Chronic Kidney Disease (CKD), has however emerged in recent years (14–22).

This paradoxical, even violent interplay between life and death that a woman experiences in her body in a dramatically crosscutting fashion emerges from these writings. Tyer-Viola et al. (16) emphasize, along with others, that at the same time, there is an attempt to forget the illness by trying to experience a "normal" pregnancy like other women, with a parallel increase in anxiety when pregnancy may modify the symptoms of the illness. There is a clear need to manage potential complications during pregnancy, and it is essential to reflect on and conduct research into improving their management (20, 23). Robin summarizes matters in the psychoanalytical field when stating that during the course of th,e illness, the "threshold of permeability to the unconscious as also the preconscious is lowered; a lifting of repression takes place, which invasively gives way to regressive reminiscences and fantasies" [(24), p. 164].

Both periods are conducive to psychological reorganization during which the most archaic experiences are heavily drawn on. Thus, the psychological processes at work during pregnancy and becoming a mother intersect or intermingle with those set in motion after the announcement and treatment of a serious somatic illness. In this case, the maternal psyche is assailed by traumatic aspects. Anxiety and diffuse anguish are also perceptible. Close family and relatives are also seriously affected.

Pregnancy in women with chronic disease, and more particularly kidney failure, is still rare, especially when the condition deteriorates and the patients are on hemodialysis.[1] The evolution of pregnancy can then become unfavorable due to the frequency of complications. The clinical and physiological disturbances observed an increase in the degree of kidney failure. With the improvement of hemodialysis techniques, the fertility of women suffering from chronic renal deficiency has improved considerably (particularly since the introduction of erythropoietin, which allows more women to have normal cycles). Although increasing successes are now being reported, the outcome of pregnancy for women undergoing dialysis remains extremely uncertain and these women have to deal with massive restrictions. "Intensification of hemodialysis, with up to five or six sessions a week (as opposed to the usual three weekly sessions), peritoneal dialysis, or even the temporary combination of these two methods, is essential for sufficient quality of the amniotic fluid" [(25) p. 121]. Any pregnancy in a woman with kidney disease should be considered as being at risk. During monitoring sessions, the full reality of bodily functions is omnipresent. Psychologically, it should also be borne in mind that end-stage renal disease involves a possibly traumatic confrontation with invasive techniques without which the patient can no longer survive. She cannot preserve herself (26) and each dialysis conjures up an end-of-life situation (27, 28). Confronted by all this, the patient's imaginary projections are crushed, subdued under an operative way of thinking, where discourse is usually merely factual and reverie is well-nigh impossible.

2. Materials and methods

The methodology adopted here is qualitative. The clinical work discussed here stems from analytically oriented psychotherapeutic follow-ups with a psychoanalytical orientation in medical services, both in obstetrics and gynecology (one case study), and nephrology (eleven studies). The sessions took place weekly for periods ranging from 1 to 6 years.[2] The first above-mentioned case study took place with a pregnant woman who had been on dialysis for about a year. The follow-up continued a few months after the birth of the child. For the eleven other follow-ups, kidney disease was known and was not yet chronic before pregnancy but became so afterward. The start of dialysis (in-centre hemodialysis for each of them) began in the year following the birth of the child or a few years later (at most two). For four of these women, psychotherapeutic follow-up started before their pregnancy (and hence the start of dialysis) and for the other seven, it started when they were put on dialysis, that is after the birth of their child.

All this took place against a background of encounters with other women we remember, who were unable to have a child and were dealing with the work of renunciation. Issues related to femininity, maternity, and fertility arose very regularly during psychotherapeutic interviews. Clearly, the disease also heavily affected family members, especially the father of the child. However, the focus here is mainly on the women themselves, who also talked about their spouses, and it is their perspective and their own representations that will be discussed here. Participants' names were anonymized. Written informed consent was obtained from all the participants for the publication of any potentially identifiable data included in this article.

After an in-depth case study, a cross-sectional survey was conducted to identify the main lines of thought. These are presented below.

3. Clinical results

3.1. A matrix experienced as "unwelcoming"

Listening to pregnant women, it is common to hear them talk about what they feel within their bodies, describing how the baby moves and kicks out. Sensoriality is omnipresent. Pregnant women suffering from nephrological disorders who consulted soon confronted the author with descriptions of the inside, functional workings of their maternal bodies. This was accompanied by a plethora of anatomical detail on the disease's progression as it hollowed out an unwelcoming and deficient environment for the unborn child. Additional medical information painted an even bleaker picture when the dialysis went a little less well than usual, adversely affecting the quality of the amniotic fluid. They, thus, considered their bodies as places where "everything (was) bad," "ruined bodies" that were no longer good for anything, entirely negative. An alternative interpretation was to split the body up into its good parts (uterus) and bad parts (the kidneys that failed to function and disrupt the rest, including the uterus).

As they prepared to bring their infant out into the world, they questioned their ability to be "good enough" mothers [in Winnicott's sense (29)] and were vulnerable to depression as they considered themselves to be "unwelcoming" to their child. For some, these two temporalities, pregnancy and illness, proved impossible to articulate and elaborate on, with the thought processes relating to one movement blocking off those on the other. They thought of themselves as a "bad" container, a "bad nurse" right from the time of pregnancy as their renal function deteriorated. The process of psychological integration of the unborn baby, which S. Missonnier calls "psychic nesting" (30), was undermined. Feeling physically deficient, their very ability to carry the child psychically became threatened (31). Their imaginings and their representations of the unborn child became extremely tentative and sketchy as they went through pregnancy not knowing whether it would be carried through

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[1] Hemodialysis usually involves the patient being dialyzed three times a week, for four-hour sessions. They are then connected to an artificial kidney that cleans the blood week after week. Kidney transplantation provides an alternative in some cases.

[2] These follow-ups are currently completed and were collected over a period of about 10 years during the author's practice as a clinical psychologist.

to term, whether the baby might suffer fetal pain, or whether their illness would develop further.

3.2. A skin rendered transparent

Pregnancy entails a dimension of going to see, of taking a look inside (32), of showing this unborn child and at the same time of keeping it secret, of preserving it from view. Let us recall that for Winnicott in *Human Nature*, "true female genital functioning tends to be hidden, when it is not in fact secret (...) the fantasy plays with the concealment within oneself, the hidden and the secret." [(33) p. 65]. Genital representations find their fullest experience in identification with the mother, who is capable of acting and conceiving a child. In young girls' games, we observe this same tendency where the dimension of secrecy is essential. "The game of *Can you keep a secret?* belongs typically to the female side of human nature, just as fighting and pushing things into holes belong to the male side. Unless a girl can keep a secret she cannot become pregnant. Unless a boy can fight or push a train through a tunnel he cannot deliberately impregnate" (1988, p. 65-66). A glimpse of the imaginative elaboration of their dominant bodily functioning can, thus, be seen in such games. That secrecy is an intimate space that one seeks to protect against the outside world but that also functions as a barrier to archaic and oedipal anxieties. Through, thus, defining the notion of secrecy, we also grasp the pleasure we can feel in keeping a secret and then sometimes sharing it. Working on this theme with pregnant women may help them develop a capacity to turn toward their internal mode.

This inward drive is further encouraged by the technological advances in gynecology and obstetrics (ultrasound, monitoring, etc.). During their interviews with the author, pregnant women with an underlying or declared CKD all described their bodies as being pried into under everyone's gaze. This increased scrutiny that is lavished on them and their baby reassured them in part, but it also had a price tag attached to it psychologically as they felt as though they were being made transparent, with their insides constantly being probed by machines and needles. They spoke of a body that was no longer merely attacked internally by the disease but also assailed at its very limits. This accumulation of micro-traumas compounded their fears daily. Patients in nephrology regularly evoked this theme, stressing how burdensome and painful this was at a time when they ought to be providing a containing envelope for their baby. This containing function was regularly put to the test. In order to cope with this, the work of primary erogenic masochism[3] that intertwines the life drive and the death drive is essential. I shall give as an example the case of Léonie.

> *We met Léonie for the first time when she was hospitalized for 3 weeks. She had to stay in bed throughout her pregnancy and even a little longer as she was to undergo another operation as soon as the baby was born. She was 5 months pregnant. She had kidney disease (due to diabetes) but also cardiac and digestive problems. She was connected to multiple catheters and an operation was scheduled after childbirth to restore her digestion. The seriousness of her medical complications weighed so heavily on the author that she felt trapped, unable to look beyond that immediate reality of her bedridden condition. Léonie explained that she cared about this baby above all else and that she had waited such a long time for it to come. She and her partner had wanted a child for several years now and the waiting became all the more painful as her additional somatic problems emerged, suddenly, on top of her already recognized heart problems. She was ready "to put up with anything, to endure everything, to wait week after week as long as (she could) keep (her) child." To begin with, the couple's desire to have a child emerged slowly and quietly, with the idea of starting a family a little later when the time came... but she now saw a major difference, talking about a pregnancy and a child that were "vital" to her. It was "what gave (her) the strength to live," as she put it. Having learned of her illness, she knew that it was going to be chronic for the rest of her life but she then wanted this child more than anything else to "prove to (herself), to show that (she could) also give life." Her body "cannot let (her) down at all levels," it must also be able to "give life." Her comments as to her inner bodily workings were harsh and brutal, with only the womb being preserved as a bubble of life. She was a carrier of death, but she would also be a giver of life... She talked of this pregnancy, this child, as coming to repair her psychically as well as physically, her body in itself. Some days, she had real death anxieties, feeling the peripheral parts of her body become numb (peripheral neuropathy being a common symptom in the later stages of diabetes). Even though she had not yet reached that condition, she was acutely aware of such complications and even evoked the sensation of herself gradually being transformed into a corpse. As the unborn infant moved inside her, knocking, this brought her out of these moments of morbid stupor. She also looked forward to examinations of the baby, where the gynecologists would tell her that he was doing well. Admittedly, he was rather small and a Cesarean section was already being envisaged before the end of pregnancy, but he was developing quite normally. Léonie kept repeating the words "completely normal" to herself. Even dependent as she was on medicine and treatment, she was going to give birth to a child in good health. She was able to carry him and ensure that he developed normally. Despite moments of doubt and worry, she felt like a good enough mother (according to Winnicott's meaning).*

Inevitably, the scopic impulse is of course exacerbated by the reinforced medical monitoring needed to ensure that pregnancy proceeds well. Each medical indicator is watched assiduously. This is psychologically demanding but also reassures the mother-to-be when the baby's and her own medical results are good. Technology and medication are indispensable to maintaining life.

3.3. The dream baby[4]

This hollow female organ, the uterus, is at the center of these women's concerns and calls upon them in particular at the phantasmic level. In reference to Klein, the mother's body is

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[3] Which we can define as the first psychic connection realizing an entanglement between the life drive and the death drive necessary for psychic life. This makes it possible to withstand the tensions of excitation that are inevitable and necessary for psychic life [according to Rosenberg (12)].

[4] Aulagnier (34); Lebovici (35); Soulé (31); Cupa-Pérard (36); Nerson-Sachs (37).

fantasized as a recipient of combined wealth, namely including the father's penis and new babies to come. The infant in the fantasy is an internal object among others such as the breast, penis, feces, and child, all related to the receptacle containing all, the "good" as well as the "bad," that is to say, the maternal interior. This recalls the box that contains all secrets, as M. Cournut-Janin has already pointed out: "Between Dora's jewelry box and the secret that Winnicott's little girl knows how to keep, the metaphor of a box and what it conceals is constructed: the female body, which is rich in 'transposable' content (penis, feces, child, jewelry, buttons, not forgetting the blood, which is promising, of the menstrual period)" [(38), p. 57]. The interior of the female body can also be full of other types of objects, which can be subsumed under the category of waste. At a fantastical level, the treasured baby or the jewel baby is evoked, but the womb may also be seen as a receptacle for non-productive discharge or even poison. In women with CKD, these phantasmic babies are present as in many women, but the feeling is all the more acute and the divisiveness all the more pronounced.

The baby as treasure is there to fill out, restore, and enhance child-like desires. In nephrology, this is especially true, as the pregnant patients feel a sense of fulfillment, a kind of narcissistic elation: being pregnant for them is a true victory. The infant is a treasure, a movement of life. Even as the disease emerges or asserts itself, it becomes eclipsed by the marvelous mundanity of the life of a woman who goes on her way just like any other, meeting a man and then becoming pregnant. These women spoke of real "healing." For women with kidney disease, the baby also comes in to mend them, to show them that their bodies are not just sick and fantasized as "bad" or "damaged" but able to bring forth life. They, thus, prove to be fertile when few thought it was possible. This baby that normalizes her life can become a saving grace (and hence is largely idealized), an ornament to the mother-to-be, while the baby too can be adorned like their mother, reinforcing their femininity phantasmically.

However, the fetus is also potentially dangerous to the mother and can encroach on or invade her maternal body. For women with CKD, pregnancy can actually degrade their kidney functions. The baby can, thus, be associated with deteriorating maternal health generating a form of ambivalence that can sometimes take on a persecutory aspect.

These women tend to think that their inside is not being appropriately fitted to receive the unborn child in optimal conditions. The liquid environment is imperfectly purified, not adequately renewed, thus reactivating fantasies of a soiled, damaged, cloacal matrix. The anal dimension is largely present. The baby can then be fantasized as being attacked by this "waste" or even being part of it. The idea of a form of "invasion" by the disease is regularly mentioned.

3.4. The "motherhood constellation"[5]

Again referring to the approach adopted by Melanie Klein, the primordial anguish of the girl, the woman, concerning her inner body relates to the fear of seeing herself ravished, damaged within her body, above all through her genital organs. The mother's body is the place where all that is desirable is located, the place for investigations of origins and the very mainspring of life. As a result, the child, especially the daughter, is full of hatred toward her mother, whom she wishes to attack in order to steal what she hides inside her body. This is followed by anguish linked to the fear of reprisals and of seeing the inside of her body stolen away and destroyed (40). Her own ability to bear children is destined to play a similar role, to prove to her the unaltered nature of her inner self, for which healthy, vigorous offspring will provide proof. Conversely, any flaw in the child's successful development will revive the mother's anxieties relating to her invisible, unverifiable, and worrying inner body and the ensuing reprisals she fears from her mother. It is also the fear of having one's internal or intimate treasures (penis, feces, or fetus) and the gem baby taken away, especially by the mother with whom the daughter had tried to rival and who may now want to take back what belongs only to her, especially the father's child. Thus, some women suffering from kidney failure spoke about their fear of a broken filiation and how they felt it was for them impossible to become a mother. Many childhood desires were re-awakened, and especially their past "cravings" for the maternal body. Their difficulties surrounding their own fertility and pregnancy came as an echo, a form of punishment for their infantile sexual desires.

Confrontation with their own mothers for these women can be painful. Ambivalence and aggression are regularly palpable and may be accompanied by a feeling of guilt. This is exacerbated when the kidney disease is hereditary.

Moreover, when faced with this debt of life (41–43) toward their mother who brought them into the world, the infant may seem to provide a way to settle matters. However, the question remains as to whether the patient will be able to carry things through to childbirth, after which there may still remain a complex debt that cannot be removed. We need to remember that these women talk of the life they are bearing within them but also of their possible early death. Well cared for, that outcome may still be remote, but the disease remains naggingly present, accompanied by the fear of their imminent demise. These women readily resort to mentioning their mother (sometimes a sister) as the person who will take care of their child if 1 day they are no longer able to do so. They also look to the coming months when they will have to organize their lives as mothers and fit all this in with their dialysis, already seeing themselves as mothers in control but only tenuously as they rely on treatment several times a week before having to recover. Their mother's presence by their side provides reassurance but also holds them within a form of dependence. They are to become mothers but regularly remain as if maintained in an infantile position, being dependent on the medical team and the machine itself in order to survive and give birth:

> *Katia is the mother of a 2-year-old girl. She says she is lucky because she was able to have her child before dialysis. She is severely diabetic and pregnancy did not help but at least she has her child by her side. She hears her laughing, she plays with her and she talks about her "ray of sunshine." She doesn't think about the future, about her failing eyesight and the treatment that is "eating out her life." All she thinks about is her daughter. After a pause in the interview, she explains that the thrice weekly trips to undergo dialysis are becoming more and more painful for her as she leaves her daughter behind. She knows her child is in good hands with her mother, but the whole thing is becoming more and more unbearable. The situation is not just a "separation, like any woman going to work, but something else mixed in." Katia feels unable to care for her daughter, unable to run toward her or carry her anymore as she devotes attention to her arm where the fistula*

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[5] Stern (39).

> *that allows her to be connected to the dialysis machine is located. She feels she is failing when off work for health reasons. Sometimes she is so tired that she finally entrusts her daughter to her mother so that she can sleep and recover. She is a mother but only "part-time." She gets depressed. She wanted so much to be a mother and in the end her own mother takes more care of her daughter than she does. She feels guilty about her rivalry and aggressiveness toward her mother, which depresses her even more. Neither does she live up to her mother's standards. She talks about her childhood and what her mother did with her and for her, which she is not sure she can do with her daughter. She finds ways around the difficulties. Some days she feels creative enough and other days she thinks that she will never be able to give her daughter what she has received. She talks about a half-hearted transmission.*

4. Discussion: Clinical treatment shrouded in uncertainty

The major challenge within our health institutions is to sustain multi-professionalism in hospitals in order to understand and cater to the patient as a whole. Due to cross-analysis conducted by different professionals, care is thought out and shared by several people with different skills. A kind of "constellation" of health and care professionals is essential here.

The recent pandemic has stimulated new thinking on health issues and innovative concepts such as "one health" are emerging. A recent French ministerial report published in February 2022 promotes this philosophy to reflect on the "lessons of the crisis." The World Health Organization defines health as "a state of complete physical, mental and social wellbeing and not merely the absence of disease or infirmity"[6] The patient's health is, thus, considered in all its complexity, in relation to those close to her and the world around her. From this perspective, psychologists have a duty to be attentive to the analysis of institutional dynamics. We actively participate in the reflections of the medical and care teams at various institutional levels involving the hospitalization service staff at multidisciplinary meetings (doctors, nurses, social workers, dieticians) during which the teams formulate new demands in our direction. These meetings are also an opportunity to discuss ongoing follow-up. Interstitial times are also important, with the psychologist meeting caregivers and physicians. The psychologists then devote time to passing on information to the medical and care teams.

As psychologists and psychotherapists working in these medical services, we are confronted with a "clinic of the real" (44), where the experiences of trauma, mourning, and loss intersect. In these psychosomatic services, the harsh realities of clinical work can severely affect us. Faced with the burden of bodily representations, we try to think with the patient, in the dynamics of exchanges, of the place the spoken word can occupy, enabling the emergence of representations of a body closer to the fantasy, drawing away from the clutches of immediacy. In the medical world, the real daily challenge is to maintain the stability of the psychological framework (in the broadest sense of the term) offered to our patients, considering the sick person in her somatic and psychic wholeness, in connection with her entourage and environment. To avoid becoming overwhelmed by the grim realities of the sickness, we need as psychologists, to find our own temporality and retain our own capacity for reverie (in the sense of Bion) with regard to our patients.

Clearly, patients suffering from chronic diseases where their very survival is at stake experience considerable distress. The process of dialysis itself is a potent reminder of mortality and proximity to death becomes deafening as first object relations move into the foreground, shrouded in uncertainty. Psychic disorganization can be massive. We regularly observe patients who are distraught and unable to find the internal resources to cope with the experience. Here, the psychologist has a duty to listen to archaic traces as the body, in dialysis, appears in all its reality and vulnerability. As practitioners, we regularly have to listen to speech devoid of prosody, lacking modulation and affects, and having to remain attentive to the unspeakable. Gradually, in the space of psychotherapeutic work, possibilities for the "reanimation" of the psyche emerge, as part of an internal space that takes up a phantasmic activity as it begins to be able to project into the external space and the space of the sessions.

Psychological or psychotherapeutic follow-up provides a reflective appraisal so as gradually to recover the pleasure of thinking and allow for the reintegration of possible traumas. During the patient's discourse, time regularly freezes over. It remains somehow suspended, sometimes in a lasting way. Faced with such discontinuity, the patient needs to find regularity in the relationship and consistency in the medical and nursing teams, as well as in our own psychotherapeutic framework. This therapeutic structure needs constancy for the patient to emerge from the situation of dependence and reappropriate experience as a "continuity of being" (as understood by Winnicott).

The arrival of the baby is marked by serious medical constraints and by the limitations imposed by the disease. In the course of psychotherapeutic follow-up, we gradually try to leave more room for reverie. The patient can then take the risk of investing more calmly in the baby, her baby, as pregnancy progresses and she feels it move within her. The challenge is to differentiate and specify the sensations specific to her pregnant body and the fetus and, thus, rely on them so that the representations surrounding and enveloping this unborn baby can develop and become richer and greater in number.

These women may sometimes wish to commence psychotherapeutic work to begin the process of mourning for their fertility and the child. The work of renunciation is accomplished gradually and transmission will take place elsewhere, moving toward other objects and other projects.

5. Conclusion

When the child is born and terminal kidney disease occurs, it may be important to "de-crystallize" this period as the different psychic movements may possibly collide and become paralyzed. Pregnancy, the birth of the child, and sometimes the child itself are all part of the history of the illness. Demands on the child, its life debt, can be embroiled in what is happening with the illness. The child is caught up in the same loop as the machine (the artificial kidney that drains the patient three times a week), making them live like the machine.

However, even when carried along with the illness, pregnancy remains a moment of life where the work of creation, both physiological and psychological, is at work. In particular, it affords an opportunity to pass on the message that beyond the disease, life goes on.

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[6] https://solidarites-sante.gouv.fr/IMG/pdf/contribution_conseil_scientifique_8_fevrier_2022_one_health.pdf

A natural follow-up to this work would be the study of early interactions between mothers on dialysis or about to be on dialysis and their children. This is a surprisingly understudied area that would be important to develop.

Data availability statement

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author.

Ethics statement

Ethical review and approval was not required for the study on human participants in accordance with the local legislation and institutional requirements. The patients/participants provided their written informed consent to participate in this study. Written informed consent was obtained from all the participants for the publication of any potentially identifiable data included in this article.

Author contributions

The author confirms being the sole contributor of this work and has approved it for publication.

Conflict of interest

The author declares that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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Dimensional model on how familial vulnerability and environmental factors impact transitional age youth psychopathology: The *Transition_psy* study

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Background: Understanding psychopathology in transitional age youth (TAY) requires a complex model, incorporating familial vulnerability and environmental factors. A trans-diagnostic and dimensional approach seems the most appropriate. *Transition_psy* study aims to assess factors playing a role in TAY psychopathology and to define predictors.

Materials and methods: This article presents part of the *Transition_psy* study results, a case-control observational study. Youth aged 17 years old were recruited between June 2020 and December 2021, from both clinical [clinical population (CP) group] and non-clinical settings [non-clinical population (NCP) group]. Participants completed self-report questionnaires. The primary outcome to assess TAY psychopathology was the Youth-Self Report (YSR). We evaluated care needs with the Health of The Nation Outcome Scales For Children And Adolescents (HoNOSCA-SR) and quality of life with the World Health Organization Quality of Life – BREF (WHOQoL-BREF). Exposure factors included familial vulnerability, childhood, and present environmental factors, such as first-degree family history of psychopathology, the Childhood Trauma Questionnaire (CTQ) and the Family Assessment Device (FAD). YSR scores were compared, between groups, according to exposure factors with ANOVA and linear regression. We performed best subsets selection of multivariable analyses based on the Akaike Information Criterion. This study was registered with ClinicalTrials.gov (Identifier: NCT04333797).

Results: A total of 220 TAY (CP = 106, NCP = 114) were included in the study. Participants were aged 17 years old. The majority were female (69.1%), single (96.8%), and born in Belgium (82.3%). Clinical data were all significantly different between CP and NCP groups. YSR scores were found statistically different according to group ($p < 0.001$), first-degree family history of psychopathology

($p < 0.001$), CTQ ($p < 0.001$), and FAD ($p < 0.001$). Predictive dimensional model suggested that TAY psychopathology can be predicted by group, CTQ and FAD. Significant positive correlation was found between YSR and HoNOSCA (rho = 0.81) and negative correlation between YSR and physical and psychological health (rho = −0.69 and −0.71, respectively).

Conclusion: This study findings allowed to present a predictive dimensional model on TAY psychopathology, including belonging to a clinical population at transitional age, childhood trauma, and family dysfunction. Further research is needed to replicate *Transition_psy* study results in other samples. The proposed model could be used in clinical practice to improve assessment of TAY psychopathology.

KEYWORDS

transitional age youth, psychopathology, dimensional approach, familial vulnerability, environmental factors, quality of life, care needs

1. Introduction

Transitional age youth (TAY), aged 16-24 years old, are a particularly at-risk population regarding mental health. Mental disorders onset before the age of 25 in 62.5% of the cases, with a peak incidence around 14.5 years old (1).

The ongoing hypothesis explaining the incidence of psychiatric disorders in TAY is multifactorial. Genetic and environmental factors, partially through epigenetics, negatively impact brain development (2). The brain maturation process presents two particularly vulnerable periods in life: (1) perinatal and early childhood, and (2) adolescence and early adulthood. During these periods, there is a greater risk that environmental factors interfere with brain maturation (2, 3).

It has already been proven that both familial vulnerability and childhood adverse events increase the risk of psychopathology (4, 5), manifested through internalizing and externalizing behaviors (6, 7). Additionally, psychopathology appears to be related to adolescent environmental factors, such as gonadal hormones, substance use, social interactions and school environment (8). It is still unclear whether these factors are a result of alterations that occurred earlier in life or strictly related to puberty (9).

All these factors contribute to the scientific and clinical complexity of TAY psychopathology and care needs. Understanding typical developmental processes remains crucial in research on the prevention of adverse life events effects on mental health (10). There is growing evidence in the literature that this complexity could be resumed in a single dimension of psychopathology, the "p" factor, measuring each individual's liability to mental disorder, comorbidities, duration and severity of disorders. The "p" factor seems positively correlated with family history of psychiatric illness, brain function, childhood developmental history, and adult life impairment (11).

Hence, a trans-diagnostic dimensional approach seems to better allow the understanding of TAY psychopathology (12, 13). TAY psychopathology is often characterized by early clinical presentations that include non-specific or subthreshold intensity and/or frequency symptoms, and by a high incidence of comorbid disorders (14). In recent years, such trans-diagnostic clinical staging models have gained importance, by allowing a multidimensional assessment while considering illness as a dynamic continuum from its absence to its most extreme expression (13). This broader strategy to identify at-risk TAY may ultimately permit to recognize early stages of severe mental disorders, offering new management strategies tailored to the patient's clinical stage, preventing the onset and/or progression of mental disorders (12). However, to date, there is little data on clinical dimensional characteristics involved in the development of psychopathology at the age of the transition and the identification of at-risk TAY.

The *Transition_psy* study focuses on the understanding of TAY psychopathology mainly in terms of quality of life and care needs (15). Based on the existing literature, we hypothesized that childhood trauma and family history of psychopathology were the main common factors to develop psychopathology at the transitional age (4, 5, 11). The *Transition_psy* study proposes a predictive dimensional model considering familial vulnerability, and both childhood and present environmental factors. The conceptual model of the study is presented in **Figure 1**. The aim of *Transition_psy* is to determine which factors play a protective or a risk role in psychopathology in the transitional age.

2. Materials and methods

2.1. Study design and setting

This article presents part of the *Transition_psy* study results. This is a case-control observational study evaluating risk and protective factors to develop psychopathology in TAY. The recruitment has been led in clinical and non-clinical settings. Clinical settings consisted in both outpatient and inpatient facilities within the urban area of Brussels: three general university hospitals and one outpatient university clinic. Non-clinical settings were both schools in the urban area of the same town and social networks. The complete protocol of *Transition_psy* study was

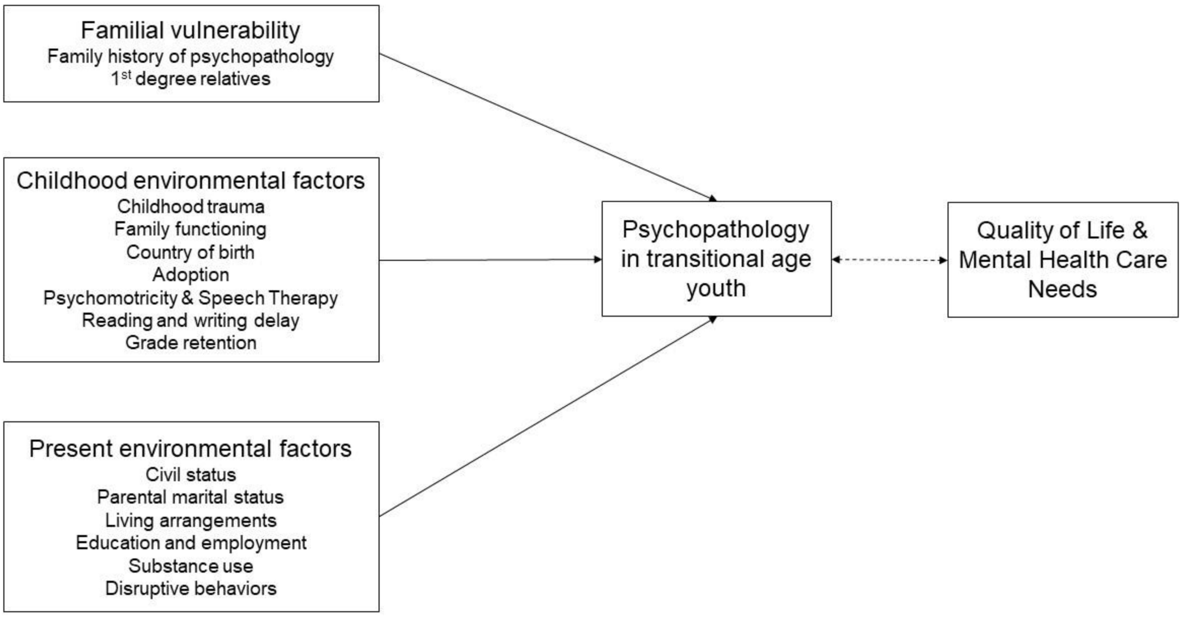

FIGURE 1
Conceptual model for the study. The single headed and continuous arrows indicate a predictive relation; the double headed and dotted arrow indicates a correlation.

described in a previous paper (15). This trial was registered with ClinicalTrials.gov (Identifier: NCT04333797) on 3 April 2020.

2.2. Recruitment and procedure

Between June 2020 and December 2021, 17 years old youth were recruited in the Brussels urban area, Belgium. Clinical sample was recruited in collaboration with the referring physician. Participants from non-clinical settings were invited to participate in the study throughout flyers and social media posts. As a compensation, they received a 20 € voucher from a multimedia shop.

To be included in the study, participants had to have sufficient fluency in French, and both parents or legal holders of parental authority and the participant had to provide informed and written consent. We excluded potential participants actively involved in another research study, those who were unable to answer the assessment tools, and patients with a progressive illness affecting short-term vital prognosis.

The clinical group was named “clinical population” (CP) because these participants were actively involved in outpatient or inpatient care, at the moment of inclusion. On the contrary, the non-clinical population was named “non-clinical population” (NCP).

At the inclusion, participants met the research assistants for a brief interview and were invited to complete the baseline assessment, consisting of self-report questionnaires available on the Research Electronic Data Capture (REDCap) platform. The time for completion was about 45 min.

Of the 428 participants considered for the study participation (CP = 309, NCP = 119), 393 were eligible to participate, among whom 254 consented to enroll in the baseline assessment. Few participants ($n = 34$) did not complete at least 50% of the assessment and they were not included in the data analysis, representing a final sample of 220 (CP = 106, NCP = 114) with a participation rate of 51.4% (see **Figure 2** for more details).

2.3. Measures and outcomes

All the measures are self-reported standardized questionnaires, previously validated in French.

The primary outcome to assess psychopathology was the Youth Self-Report 11–18 (YSR), a specific instrument for 11–18 years old adolescents performing a detailed assessment of participants’ psychopathology (16, 17). This is a 112-item questionnaire on a three-point Likert scale (0–2). Global internalizing and externalizing problem behavior scores can be obtained. For this study analysis, we used the total scores to describe the overall TAY psychopathology (18). We also used the General Health Questionnaire-12 (GHQ) (19, 20), the 12-item version, as a screening tool for psychopathology in the sample. Each GHQ item scored on a 4-point scale (0–3); total scores range from 0 to 36 with higher scores representing greater psychopathology. This tool has been proven effective in primary care settings (21, 22).

We used two measures to evaluate care needs and quality of life, respectively: the self-rated Health of The Nation Outcome Scales For Children And Adolescents (HoNOSCA) (23, 24) and the World Health Organization Quality of Life- BREF (WHOQoL) (25, 26). The HoNOSCA (23, 24) is a 13-item instrument, scored on a five-point scale (0–4), measuring the severity of physical, personal, and social problems of children and young people with mental health problems. The total score, ranging from 0 to 52, represents the overall severity of care needs. The WHOQoL (25, 26) is a 26-item questionnaire on a five-point scale (1–5). It is possible to calculate

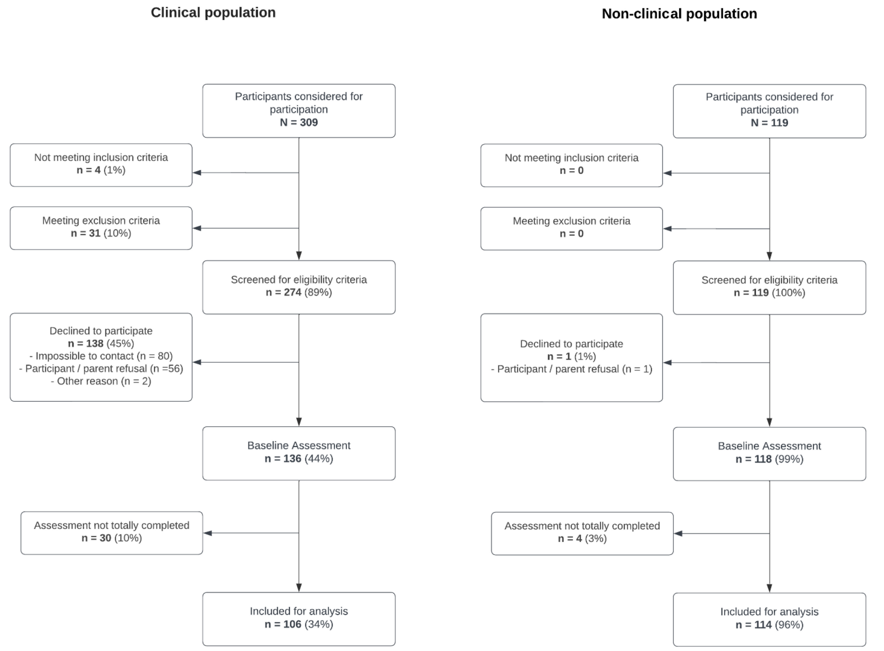

FIGURE 2
Transition_psy study recruitment flowchart.

four separate scores for each broad domain ranging from 0 to 100. The four domains of quality of life are (1) physical health, (2) psychological health, (3) social relationships, and (4) environment. These two measures were used to examine the correlation between the TAY psychopathology and their actual degree of care need and impact on quality of life.

We collected sociodemographic data, such as: sex, civil status, country of birth, parental marital status, living arrangements, enrollment in education and/or employment. Additionally, participants were asked to report clinical data, mainly related to mental health: psychiatric and/or psychological consultations, psychiatric inpatient care and psychotropic drugs use.

Exposures have been assigned to three main categories according to chronological criteria: (1) familial vulnerability, (2) childhood environmental factors, and (3) present environmental factors.

To evaluate familial vulnerability, participants were asked to provide data about mental illness history of their first-degree relatives (parents). Summarized scores could range from 0 to 2, if they had no relatives with mental illness history, one or both parents, respectively.

Childhood environmental factors were assessed using two standardized tools: the Childhood Trauma Questionnaire (CTQ) and the Family Assessment Device (FAD), together with data about history of migration, adoption, previous psychomotricity or speech therapy, grade retention at school and reading or writing delay. Concerning these two last items, the threshold age for delay was set at 8 years old (27, 28). The CTQ is a 28-item instrument measuring trauma during childhood (29, 30). Each item scores on a five-point Likert scale (1–5) and the CTQ total score ranges from 28 to 140 (31). The higher scores represent greater intensity of childhood trauma. The FAD is a 12-item tool on a four-point scale (1–4) (32, 33), with a total score from 12 to 48 (34). Higher scores represent worse levels of family functioning.

The present environmental factors considered in the study were: some of the sociodemographic variables (youth's civil status, parental marital status, living arrangements, enrollment in education and/or employment), substance use (alcohol, tobacco, cannabis, or other drugs), and disruptive behaviors (such as stealing, vandalism, animal cruelty, assault, and battery).

2.4. Statistical analysis

We performed the "*a priori*" analysis to calculate the sample size with the G*Power software, version 3.1.9.7 (35). To reach a medium effect size (i.e., $r = 0.3$), with an α-error of 5% (two-sided) and a β-error of 80%, the required total sample size was 84 participants for correlations and 144 for linear regressions.

All statistical analyses were conducted using the software Statistical Package for the Social Sciences Version 27 (SPSS, Inc., Chicago, IL, USA).

We performed descriptive statistics to study sociodemographic and clinical characteristics in the total sample and in the two groups (CP and NCP): absolute and relative frequencies were presented for all qualitative variables; means with standard deviations were used to describe normally distributed quantitative variables. The normality of the distributions was established graphically (histogram, and normal probability plot). The frequencies in categories were compared between groups with χ^2 test for the majority of variables; when the absolute count was less than five, we performed Fisher's exact test. The homogeneity of variances was verified with Levene's test, and the means of quantitative variables were compared between the two groups with independent samples T-test.

To compare the means of quantitative variables (GHQ and YSR) according to familial vulnerability and environmental factors between the sub-groups, we performed ANOVA for each categorical variable and linear regression for the quantitative variables (CTQ and FAD), presenting regression coefficients (b) and 95% confidence intervals. The interaction of each variable with the group was assessed in the models. For significant variables, we then performed multivariable analyses and tested all combinations of explanatory variables to choose the best model based on the Akaike information criterion (AIC). We verified the normality and homogeneity of variances of residuals with graphical representations. When appropriate, the p-values of post-hoc pairwise comparison tests were adjusted with Tukey's honestly significant difference method.

Correlation between WHO-QoL, HoNOSCA, YSR, and GHQ was calculated with Spearman's rho coefficient.

Missing data were treated with pairwise deletion. The statistical significance level was set at 0.05 (two-sided).

3. Results

3.1. Sociodemographic and clinical characteristics

A total of 220 participants, aged 17 years old, were included in the final sample. The majority of participants were female (69.1%), single (96.8%), and born in Belgium (82.3%), but there were no significant statistical differences between groups regarding sex ($p = 0.334$), civil status ($p = 0.714$), or country of birth ($p = 0.669$). There were significant statistical differences regarding parental marital status ($p = 0.008$), living arrangements ($p = 0.009$), and enrollment in education and/or employment ($p = 0.044$) between CP and NCP participants. Detailed sociodemographic characteristics of participants are presented in Table 1.

Table 2 shows clinical data in the total sample and between groups. CP participants had significantly greater rates of psychiatric and/or psychological consultation ($p < 0.001$), inpatient care ($p < 0.001$), and psychotropic drug use ($p < 0.001$), comparing to NCP participants. CP participants had significantly higher GHQ ($p < 0.001$) and YSR ($p < 0.001$) scores than NCP participants. CP participants also presented higher HoNOSCA scores ($p < 0.001$) and lower WHOQoL scores, in all four domains ($p < 0.001$).

3.2. Familial vulnerability and environmental factors

We decided to present the analysis of YSR scores according to the group and familial vulnerability, childhood and present environmental factors as primary results (Table 3). Findings regarding GHQ scores according to the most significant factors are summarized in Table 4. The analyses of every factor using the GHQ scores are presented in the annex section (Supplementary Table 1).

Youth Self-Report scores were statistically different according to the group (CP and NCP), in each analyzed factors. The most significant factors to determine differences in YSR scores were the first-degree family history for psychopathology ($p = 0.022$), CTQ scores ($p < 0.001$), FAD scores (<0.001), parental marital status ($p = 0.027$), and disruptive behaviors ($p = 0.014$). Results on the interaction between the group and each factor did not show a significant difference, except for CTQ scores (Table 3). Regarding first-degree family history for psychopathology, post-hoc pairwise comparison tests showed a significant difference between participants without family history and those with one or both parents with a mental disorder ($p < 0.001$ and $p = 0.004$ accordingly), but no statistical difference between these last two categories ($p = 0.998$).

We found similar findings on GHQ scores in both group and factors. The most significant factors to determine differences in GHQ scores were the first-degree family history for psychopathology ($p = 0.028$), CTQ scores ($p < 0.001$), FAD scores (<0.001), and other drugs use ($p = 0.049$) (Table 4).

3.3. Predictive dimensional models

We studied the most predictive models on YSR scores with selection of the most significant factors identified in the previous logistic regressions. The two models with the smallest AIC were selected. *Model 1* included the following variables: group, CTQ, FAD, and first-degree family history of psychopathology (AIC = 790.13). In *model 2*, the variable first-degree family history of psychopathology was excluded (AIC = 790.17). The significance of each predictor in the predictive dimensional models is shown in Table 5.

Finding about the most predictive two models for GHQ scores are presented in the annex section (Supplementary Table 2). Group, FAD, and first-degree family history of psychopathology are included in both models, whereas CTQ is only present in the second best model.

3.4. Quality of life and care needs

The correlation matrix (Figure 3) assesses the strength and the direction of the relationship between the four domains of WHOQoL, HoNOSCA, GHQ, and YSR. Each correlation coefficient was statistically significant ($p < 0.05$). The higher positive correlation coefficient (rho = 0.81) was found in the relationship between YSR and HoNOSCA. GHQ and HoNOSCA are also positively correlated, but with a lower coefficient (rho = 0.66). Similar results were found in GHQ and YSR (rho = 0.61). GHQ, YSR, and HoNOSCA correlate negatively with all four domains of WHOQoL. However, the correlations with the highest coefficients are found between the first two domains of WHOQoL (1 = physical health; 2 = psychological health) and the other measures scores: GHQ (rho = −0.64 and −0.67, respectively), YSR (rho = −0.69 and −0.71, respectively), and HoNOSCA (rho = −0.70 and −0.71, respectively). The four domains of WHOQoL are all positively correlated among them; even if statistically significant, these correlations do not appear very strong. The highest one is the correlation between domains 1 and 2 (rho = 0.71).

TABLE 1 Sociodemographic characteristics of the sample and comparisons of the groups ($N = 220$).

Variables	Total ($n = 220$)		CP ($n = 106$)		NCP ($n = 114$)		
	n	%	*n*	%	*n*	%	*p*-value
Sex							
Male	66	30.0	32	30.2	34	29.8	0.334
Female	152	69.1	72	67.9	80	70.2	
Other	2	0.9	2	1.9	0	0.0	
Civil status							
Single	213	96.8	102	96.2	110	97.4	0.714
Cohabitant	7	3.2	4	3.8	3	2.6	
Country of birth							
Belgium	181	82.3	86	81.1	95	83.3	0.669
Other	39	17.7	20	18.9	19	16.7	
Adoption							
No	214	97.3	103	97.2	111	97.4	1.000
Yes	6	2.7	3	2.8	3	2.6	
Parental marital status							
Married	105	47.7	40	37.7	65	57.0	**0.008****
Divorced/separated	93	42.3	56	52.8	37	32.5	
Other	22	10.0	10	9.4	12	10.5	
Living arrangements							
Family	202	91.8	92	86.8	110	96.5	**0.009****
Other	18	8.2	14	13.2	4	3.5	
Education and/or employment							
Yes	207	94.1	96	90.6	111	97.4	**0.044***
No	13	5.9	10	9.4	3	2.6	

$^{*}p < 0.05$ and $^{**}p < 0.01$. CP, clinical population; NCP, non-clinical population. Bold values are the statistically significant *p*-values.

4. Discussion

This paper presents part of the *Transition_psy* study results. It consists of a case-control observational study aiming to model the mechanisms involved in the TAY psychopathology.

4.1. Sociodemographic and clinical characteristics

Some sociodemographic characteristics were are significantly related to the groups. Our findings show that CP participants have more frequently divorced or separated parents which is consistent with previous literature (36, 37). The differences observed in living arrangement conditions, on the contrary, is possibly related to the recruitment strategy of the *Transition_psy* study, as the clinical facilities collaborated with Youth Aid Residential Services, resulting in greater rates of CP youth not living with their families. Enrollment in education and/or employment is lower in the CP group. According to the Belgian Superior Health Council (38), psychopathology is associated with a higher risk for school drop-out (39), and reduced work activity (40).

The statistical differences in clinical characteristics confirm that CP and NCP groups properly represent the clinical and non-clinical populations that we aimed to target in this study. Both psychopathology scores (GHQ and YSR) are relevant to distinguish between CP and NCP participants; these findings are consistent with previous research (41–43). Concerning quality of life, WHO-QoL scores in the CP population are significantly lower than in a reference healthy population (25). In particular, psychological health scores (Domain 2) in the total sample and CP group are lower than the means in the reference healthy population, whereas the environment scores (Domain 4) are higher in this study sample (25). Milestone European study showed that HoNOSCA is an appropriate instrument to assess the severity of mental health problems in TAY (44), which is consistent with our study sample.

However, part of the NCP participants already had previously had psychiatric and/or psychological consultation (40%), and a small portion of them already have received inpatient care in psychiatric units or psychotropic treatment. If these results could be partially explained by epidemiological data on mental health care needs in the general population (45), we should also highlight three additional factors. First, mental health literacy appears to have improved in youth over the last years (46), resulting in higher psychological or psychiatric consultation rates that might

TABLE 2 Comparison of clinical data by groups (*N* = 220).

Variables	Total (*n* = 220)		CP (*n* = 106)		NCP (*n* = 114)		
	n	%	*n*	%	*n*	%	*p*-value
Psychiatric and/or psychological consultation							
Yes	150	68	104	98.1	46	40.4	$\mathbf{<0.001}^{***}$
No	70	32	2	1.9	68	59.6	
Inpatient care							
Yes	49	22	46	43.4	3	2.6	$\mathbf{<0.001}^{***}$
No	171	78	60	56.6	111	97.4	
Psychotropic drug							
Yes	40	18	39	36.8	1	0.9	$\mathbf{<0.001}^{***}$
No	180	82	67	63.2	113	99.1	
	Mean	SD	Mean	SD	Mean	SD	*p*-value
GHQ	17.05	8.24	20.81	8.13	13.55	6.70	$\mathbf{<0.001}^{***}$
YSR	61.20	10.17	66.52	8.95	56.25	8.64	$\mathbf{<0.001}^{***}$
WHOQoL							
Domain 1	66.26	18.59	56.44	17.91	75.39	14.04	$\mathbf{<0.001}^{***}$
Domain 2	53.44	21.71	40.53	19.30	65.45	16.29	$\mathbf{<0.001}^{***}$
Domain 3	62.60	19.15	57.43	19.84	67.39	17.23	$\mathbf{<0.001}^{***}$
Domain 4	72.74	17.15	67.58	17.97	77.53	14.89	$\mathbf{<0.001}^{***}$
HoNOSCA	17.92	10.21	24.20	9.02	12.09	7.42	$\mathbf{<0.001}^{***}$

$^{***}p < 0.001$. CP, clinical population; GHQ, General Health Questionnaire; HoNOSCA, Health of The Nation Outcome Scales For Children And Adolescents; NCP, non-clinical population; SD, standard deviation; WHO-QoL, World Health Organization Quality of Life (domains: (1) physical health, (2) psychological health, (3) social relationships, and (4) environment); YSR, Youth Self-Report. Bold values are the statistically significant *p*-values.

not be related to a general increase in psychopathology nor in mental health care needs. Second, we acknowledge that data were collected mostly during the COVID-19 pandemic. It has already been proven that mental disorders increased during the pandemic, particularly in youth (47). Last, since NCP participants volunteered to participate in this research, they could be more likely to be concerned by a past or present experience of psychopathology.

Participation rates were quite different according to the two groups (CP and NCP). Firstly, the recruitment procedure was different in CP and NCP groups. Since CP youth were enrolled in the study in collaboration with their clinician, many of them (45%) declined to participate. On the contrary, NCP youth were asked to express their interest to participate, spontaneously, and they received compensation; as consequence, they were less susceptible to decline the participation to the study. Secondly, we hypothesize that in the CP group, the intensity of the psychopathology might have discouraged participation, which lead to higher rates of refusal and impossibility to reach out the participant by phone.

4.2. Familial vulnerability and environmental factors

We explored the role of several factors on TAY psychopathology, which have been grouped into three main categories: (1) familial vulnerability, (2) childhood environmental factors, and (3) present environmental factors. Due the recruitment selection of participants, the two groups differed significantly in terms of psychopathology. The interaction of each factor and the group was explored.

Youth psychopathology appeared significantly different according to the presence of parental history of psychopathology, in both CP and NCP groups. However, the effect of one or both parents with history of psychopathology was not cumulative, the only significant effect on TAY psychopathology was whether at least one parent presented a positive history of mental disorder or not. It is already known that parental mental illness represents a double burden for children and adolescents in both genetic transmission (4, 48) and family-related factors, such as a worse family environment and interaction between parents and children (49). Our study findings on familial vulnerability support the link between genetics first-degree familial mental health disorders and TAY psychopathology.

All types of childhood trauma have been proven to be a common matrix in the emergence of non-specific psychopathology, playing a crucial role in the factor "p" model (11). Our study findings are consistent with the previous literature. Childhood trauma (CTQ) was statistically correlated with TAY psychopathology, and the interaction with the group was significant. This leads us to suggest that childhood trauma might trigger to seek for help at the transitional age. Further research in other TAY samples should be performed to verify this hypothesis.

Family environment plays a mediating role in the relationship between stressors and children and adolescents'

TABLE 3 Youth-Self Report (YSR) scores according to group and familial vulnerability, childhood or present environmental factors.

Variables	Total (n = 220)		CP (n = 106)		NCP (n = 114)		p-value		
Family vulnerability	Mean	SD	Mean	SD	Mean	SD	Variable	Group	Interaction variable-group
1st degree family history[a]									
None	58.65	9.55	64.67	8.67	55.23	8.28	0.022*	<0.001***	0.681
One parent	66.53	8.59	68.25	8.24	58.50	5.09			
Both parents	66.69	12.01	68.63	10.41	63.60	14.98			
	b [95% CI]						p-value		
Childhood environmental factors	Variable		Group		Interaction variable-group		Variable	Group	Interaction variable-group
CTQ[a]	0.44 [0.22; 0.37]		−0.38 [−4.99; −2.80]		0.12 [0.01; 0.16]		<0.001***	<0.001***	0.025*
FAD[a]	0.36 [0.31; 0.58]		−0.42 [−5.38; −3.16]		0.04 [−0.08; −0.19]		<0.001***	<0.001***	0.430
	Mean	SD	Mean	SD	Mean	SD	Variable	Group	Interaction variable-group
Country of birth									
Belgium	61.03	10.32	66.64	8.78	55.96	8.54	0.715	<0.001***	0.438
Other	61.97	10.17	66.00	9.87	57.74	9.21			
Adoption									
Yes	61.83	8.40	67.67	4.16	56.00	7.55	0.900	0.003**	0.844
No	61.18	10.22	66.49	9.06	56.26	8.69			
Psychomotricity									
Yes	63.65	8.90	66.55	6.09	58.33	11.26	0.633	<0.001***	0.642
No	61.00	10.26	66.52	9.25	56.14	8.52			
Speech therapy									
Yes	62.61	10.33	67.61	7.71	54.35	8.74	0.798	<0.001***	0.134
No	60.66	10.08	65.91	9.58	56.74	8.59			
Reading delay[b]									
Yes	62.94	10.84	68.33	9.73	57.56	9.46	0.453	<0.001***	0.899
No	60.97	10.07	66.43	8.78	56.20	8.62			
Writing delay[b]									
Yes	58.50	11.50	67.67	10.07	54.57	10.23	0.891	<0.001***	0.642
No	61.31	10.06	66.66	8.78	56.43	8.58			
Grade retention[b]									
Never	59.23	10.16	67.23	9.23	55.04	7.89	0.409	<0.001***	0.059
Once	63.65	9.31	65.78	8.55	60.16	9.63			
Twice or more	64.69	10.31	66.48	9.45	57.20	11.45			
Present environmental factors	Mean	SD	Mean	SD	Mean	SD	Variable	Group	Interaction variable-group
Civil status[b]									
Single	61.24	10.25	66.58	9.07	56.34	8.73	0.473	0.001**	0.797
Cohabitant	59.86	7.56	65.00	5.48	53.00	1.73			
Parental marital status[b]									
Married	59.00	10.09	65.67	9.19	54.89	8.31	0.027*	<0.001***	0.658
Divorced/separated	64.23	9.90	67.68	8.72	59.00	9.38			
Other	58.91	8.58	63.40	9.08	55.17	6.28			

(Continued)

TABLE 3 (Continued)

Present environmental factors	Mean	SD	Mean	SD	Mean	SD	Variable	Group	Interaction variable-group
Living arrangements[b]									
Family	61.21	10.26	67.08	8.76	56.31	8.69	0.26	**<0.001*****	0.604
Other	61.06	9.74	62.86	9.70	54.75	7.85			
Education and/or employment[b]									
Yes	61.12	10.30	66.67	9.19	56.32	8.70	0.514	**<0.001*****	0.900
No	62.54	7.77	65.10	6.39	54.00	6.08			
Alcohol[b]									
Yes	61.69	9.70	65.15	8.63	56.40	7.38	0.284	**<0.001*****	0.446
No	60.42	10.88	67.38	9.38	56.02	10.42			
Tobacco[b]									
Yes	63.77	10.17	68.83	8.12	56.77	8.50	0.117	**<0.001*****	0.286
No	60.19	10.02	65.33	9.18	56.10	8.72			
Cannabis[b]									
Yes	63.53	9.67	67.54	8.71	57.28	7.64	0.274	**<0.001*****	0.911
No	60.24	10.24	65.93	9.10	55.97	8.91			
Other drug[b]									
Yes	65.43	7.37	67.17	6.31	55.00	0.00	0.952	**0.021***	0.839
No	61.06	10.23	66.48	9.11	56.27	8.67			
Disruptive behaviors[b]									
Yes	65.98	9.26	70.28	7.43	58.19	6.95	**0.014***	**<0.001*****	0.331
No	59.97	10.05	65.10	9.11	55.94	8.87			

[a] Data is missing for 24 participants (16 CP and 8 NCP); [b] Data is missing for 9 participants (7 CP and 2 NCP); $*p < 0.05$; $**p < 0.01$; and $***p < 0.001$. CTQ, Childhood Trauma Questionnaire; FAD, Family Assessment Device; CP, clinical population; NCP, non-clinical population; SD, standard deviation; YSR, Youth Self-Report. Bold values are the statistically significant *p*-values.

TABLE 4 Summary of the GHQ scores according to most significant factors.

Variables	Total (*n* = 196)		CP (*n* = 90)		NCP (*n* = 106)		*p*-value		
Family vulnerability	Mean	SD	Mean	SD	Mean	SD	Variable	Group	Interaction variable-group
1st degree family history									
None	15.38	7.78	18.78	8.58	13.44	6.59	**0.028***	**0.001****	0.743
One parent	21.18	8.54	22.61	7.97	14.50	8.60			
Both parents	22.31	7.78	24.63	6.89	18.60	8.14			
	b [95% CI]						*p*-value		
Childhood environmental factors	Variable		Group		Interaction variable-group		Variable	Group	Interaction variable-group
CTQ	0.24 [063; 0.020]		−0.36 [−3.97; −1.94]		0.02 [−0.06; 0.8]		**<0.001*****	**<0.001*****	0.782
FAD	0.26 [0.14; 0.38]		−0.37 [−3.99; −2.02]		−0.32 [−0.54; 0.59]		**<0.001*****	**<0.001*****	0.592
Present environmental factors	Mean	SD	Mean	SD	Mean	SD	Variable	Group	Interaction variable-group
Other drug									
Yes	14.00	6.25	15.83	4.31	3.00	.	**0.049***	**0.012***	0.504
No	17.15	8.30	21.11	8.22	13.65	6.66			

$*p < 0.05$; $**p < 0.01$; and $***p < 0.001$; complete results are available in the annex section. CTQ, Childhood Trauma questionnaire; FAD, Family Assessment Device; GHQ, General Health Questionnaire; CP, clinical population; NCP, non-clinical population; SD, standard deviation. Bold values are the statistically significant *p*-values.

psychopathology (50, 51), but the impact on youth still needs to be established. In our TAY sample, family functioning appeared to be significantly correlated with psychopathology. We can affirm that these findings about family functioning in TAY provide consistent evidence about the relation with youth psychopathology.

TABLE 5 Best model selection: significance of the predictors on YSR.

	Variables	AIC	*p*-value	Model coefficients	95% confidence interval
Model 1	Group	790.13	<**0.001*****	–0.377	(–9.997, –5.340)
	CTQ		<**0.001*****	0.284	(0.094, 0.310)
	FAD		**0.044***	0.152	(0.005, 0.377)
	1st degree family history		0.159	0.084	(–0.580, 3.507)
Model 2	Group	790.17	<**0.001*****	–0.383	(–9.994, –5.617)
	CTQ		<**0.001*****	0.305	(0.107, 0.310)
	FAD		**0.047***	0.149	(0.003, 0.365)

*$p < 0.05$ and ***$p < 0.001$. AIC, Akaike Information Criterion; CTQ, Childhood Trauma Questionnaire; FAD, Family Assessment Device; YSR, Youth Self-Report. Bold values are the statistically significant *p*-values.

Among present environmental factors, only parental marital status and disruptive behaviors were found to be significant. As discussed above, parental marital status was statistically different according to the group. Our TAY sample psychopathology appeared to correlate significantly with this factor, as is the case for children (37). The main hypothesis explaining the correlation between psychopathology and disruptive behaviors seems to be the possible redundancy of this variable with two YSR sub-scales (rule-breaking behavior, and aggressive behavior) (16). However, we should point out that total scores for psychopathology were significantly correlated to disruptive behaviors in our study sample.

The results on substance use should be discussed. None of the assessed substance was significant to TAY psychopathology. This finding could be explained in two different ways. On one hand, we could hypothesize that substance use does not intervene in the emergence of psychopathology, it is rather a consequence of psychopathology as a subsequent manifestation. On the other hand, in occidental societies, substance use is increasing in youth, and the age of first drug consumption is lower than it was in the past, and this is certainly the case in Belgium (52). Hence, substance use in youth might be not uniquely correlated to psychopathology, but also connected to societal trends.

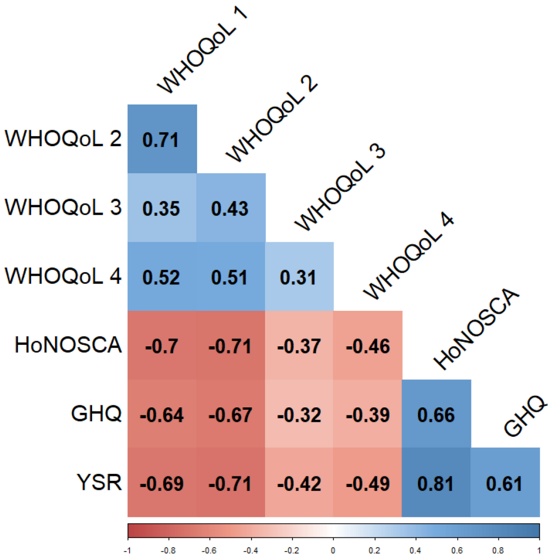

FIGURE 3
Correlation matrix between WHO-QoL, HoNOSCA, GHQ, and YSR. Correlation coefficient, method = Spearman's rho; all coefficients are bold because they are statistically significant ($p < 0.05$). GHQ, General Health Questionnaire; HoNOSCA, Health of The Nation Outcome Scales For Children And Adolescents; WHO-QoL, World Health Organization Quality of Life (domains: (1) physical health, (2) psychological health, (3) social relationships, and (4) environment); YSR, Youth Self-Report.

Similar considerations can be pointed out about enrollment in education and/or employment. However, in this case, the causative role of psychopathology on school drop-out and reduced work activity has already been recognized (39, 40).

4.3. Predictive dimensional models

The best subsets selection aimed to present dimensional models for the most predictive outcomes on TAY psychopathology. Results on YSR identified the three best predictors in youth: group, childhood trauma (CTQ), and family functioning (FAD). Family functioning was already correlated with youth psychopathology in urban areas, particularly regarding internalizing symptoms (53). Recent studies showed a positive correlation between adverse childhood experiences and neurodevelopmental disorders in children, leading to a greater risk of poor health outcomes in childhood and adolescence through the mediation of maladaptive stress calibration (54–56). Thus, the presence of childhood trauma and poor family functioning in strong association with the psychopathology in our TAY sample seems consistent with these models. These findings also need to be interpreted in light of some CTQ specific characteristics. The majority of items in the CTQ are related to past experiences with family and parents (29, 30), consequently concordant results between CTQ and FAD seem consistent with previous studies in University students (57).

The first-degree family history of psychopathology, even if significant in the linear regression, appeared to be redundant to predict TAY psychopathology. This might be related to different hypotheses. In particular, we found an already proven correlation between maternal mood disorder, youth comorbidity, and worse family functioning among bipolar youth (58). Additionally, high parental stress is a major risk factor for childhood maltreatment. Childhood maltreatment has been proven to worsen psychopathology, in particular symptoms related to neurodevelopmental conditions (54, 59), but also to cause psychopathology if combined with other genetic and environmental risk factors (60). For these reasons, we highlight that childhood trauma and poor family functioning might be strong enough explanatory factors associated with belonging to a clinical population.

The influence of genetics on psychiatric disorders (4, 48) is far from being rejected with these findings on familial vulnerability. However, dimensional models showed that, in clinical practice, assessment of childhood trauma and family functioning in the clinical population might be more efficient in the prediction of psychopathology in TAY.

The most predictive dimensional models on GHQ showed three similar significant factors: group, family functioning (FAD), and first-degree family history of psychopathology. These findings are essential for the screening of TAY psychopathology in primary care settings. General practitioners and other first-line health professionals might benefit from this model to efficiently screen psychopathology in youth and, if needed, refer to specific psychiatric care.

4.4. Quality of life and care needs

The positive and strong correlation between YSR and HoNOSCA confirms that as TAY psychopathology increases, care needs are greater. HoNOSCA has already been proven to be a cheap and efficient tool to monitor care needs in youth at the transition boundary (61). This instrument should be considered in clinical practice because of its specificity to correlate with psychopathology in detail. The correlation between GHQ and HoNOSCA is positive and significant, but less strong than the correlation between YSR and HoNOSCA. These findings are consistent with the fact that GHQ is a more sensitive tool to screen psychopathology, when compared to YSR, but may be less specific to determine care needs in depth. As already discussed above, GHQ finds its interest in primary care. In general, these results about positive correlation between TAY psychopathology and care needs support the encouraging trend of mental health literacy in youth (46), which is the best strategy to improve early intervention (62, 63).

Youth psychopathology and all quality of life domains are negatively correlated in the *Transition_psy* study sample. In particular, greater correlations are found in physical and psychological health, meaning that these two domains are more related to youth psychopathology in our study population. In the clinical sample of the Milestone European study, the most impacted domains were psychological health and social relationships, even if not significant in the comparison between usual care and managed transition (61).

Concerning quality of life itself, our findings show that all four domains correlate positively among them in a significant way. TAY's physical and psychological health positively and strongly correlates. Environment moderately correlates with both physical and psychological health, whereas the correlation between the social relationships and the other three domains is not very strong. To date, few studies are available in the literature showing WHO-QoL inter-domain correlation in TAY. One of these was conducted in war-affected youth in Sierra Leone; these findings show lower positive correlation between physical health with psychological health and environment, whereas the correlation between environment and psychological health seems more similar (64). Further research should be led to test more detailed hypotheses on this subject.

4.5. Limitations

Certain limitations in this study should be further taken into account, mainly the differences about the CP and NCP groups and the impact of COVID-19 pandemics on youth mental health.

Participant groups (CP or NCP) were selected according to the participants' active involvement in inpatient and/or outpatient mental health care, in the recruitment setting. However, all data were collected *via* self-report questionnaires, resulting in the absence of clinician and/or parents information. Among the CP population, a small proposition of participants did not self-reported being involved in mental health care even if they were recruited in one of the clinical settings. This example illustrates that information bias is a key problem in the assessment of research study designs and a distortion of results must always be considered (65).

Selection bias must also be taken into account in this study. A considerable proportion of NCP youth declared involvement in inpatient or outpatient mental health care or psychotropic drug use previously to the recruitment phase. Even though they were not actively seeking mental health care, their previous life experiences may have caused a distortion in results, probably by reducing differences between the two groups. Additionally, a selective refusal to participate in the study was perhaps observed. Participants needed to be motivated to answer questions about their mental health status and psychological vulnerabilities. It is possible that some responders dropped out of the study for two opposite reasons: either their psychological discomfort was too high to participate in a mental health study (most probably in the CP group) or, on the contrary, they did not feel mental health was a priority during the considered period (particularly among the NCP group). Also, recruitment strategies between the two groups were slightly different as described in the methods section (15, 66).

Additionally, data were collected between June 2020 and December 2021, mostly during the COVID-19 pandemic phase. It has already been shown that mental health problems urged during this period, particularly in adolescents and young adults (47). The impacts of the COVID-19 pandemic on mental health research are not yet properly acknowledged but this element must be considered in our study.

5. Conclusion

A complex model, including familial vulnerability and environmental factors, is needed to understand the emergence of psychopathology in TAY. To date, the trans-diagnostic and dimensional approach seems to be the most appropriate one.

With this study, we propose a predictive dimensional model on TAY psychopathology that includes belonging to a clinical group at the transitional age, childhood trauma, and family functioning. To date, few studies have been conducted on how to predict psychopathology in youth. Further research is needed to replicate these findings and to study other factors' role in TAY psychopathology.

In terms of implications in clinical practice, early and specific assessment of the emergent psychopathology in TAY is crucial to allow early intervention and to improve lifelong prognostic. The proposed predictive dimensional model might be implemented in clinical practice to alert professionals about the presence of psychopathology in TAY.

Additionally, HoNOSCA seems an efficient tool to establish care needs in psychiatric settings at the boundary between Child and Adolescent Mental Health Services and Adult Mental Health Services. GHQ could be used as a consistent screening tool of psychopathology in youth in primary care settings.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving human participants were reviewed and approved by IRBs of Queen Fabiola Children's University Hospital, Brugmann University Hospital, and Erasmus Hospital. The study was conducted in accordance with the International Conference on Harmonization (ICH) for good clinical practice (GCP). Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

Author contributions

VD was the chief investigator who conceptualized the study and wrote the grant funding proposition, together with MD, HN, CK, and JR. SM, JR, and EB-S collected and analyzed the research data and prepared the first draft and subsequent versions of this manuscript. SM and JR were the research assistants and wrote the ethical approval documents. CL and HS contributed to the statistical analysis and results interpretation. VD, HN, MD, CK, CL, and HS revised the manuscript. All authors reviewed and approved the submitted version of this manuscript.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

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Transcultural skills for early childhood professionals

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Context: Transcultural skills are especially useful for those involved in the perinatal period, when parents and babies must adapt to one another in a setting of migration a long a focus of transcultural clinical practice.

Objective: The aim of this article is to provide useful transcultural skills for any health care worker (e.g., psychologists, child psychiatrists, midwives, family doctors, pediatricians, specialized child-care attendants, and social workers) who provide care or support to families during the perinatal period. It highlights the cultural aspects requiring attention in relation to representations of pregnancy, children's needs, obstetric complications, and postnatal problems. Taking into account the impact of culture on clinical evaluation and treatment can enable professionals to distinguish what involves cultural representations of pregnancy, babies, and sometimes of disease from what is associated with interaction disorders or maternal psychopathology.

Methods: After explaining the relevance of transcultural clinical practices to provide migrant mothers with better support, we describe 9 themes useful to explore from a transcultural perspective. This choice is based on the transcultural clinical practice in our specialized department.

Results: The description of these 9 themes is intended to aid in their pragmatic application and is illustrated with short clinical vignettes for specific concepts. We describe situations that are extreme but often encountered in liaison transcultural clinical practice for maternity wards: perinatal mourning with cultural coding, mediation in refusal of care, cultural misunderstandings, situations of complex trauma and of multiple contextual vulnerabilities, and difficulties associated with acculturation.

Discussion: The transcultural levers described here make it possible to limit cultural misunderstandings and to promote the therapeutic alliance. It presupposes the professionals will concomitantly analyze their cultural countertransference and acquire both the knowledge and know-how needed to understand the elements of cultural, political, and social issues needed to develop clinical finesse.

Conclusion: This combined theoretical-clinical article is intended to be pedagogical. It provides guidelines for conducting transcultural child psychiatry/psychological interviews in the perinatal period aimed at both assessment and therapy.

KEYWORDS

culture, migration, transcultural competencies, perinatal, maternity ward, pregnancy, birth

1. Introduction: the importance of acquiring transcultural skills

There are a thousand and one ways to think about, educate, and take care of babies across the world, and they are all equally effective in enabling good child development—both emotional and physiological. Several sociological, anthropological, and psychological works confirm this universality without uniformity (1, 2). Among the differences that may affect our clinical evaluation, we should take into account the distal interactions in the mother-baby relationship and the more proximal interactions in some cultural groups (3–5). In many cultural groups, for example, tactile and motor interactions count more than visual and verbal interactions: mothers carry babies frequently; massages shape babies, drain their impurities, and stimulate them. That is, babies are considered malleable, impure, from a world of invisible beings (6). They must be embodied and shaped for the world of humans. Elsewhere, it is eye contact and speaking that have become most important in interactions to compensate for the distal character of other interactions, e.g., swaddling to avoid skin-to-skin contact, strollers later on (1). There are numerous examples around the world of important differences in how to put babies to sleep, feed them, play with them, care for them, and address them. There is not one good way or one bad way to interact with babies.

The fundamentals, such as the theory of attachment described by Bowlby (7), must also be taken into account in the context and the environment in which infants develop, along with their mothers and the various, often multiple, attachment figures (and not only one or two people who care for the baby consistently and continuously to aid their social and emotional development).

The absence of uniformity also lies in the way of thinking of a family (8). The concept of a family is enlarged beyond the nuclear in many cultures: the opinion of a maternal uncle in a matrilinear society (such as Haiti, some regions of Indonesia, and Cameroon) about a child's education can be more important than that of the father, for example. In other places, the role of the elders and even the ancestors is important (Asia), and children belong more to the group than to their two parents.

These differences create the need for professionals to decenter, that is, to not interpret the unknown in terms of the known, for that risks eroding human complexity and inducing cultural misunderstandings. On the contrary, we must leave room for doubt when we are faced with ways of doing things that seem surprising to us.

Knowing how to adapt the framework and the questions in an interview with patients or clients allows us to gain access to their different cultural representations. In our health care departments in France, the republican ideology is supposed to protect against the risk of pointing out differences in patients from other cultures and other religions, so that patients can feel they are receiving care with equal rights and without either judgment or exclusion. These universalist positions avoid any insistence on the difference between communities or cultural groups (9). Others nonetheless underline the need to recognize the specific needs of minorities and to find appropriate responses to these needs (10–13).

The curriculum allocates very little space to the consideration of cultural elements in health care relationships (14). Care providers are often very prudent, even inhibited, about approaching cultural questions, sometimes to the point of being implicitly reluctant to use an interpreter in situations where a language barrier exists, although this can be considered to constitute a form of institutional abuse/mistreatment (11). Another pitfall has been observed in France: the logic of integration that is supposed to erase or at least erode these differences led professionals of early childhood to advise in the 1980s that the use of French should be privileged in personal family spheres. One consequence of this was an impairment of the language quality in family interactions in non-French speaking families. It is clearly recognized today, however, that using a mother tongue other than that of the host country does not slow down the acquisition of the host-country language. On the contrary, the more languages one learns, the easier it is to learn others (15).

The objectives of this article are simultaneously theoretical, clinical, and pedagogic to demonstrate the importance of taking these elements into account to obtain a better clinical understanding and evaluation of the situation in the perinatal period and thus be able to provide optimal support. The aim is to help care providers to acquire/adopt transcultural skills, that is, an overall set of aptitudes and attitudes that allow them access to these essential elements of understanding. Academic training is needed to impart transcultural skills to early childhood professionals, regardless of their discipline (16–18). Transcultural skills consist in adopting a dynamic posture toward the acquisition of skills (such as knowing how to work with an interpreter, to adapt one's manner in conducting an interview, general culture) and attitudes (nonjudgmental, and nondiscriminatory toward minority populations, decentering), which help to take the impact of culture on symptoms into account (19). The clinical evaluation is thus more detailed, without creating the feeling of stepping past the lines of our republican institutions. We know that these cultural elements suffuse/pervade/dominate/invade the thoughts of hospital patients, in particular during this period of pregnancy and childbirth. Tact is needed to show the usefulness of this exploration and its benefits for clinical evaluation, treatment, and support of mothers in the care related to mothering.

2. Implications for the clinical situation/for clinical progress: value of the transcultural approach in the perinatal period

2.1. Psychological vulnerability

The concept of psychological universality and of universality in ways of mothering a young child makes it impossible to label the way something is done in one culture to be the best way of doing it in any culture or to define any vulnerability as greater in the babies of a given cultural group. On the other hand, outside of their cultural group, families can find it distressing to give birth in a foreign country. Migration can induce a loss of bearings and undermine a sense of parental competence. Confronting different mothering techniques and questioning themselves about how to raise a baby born in a foreign land are factors that make parents especially vulnerable during this period. Babies themselves can appear strange and foreign to their parents' eyes simply because they were born in another country (11). What will their parents be able to transmit to them of their cultural values, of their identity? Migration, especially if recent and if the couple or the mother is isolated, induces a sudden break from previous reference points (3, 4, 20, 21). The absence of a word, a practice, of support at the start of their baby's life/at the start of their life as parents—a moment when the group is primordial—can make isolated parents still more insecure (20). That is, numerous cultural practices exist to protect mothers and babies and to enculturate babies: for example, among Hindus, both naming according to specific astrological criteria and a head-shaving ritual for purification. In several non-Western cultures, the mother must comply with a period of rest and social withdrawal after giving birth. During this time, the baby is presented to the world and the mother is released from her domestic tasks, surrounded and supported by older women. Pregnancy and childbirth are among the life stages that require rites. Among these, Van Gennep (22) distinguished rites of protection from rites of passage. The first are intended to protect the mother and child from perinatal risks but also from dangers and the malevolence of both the visible and invisible worlds. The rites of passage, on the other hand, make it possible to "forbid, from birth, even pregnancy, the fusion of the mother and the baby, to unify the body and the mind or soul (the psyche), and to manage the anxiety of death inherent in humanity" (23). The childbirth is thus marked by a period of reclusion (margin period), followed by rites of reintegration and a social return from childbirth, which is the moment of both the mother's churching ceremony and the baby's second (psychic) birth (23–25).

This planned isolation period of 40 days exists in several cultures, not solely in Muslim areas. In the words of Ben Mohammed (26, p. 184) about North Africa, "the 40 days following the birth of a child are particularly dangerous for the mother and child. The tomb remains open during that time." The mother–child dyad is isolated, especially during breastfeeding, so as not to attract "the evil eye of envious women who might make the milk go dry." Other mothers have stated that in China too, the rule is "confinement" for a month after giving birth: neither the mother nor the baby is allowed to leave the house, except for medical appointments. And in this case, the mother must cover her head. She must also avoid tiring activities such as reading or watching television and ideally, she must stay mostly in bed. She must avoid washing her hair and therefore must settle for a quick sponge-down with hot water and lemongrass oil. At the end of the month, a big party is organized, bringing together all family members.

Distance from the country and the absence of the group can make these practices more difficult. Some families may feel insecure about whether they have performed these practices correctly. Whether or not we are superstitious, we are carried by a collective unconscious, and we in the West also have our own beliefs without always realizing that they are cultural and not universal. We need to feel that we belong to a group at moments of vulnerability or during life passages. This cultural dimension can become important in particular when there is a problem or an illness. Inversely, the fact of feeling active is protective and can help a parent to emerge from a traumatic shock. It is not unusual to see a mother or an isolated couple frozen due to a recurrent trauma or just by the fact of not knowing, not recognizing, or not daring to know how to express it because of the language barrier and the cultural distance from the professionals attempting to offer support.

Sometimes, in the absence of the group, only the father is present; he may find himself in a place that is not supposed to be his. In some cultures (25) it is even bad luck for the father to be present at the birth. When he finds himself invested and involved in mothering the newborn, the risk is that he may deepen the gap with traditional mothering and induce guilt and a feeling of incompetence in the mother (27). In these situations where the initial milieu is traditional, what is missing is other women (cousins, aunts, and grandmothers), co-mothers and especially the mother's mother. Rodriguez describes this particularly for cesarean births to migrant women (27). Some studies nonetheless qualify the ambivalence of this place: the presence of the father alone may mitigate the mother's feeling of isolation and reduce some of the risks associated with it. Lameche (28) described this in a study of a newly arrived North African population. The report RéMI related to the cesarean rate, which appears to rise less when the partner is present (29).

Forms of breastfeeding can also be transformed during migration: faster passage to mixed breastfeeding, because of preoccupation about the need to work and make a living as rapidly as possible. Some mothers say they fear the child "will get used to being carried" and avoid doing that frequently because there is no one else to do it (30).

2.2. Vulnerability that can be transformed into psychopathology

The risk of psychopathology is greater among migrant patients, especially for pre- and postpartum depression, which affects, respectively, 38 and 50% of migrant women, with a higher risk for more recent migration (31–33). Complex post-traumatic stress states are also overrepresented (34, 35). In the first place, we often find in clinical pictures of complex PTSD impaired trust in others. That is, trust has been endangered in repeated, intentional episodes of trauma. Dissociative symptoms, somatization, and feelings of hatred and anger are also frequent in the general migrant population. The repetition and/or reactivation of traumatic events contributes to them. The traumatic valence of a migrant's journey may be found at different three time periods: before migration, during the voyage, and after arrival. Before the migration, some departures are associated with traumatic situations, civil war or conflicts, or a flight from complex social or family conditions. Some mourning or trauma can recur, sometimes months or even years later. The period of pregnancy is propitious to the emotional reactivation of these unresolved past events (3).

During migration, the journey can be affected by trauma. Some migrant women traveling alone experience sexual violence during their travels (36).

Afterward, the living conditions in the host country, sometimes precarization, the waiting time to the regularization of their socio-administrative situation, job hunting, and looking for housing can generate discouragement, anxiety, and disillusionment about the initial migration plan. Some symptoms are directly linked to precarization or isolation from relationships (37, 38). Micro-aggressions can also have an impact[1] (39). Finally, very preterm birth or obstetric complications can present the risk of the infant's death or disability and have a traumatic impact on families.

2.3. Physical vulnerability

In France, the number of at-risk pregnancies, cesarean births, and preterm births appears higher among migrants, particularly those from sub-Saharan Africa (31, 40, 41). Nonetheless, the literature on this point is not entirely consensual (42). The factors advanced to explain the excess risk of cesareans among migrant women are limited and inadequate. It could be linked to insufficient access to prenatal care, representations of pregnancy follow-up that are different from those of the country of origin, communication barriers, and often precarious and inhospitable conditions on arrival in the host country/France, fragile maternal health, a high body mass index, feto-pelvic disproportion, or even simply isolation (40, 43–46).

The precarity associated with migration is an important perinatal risk factor, as are isolation, absence of marital support, administrative status, and social insurance coverage (31, 45, 46).

3. Actionable recommendations: the interview in the perinatal period

Our transcultural team is composed of psychologists, nurses, and psychiatrists trained in transcultural clinical practices with Professor Marie Rose Moro, a leader of transcultural psychiatry in France, specialized in the clinical care of métissages (cultural mixing) in babies, children, adolescents, and their families.

We have deduced interview themes that allow a transcultural approach in child psychiatry during the perinatal period. These levers come from our diversified clinical practice applying transcultural methods, adapted and personalized to the situations for which this expertise is required. These include liaison work in the hospital, discussion and support groups with pregnant women or recent parturients (47, 48), transcultural consultations with a group of co-therapists from various cultures (3, 4, 49), mediation at the hospital with an interpreter-mediator, and institutional work (team supervision, university instruction or professional training programs, and reflexive interventions in multidisciplinary staff meetings). These guidelines also consider the bases of ethnopsychoanalytic theory (50), combined with approaches described in the international scientific literature (4, 14, 16–18).

3.1. Start the interview by creating the basis for an appropriate relationship

The interview's framework presupposes establishing the conditions for a balanced and horizontal relationship. The interviewer exits the position of expert and instead shows a posture of cultural humility (51). Sitting in a circle, sometimes sharing some water, can facilitate this first encounter (52, 53). Showing an authentic interest in the other's culture and representations can transform the relationship. This is done by asking in detail about their cultural identity, beyond their country or nationality. What are the family's cultural affiliations? What language do they speak? What city or village do they come from? Language is an important aspect of identity. Exploring the languages the patient speaks is often the occasion to praise those patient who are multilingual for this accomplishment. Both ethics and quality of care require finding an interpreter to ensure mutual understanding; it is thus necessary to identify the language the interpreter should speak. The help of family or friends has its limitations for announcing bad news (for example, a suspected malformation on ultrasound or complications in neonatology). Thus, partial mastery of a language cannot suffice in these contexts to ensure mutual understanding, especially as there can be an initial level of stunned speechlessness even in their native language, it is often necessary to repeat news that is difficult for everyone to understand. The interpreter, who is often someone with whom the patient can identify, enables the mother to assert herself, to not fear being judged: the cultural distance is thus reduced, and the interpreter facilitates bridges between the two countries by knowing the codes to both worlds. Finally, the expression of emotions is often facilitated by use of the mother tongue which, in turn, facilitates the narration [Ricoeur's narrative identity (54)].

Sometimes reluctance linked to financial cost or time are mentioned, or those linked to individual or collective unconscious factors. Studies show that the use of interpreters and mediators offers cost benefits for public health (55). It is important for therapists to show that they are making an effort to adapt to and respect the patient by seeking to share the same language.

The notion of hospitality in the welcome is also important, for it can contrast with the exterior world, sometimes associated with the austerity of migration policy (56). The tone of voice is almost as important as the content (57).

One of the principles of receiving patients in a place providing care is to create a relationship of trust, which is all the more important for women who have experienced intentional and repeated traumas. For that reason, it is important to explicitly state the meaning and nature of a "talking" therapies, of seeing a psychologist or psychiatrist, without this being associated with the mental illness of the "asylum" sector. Finally, the role of each professional must be clearly explained, especially for more highly medicalized pregnancies. The psychological support must be related to the risk of vulnerability, while assuring the patient that there is no association with a severe overt mental disorder.

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[1] Insults or attitudes, "intentional or not" that "communicate hostile or contemptuous messages targeting people solely on the basis of their membership in a marginalized group."

The families may not be able to understand the notion of interaction disorders unless the therapist describes them clearly.

Finally, one way to show that it is possible to approach the cultural dimension in the framework of a medical interview is to begin the interview by self-disclosure, that is, short personal revelations (while remaining professional) used to create empathetic understanding: they may concern hindsight about the experience of other patients or be a personal shareable and generalizable experience.

This self-disclosure aims to create cultural proximity (if the interviewer is comfortable with it), but also to demonstrate the diversity that authorizes patients to assume their culture and thus to be able to refer to it as a resource, to subjectively structure an issue. For example, near the end of an interview during the perinatal period: "Where I'm from, in North Africa, we organize this protection, that birth ritual, when a baby's born; and where you come from?" This type of interview sometimes humanizes the professional and promotes the therapeutic alliance.

3.2. Elaborate a narrative that mitigates the risk of migration cleavage and considers the possible traumatic dimension (re-establish continuity between before and after)

Child psychiatry and transcultural psychiatric interviews are often based on narrations (3, 5, 11, 52). The therapist accepts and welcomes the cultural narratives that serve as a container for the intimate individual material relating to the current situation of the mother, family, and baby.

Bridges are built between life "back there" and life here to avoid the risk of a migratory divide and to share cultural representations. Indeed, parents often tell their story starting from the day they arrived in France. If we do not actively look for information about their premigration history, it is impossible to have access to it. This reaction can be linked to the reading of migration trauma, with too great a separation between before and after. The cleavage of migration is a phenomenon described by Nathan (58): the experience that every migrant has of mental separation between before and after migration. It resembles the loss of a psychological and sensory envelope with which the migrant decodes the exterior world and stimulates the need to create another one, like a new skin.

Evoking the patient's migration experience, its determinants, its course, what she thinks of life before and after migration—these questions make it possible to help her establish a continuous narrative of the different periods of her life. Often the heroic valence of the journey is manifest in this account and deserves to be underlined for therapeutic purposes. These elements must be explored progressively; they can finally allow the patient to recover a feeling of the continuity of existence, the continuity of her existence (the *self-continuity* described by Chandler and Lalonde (59)). We search constantly for coherence in their pathways, so often characterized by family separation, cultural ruptures, and changes in their care. This coherence and this life story are necessary to cross/bridge the passages of life (52). In the face of this disillusionment and sometimes precarization, it is useful to ask about her professional identity, her education, her place in the family before migrating, and to recall who she is. Sometimes disillusionment and/or trauma seem to deprive the journey of its meaning. This must be reactivated to give her the strength to continue. Contrary to what is often thought, asking about the meaning of the journey or exploring the other's cultural affiliations is not uncomfortable when the therapist's interest is authentic. Finally, it is necessary to welcome disillusionment, to share the disarray sometimes, often to help actively on the social level, assuming this professional role, despite being a health care provider. That is, a patient cannot engage in psychotherapeutic work until her material survival and shelter are assured. In pregnancy or at childbirth, this need for a safe place can become obsessive and constitute a form of primary material preoccupation together with a primary maternal preoccupation (constructing a home).

She must be restored to her story both gradually (60) and prudently to take her own rate of progress into account: the traumatic valence of a journey exposes her to the risk of reactivation and emotional overflow. Time is needed to transform and develop these aspects at a psychotherapeutic level. Putting it into words—into a narrative/story—is sometimes painful. Regardless of the country one leaves, regardless of the reasons for leaving, there is inevitably a loss causing narcissistic injuries the extent of which is sometimes difficult to measure, but also, simultaneously, producing unsuspected gains (13).

3.3. Reconstruct the networks of belonging

On a practical level, the networks of kinship and social belonging are reconstituted during the interview: Which family members stayed behind in the home country? Who in the family is in France? What contacts do they have (telephone, frequency)? Has the baby's birth been announced? Are there any friends, people from the community, at hand? Is the parent able to speak his/her mother tongue? Even for the most isolated individuals, encouraging them to talk about family during a consultation, helping them to telephone family members, or encouraging them to do so, can help them feel less lonely, and loneliness is a very deleterious feeling during the perinatal period.

Do not be afraid of mentioning the people missing in France; on the contrary, make them exist in the session: the grandmothers, the child's father, even the children who remained behind. Prevost et al. (61) shows for example in her qualitative study of the experience of migrant mothers giving birth in France after leaving a first child behind in the country she left that there is a massive reactivation of the missing child during the new pregnancy. Often the women have left them to protect them and to bring them over later, once the material conditions are correct. Professionals may often not dare to raise the question of the children left behind for fear of making the mother feel guilty with anxieties about having abandoned them. Some professionals are also comforted by the idea that culturally favored entrusting of children does not engender distress. Recent studies point out the consequences and intrafamily relationship stakes that it raises. Even though this practice is recognized and valued in society, it is not without risk of psychopathology for both mothers and children (61, 62).

Finally, taking the cultural place of the extended family into account sometimes makes it possible to adapt the hospital's framework. Knipiler (63) described how the experience of a neonatology unit stay aggravates the experience of isolation and how the extended family can be allowed in neonatology departments that are aware of the limits of the concept of the nuclear family in Western areas.

The interpreter can also function as someone with whom the patient can identify; this can also help her to escape her sense of isolation. Finally, therapists/care providers/professionals can creatively employ local groups and associations. They can also discuss with the patient the difficulties of contacting the family, despite today's social networks which make it easier than in the past: these difficulties sometimes follow traumas (and/or the desire to protect the family "back there"). Finally, when mother has had not news of her family or friends because she had to flee rapidly, referring her to the Tracing services provided by the International Red Cross[2] can spark new hope.

3.4. Help her to escape a feeling of powerlessness and facilitate her psychological reinvigoration

Therapists faced with the patient's psychic exhaustion from confronting recurrent adversity must often take an active position, adapted to the patient's tempo of progression to escape their own feelings of powerlessness. We help to coconstruct solutions based on what emerges from the narrative, facilitated by the interview levers described here.

- An example of a woman's prenatal mourning with an acute traumatic reaction

> *Joëlle arrived in France recently. She came from the DR Congo and was hospitalized after her delivery. One of her twins died in utero. The other twin is alive, but Joëlle is stunned and does not seem able to succeed in paying attention to him.*
>
> *Joëlle explained that she cannot either eat herself or breastfeed the baby because the other twin is "out in the cold and frozen" an allusion to the morgue/mortuary. Her principal preoccupation was that "he's suspended between the world of visible and invisible beings," the latter being, in other words, the world of the dead and spirit creatures. She understood that we were not judging her and that we would not hesitate to talk about different cultural representations of the pregnancy and ontological representations of children. She understood this because of our explicit questions about how to welcome a child into the world, what birth rituals should be done, what mourning rituals were needed. She could not be primarily preoccupied with the living baby as long as his twin was not buried in due and good form. Moreover because the two were linked, neither could be appeased and recognized in the world of the living or the dead. But Joëlle's very great precarity, her isolation from family and any community (because she had arrived very recently in France), and her lack of knowledge of the organization of funerals in France aggravated her experience of isolation and prevented her from acting.*
>
> *Helping her to escape this sense of isolation was the first step to re-energizing her. First, we talked about her family. Even thought they were back in her country, far away, we made them re-exist in this interview. She said she had not been able to tell her mother, to whom she is nonetheless close, of her delivery, both because she lacked the money to make the call, but also to protect her. Wouldn't her mother herself feel helpless, even sad about Joëlle even as she was in her own difficult situation in RD Congo, a situation that Joëlle had fled precisely to be able to later help her family? In every culture, mourning is shared by the community/collectively. Culture surrounds/lines these difficult life passages to generate meaning and master the violence of the unexpected. What can be done when the person is far from her cultural group? We thus lent Joëlle our work telephone so that she could call her mother and connect with her and her country. This simple act probably prevented the prolongation of her hospitalization. The next day, she told us that her mother was planning to organize a ceremony there, with the help of the extended family, the day of the funeral. We obtained information about - and told her - the specific date this month of the burials of fetuses and newborns who died at the maternity hospital, and where it would be. There is a support system covering the costs of burial for families in situations of great precarity and/or who do not want the burden (fetuses who died in utero or medical terminations of pregnancy). We also informed Joëlle that she could see a priest at the hospital.*

Sometimes, it is other mothers who help the patient to find solutions and to escape from this psychic paralysis, even in the most extreme situations (very great precarity, extreme preterm birth). We have observed this in particular in support groups around cultural questions about welcoming a child (30, 47, 49). These groups help to mitigate the feeling of isolation and constitute a creative and relevant form of care at the maternity ward. We can cite here a conversation around African massages. The practice was relegitimized in one support group (47). One mother thought the practice was forbidden here, that the vigor involved could be badly interpreted by professionals. This led another to say that she would have liked to do this, that it was important to her, but she really did not know how to do it well, because it is grandmothers who handle the initial care after birth "back home," mothers being considered too gentle/subdued to do these massages. Hearing this, another mother from a related cultural group improvised the massage gestures on her baby for the rest of the group.

3.5. Promote harmonious cultural métissage

One of the coping strategies described in the literature (64) is the maintenance of continuity despite an unstable and shifting situation. It is thus desirable to facilitate access to familiar things—things the patient knows—to help her regain her bearings. This enables a transition to integrate the values of the host country more easily and to construct a harmonious cultural métissage or mix between two cultural worlds. The progressive learning of new cultural norms, as well as the maintenance of religious beliefs and practices within the host country are part of the strategies for achieving this construction. Links with peers from the cultural community already more accustomed to the host country and who have often endured identical

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[2] Tracing department and Restoring family links program provided by IRCR https://familylinks.icrc.org/

or similar situations can play a major role, as sharing experiences mitigates isolation and discouragement. A double support network, in the host country and the country of origin, may have a protective effect on the mental health of migrants (14, 64).

A harmonious cultural métissage protects against the stress of adaptation to the culture of the host country. Metissage is an identity building continuum (or a set of processes) that integrates the feelings of belonging to many cultural universes. It is found among children from mixed couples or couples from migrant backgrounds or among child migrants, who tend to adopt progressively some of the cultural norms of the adopted country.

These involve maintaining a feeling of belonging to the original culture, while adopting new affiliations in the host country—all as part of a creative process integrating the values of each culture. Acculturated individuals[3] may thus be the most highly exposed to this risk of psychic vulnerability (65). That is, we encounter in our everyday transcultural clinical practices couples or isolated mothers whose sense of identity is sometimes lost due to their cultural mixing.

Behind their resistance or their lack of knowledge of cultural theories, we often see a willingness to understand the cultural sense of the symptoms of their baby or child, leading them to rethink the disease from a cultural perspective, because medicine has not led to recovery, or because the coding of the symptoms clearly evokes a cultural interpretation. Often this occurs only because of the patient's cultural group, and one of the means of treatment is precisely to link it to the culture of the country they come from, in one way or another (66).

Within a single country, the effects of globalization and accessible media and social networks engender rapid transformations. Knowledge is mixed in a single generation, which is evidently valuable, but is also a loss of bearings and identity across a single generation. The painful moments of life (a chronic disease that medicine cannot cure) or its passages (birth, death, becoming an adolescent or reaching adulthood), when they are difficult, sometimes call for cultural readings.

3.6. Construct a narrative of filiation

It is also our goal to attempt to elaborate a narrative surrounding the child and to develop a form of narrative of parenthood (67), thereby placing the child or the future child within the family history. The focus is also on the notion of a cultural mix or blend: help with the construction of self between the two cultural worlds the patient belongs to and the creative use of the wealth from both (without this appearing incompatible). "There is no filiation without putting a history into words, without a narrative, without a narrative on filiation" (Golse & Moro, op. cit.). Putting the baby's history into words is paramount. The feeling of containment, protection, and roots procured by the family and the entire group of belonging is primordial because it enables the mother and child to be part of an intergenerational web—filiation and affiliation (68–70).

In the perinatal period, asking about the history of the child's name makes it possible to identify first of all who he is and often leads to an important story of his history and his parents' investments. Given the concept of an "exposed child" at risk of vulnerability because he is the first to be born in France, this work of making him a history is very important. It also reduces the risk of reciprocal feelings of strangeness.

Without the group, it is sometimes difficult for mothers to achieve this, because of the massive nature of their worries, their numerous disruptions, exposing them to a risk of "mental blank," withdrawal, silence, and encryptment. Therapists can sometimes find themselves in the midst of extremely problematic issues: the reaffiliation of a baby born from a rape, which can be thought of/handled in several ways: by naming, or through religion, membership in a matrilinear line or a new mixed cultural group, etc. (71). In these cases, we sometimes use a valuable narrative support, the transcultural genogram (72).

It makes it possible to represent families who come from elsewhere, taking into account migrations as well as blended definitions of the family object in its psychoanalytic and anthropological sense. It thus provides support for the narration of family stories for migrant families who sometimes find it hard to conceive and transmit these bonds.

3.7. Make the family the expert, decenter oneself

At all of these stages, it is essential to adopt a decentered posture (distancing oneself to avoid ethnocentrism) in particular when one has a doubt, a feeling of strangeness, of missing the content of the clinical encounter. To learn from the family, it is important not to fear facing otherness. Assuming that some things are not spoken of/kept quiet, we recommend showing (and thinking) that you are ready to listen without judgment.

The interview is based on co-construction: the patient is given an expert role and a mother is invited to express herself the way she wants, to be fully heard, avoiding turning what is unknown into what is known. This can be done through narratives such as: "what is usually done in your country to welcome a baby? What are the rituals at birth?" The mother can therefore become an ambassador for the collective or family knowledge that she wishes for and chooses. What is most important is the meaning for her of whatever cultural or religious practice she decides to honor, as migration is bound to trigger changes, choices, and mixing. It is important to avoid the pitfall of cultural generalities: for example, that a woman who comes from a rural area does not say the same things as a woman from the city.

Finally, we aim to co-construct solutions: "even if we cannot do exactly the same as in the home country, something can be done, for instance, asking the family to carry out part of the ritual over there," or else over here, even if it is not on the day initially required, or carry it out symbolically and partially. Culture is a dynamic process, constantly changing, and certain adaptations in practices on foreign land are perfectly valid.

This approach often helps people to escape a feeling of powerlessness and enables skills to emerge despite a traumatic past.

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[3] A cultural transformation of identity that takes place in contact with the culture of the host country. The stress associated with the phenomenon of acculturation is recognized as a risk factor for the decompensation of several psychiatric disorders.

Here we cite the example of African massages of babies, or the technique of putting Tamil babies to sleep with rapid, jerky movements, the tonicity of which can surprise the untrained eye (1). The error of interpretation would be to consider them to be shaken babies, even as the techniques of this form of mothering, transmitted from generation to generation, protect the baby from any shock. The transcultural posture consists, on the contrary, of leaving room for a tolerable doubt: leave yourself the time and give yourself the means to apply the procedure used in complementarism that combines a (non-simultaneous) anthropological and psychological/medical reading (73). Therapists must be capable of listening non-judgmentally and non-ethnocentrically, even if they do not understand.

- A Kurdish family and its very preterm baby

> *Elif, a young woman of Kurdish origin who arrived in France shortly before giving birth to a very premature baby. Her social and economic situation was precarious. She did not speak French. The professionals in the neonatology department were astonished because the she was discharged but has not been to visit the baby for the entire first week in the department. The parents did not understand the hospital staff's explanations. They could not be reached by telephone. The team was feeling alarmed and powerless about the situation. At the end of a week, the parents returned with clothing and a stuffed animal for the newborn. They were accompanied by a friend who interpreted for them and the staff. They then explained the status of babies in their culture. For them, babies do not yet belong in the world of humans when they are born. They are suspended between life and death, between the world of the visibie and the world of the invisible. The parents were therefore not yet authorized to meet them. Now, after seven days of life, he can be named and welcomed to the world of humans.*

It is not rare for several parents from diverse cultural minorities to say that they are not coming to see their very preterm child because he does not belong to them yet. They entrust the infant confidently to the medical world. This does not at all prejudice their future investments in the child.

3.8. Elaborate a meaning

This transcultural posture allows access to cultural representations and sometimes to etiological theories. These cultural theories (74) give meaning to distress or suffering or a difficult event and may offer comfort beyond the medical cause or even allow the family to take action and thus contribute to the child's wellbeing. It is frequent to see families give a cultural meaning to the chronic disease of a child whom medicine cannot cure or to explain a death, a singularity. From this sense of/search for meaning comes a cultural therapeutic logic that allows patients to overcome their feeling of powerlessness. They can for example, rename a child, provide a dowry for the marriage of unmarried parents, recognize the child's ontological status, and perform the appropriate rituals to keep the infant in the world of humans. For example, the ontological representation of autism in West Africa can enable parents to consider this singularity acceptable (and culturally coherent). In some cultural groups, when a person is unwell, an external cause is sought (attack by witchcraft, badly named child, unprotected family alliances, unpaid dowry, etc.). To harm a group or a family, the most vulnerable in the group can be targets, a child, for example. Culture intervenes to enable thinking and healing, by allowing action and mastery of the associated anguish by rites of holding, cultural protections, and by giving meaning to the meaningless (75).

In some situations of unsuccessful treatment, this development of these etiological levels and anthropological treatments can take place in transcultural consultations in a department offering therapist groups with a flexible number of therapists trained in transcultural clinical practices (3).

Some interview questions can help to develop a narrative around the elaboration of a meaning: how is a child welcomed in the home country? How would the problem of a pathological pregnancy or a maternal psychological pathology be described? What would be done? How would the problem or disease be named? Have others been harmed and what could be done (or wasn't done that is possible today)?

There again, the etiological theories must be considered in a context and cannot be generalized: they depend on the cultural group (or groups) involved, the relation to migration, what the combined symptoms suggest, the family's singular history, and the individual and collective knowledge of these situations. They may be part of a global framework of intergenerational distress, without an association with a theory for a specific disease.

- Refusal of a cesarean for a "sleeping baby"

> *We were called into the delivery room for a young woman from Somalia, Faduma, to attempt to resolve a situation in which she was refusing a cesarean, delivery despite the risk of fetal death, with an argument based on cultural elements. Faduma's partner, the father of the unborn child and her only source of support in France is also opposed to a cesarean.*
>
> *The medical team informs us that the interviews have thus far been conducted through a staff member who speaks Arab, as Faduma does not speak French. Faduma has been in France for only months; she fled her country for reasons related to family trauma (fear of a forced marriage, with vague notions of excision mentioned). In her journey, Faduma crossed Libya and learned Arab. She was also violently raped. Because she was post-term, labor had been induced several hours earlier. The fetus responded poorly to the induction and appeared to present fetal distress (heart rate decelerations). The cervix remained too closed to allow vaginal delivery.*
>
> *On arrival, we decided to sit down at her level. We established a framework at a pace contrasting with the context of a life-threatening emergency. We used mediation techniques, including an interpreter in Oromo, a language which the mother spoke more fluently than Arab; given the emergency situation, this professional interpreter worked by telephone. In the presence of the midwife and the obstetrician, we retranslated the medical reasons and the risks and also translated for them her precise cultural logic (see reference mediation).*
>
> *What we learned during the interview in Oromo illuminated three essential points:*

> *On one hand, Faduma insisted that she could not be mistaken about the date of the child's conception. Was she insisting that her partner's paternity was unquestionable paternity and that she had no doubt that she did not conceive during her rape in Libya? Might it not influence the fetus's fate if the conception resulted instead from rape? She thinks she is at term especially as she feels a discordance between what the monitoring is saying and her own physical sensations that she wants to interpret. She says she is attached to traditional techniques of childbirth. According to her religion and practices, prenatal care should not be too medicalized (and she was identified only late at the maternity ward) and one does what "God has decided." Through ignorance, she said, she accepted an epidural, and it was preventing her from perceiving the painful contractions. She felt that the baby could still wait to come out "naturally" and thus refused the "surgery." When we asked her to describe what she thought the surgery is, a second point of understanding emerged: she considered it similar to a "long laceration," with a statement that reactivated the experience of her infibulation[4] but also the fear of disinfibulation associated with a cesarean.*
>
> *Finally, she explained to us an ontologic conception that can make sense of a pregnancy that can sometimes last a long time, well beyond 9 months and can nonetheless be considered at term. This ontologic theory of the "sleeping baby" is culturally coherent and is known in several cultural areas, including in North Africa and sub-Saharan Africa. It developed to make the inconceivable conceivable at a time when infant mortality in those regions was high: the same fetus who left for the world of the dead during a miscarriage or an in utero death can return several months or years later.*
>
> *This transcultural mediation took place in a particularly benevolent ambiance: Faduma only spoke when we assured her that we would not force her to have the cesarean and that we would respect her decision. The obstetrician took her traditional representations seriously and responded clearly and appropriately to Faduma's precise fears. She thus reassured her about the type of surgical incision. She also answered Faduma's questions, including her worry that the fetus's poor state was because she could not eat (concern related to the infusion).*
>
> *Can we represent the experience of strangeness/ foreignness? Of cultural distance? Seeing so many "scopes"… when the pregnancy can be hidden to protect it (from the evil eye).*
>
> *She and her partner finally agreed to the cesarean, and their first interactions with the baby went well. The physical health of both mother and baby was also good.*

Other mothers mentioned that cesarean birth is viewed as a cultural, religious, or even sexual transgression. They risk being repudiated by their husbands, branded as sterile, and accused of the baby's malformations (76).

We note that anthropological studies about the perinatal period and obstetrics in traditional rural settings report negative cultural representations of cesarean births. Cesarean birth is most often feared or refused because it demonstrates to the community the mother's inability to give birth/to know how to give birth (77). A woman from a city or somewhere else would not necessarily say the same thing.

3.9. Show that she has mothering skills and be careful not to quash them

A favorable outcome in the interactions between parents and babies is most likely when the parents' skills are underlined rather than criticized (3). This is especially important when migration has damaged these feelings of competence, for the reasons cited in part l. The logic of mixing ways of doing something or welcoming other ways of doing it requires the professional to have the tact to not impair these feelings. This requires pointing out where the mother can genuinely feel competent, but also recognizing the difficulties that others would also legitimately have, by contextualizing the situation.

- Avoiding cultural misunderstandings with a traditional Bambara woman from Mali

> *Aminata is a young Bambara mother from Mali, referred for a transcultural consultation by the staff from the maternal center[5] that initially sheltered her and her baby. At the time of our evaluation, the baby, Ramata, was a year old and she had been placed in a foster family. Some developmental delay was beginning to be noticeable. Ramata was growing up a different language environment than that of her mother, who had arrived in France soon before her birth and does not speak French. Aminata reported that her daughter was eating better when she was not present, partaking of little jars of baby food, whose composition was unknown to Aminata.*
>
> *Aminata described in a consultation her experience of the birth of this very preterm baby. The meeting with this baby arrived too early and was difficult; she was a stranger to her. Aminata had no representation of preterm birth/did not understand what preterm birth was and seemed stunned by the situation. Added to that were her language barrier and a life path that seemed studded with multiple traumas. Under these circumstances and the various misunderstandings between Aminata and the different teams caring for her, several professionals suspected she had cognitive difficulties. This led to a request for a transcultural consultation.*
>
> *The often chafing cultural distance was a source of misunderstandings on several occasions, in particular on the subject of protecting the*

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[4] Closure of the vulvar orifice during excision.

[5] Define maternal center.

child. When meetings with Aminata resumed in a larger space and in Bambara, her native language, this young mother's explanations made sense and seemed legitimate. For example, her representations of foster care enabled Aminata to accept assistance from the child protection services easily without understanding that she would not be able to control how frequently she saw the baby. Ramata is growing up in a cultural milieu different from her mother's, a fact that deepens the latter's feeling that her child is strange/foreign and incomprehensible. To feed her child well, as s/he did not gain (but did not lose) weight at least during the winter illnesses, she fed her/him as much as possible, but did not succeed in following the hospital's dietary prescription. She was also very anxious when the baby physically vulnerable because of his preterm birth, had a fever, because in her country, people see doctors for a fever, which they associate with a serious disease. She relied on the help of professionals during this period, but without any feedback from them. They were unaware of how worried Aminata was, and of how she felt progressively dispossessed of her maternal role.

The staff at the center interpreted the difficulty in their meetings to be due to a poor affective investment in the maternal relationship and her difficulty in bonding with Ramata. But did this not result from a separation of mother and baby that occurred too early? The staff's difficulty in decentering and their worries led the team to read her behavior through a prism that impaired her feeling of maternal competence. The misunderstandings concerned how she carried the baby, which they described as too "tonic", her failure to attach the baby to the stroller, his sleeping in her bed … Also some of the naive unfiltered questions she asked, due to her level of trust in these professionals: such as her mention of excision which she was not planning to do because it is forbidden in France, but an issue she wanted to raise related to including her child as part of her extended family, as her aunt was a blacksmith and exciser… In this situation, we shared an exercise in decentering with the team. Everything described here was normal in Aminata's culture. There are no cradles in African villages Babies are carried proximally. Is it Aminata who is wrong, or is it that we do not understand the complexity to which we are exposing her with our tools, such as cradles? How can we ask her to decipher prescriptions that would be complex for us as well when she cannot read? How can we make her understand the concept of contextual danger? If we say, "no, not like that, it's dangerous," the harsh word erases all of her maternal skills.

Moreover, what is dangerous here is not necessarily dangerous elsewhere in another context.

We need to ask ourselves how we can explain what seems to be inappropriate without erasing her maternal competence. In exploring in detail Aminata's pathway through her narrative facilitated by the transcultural levers described above, we understand that Aminata comes from the countryside, that she is isolated and very far from her points of reference, but also from our points of reference as professionals.

4. Discussion: how can cultural misunderstandings be resolved?

All of these postures and interview themes must allow an evaluation of the situation and a detailed understanding, while simultaneously avoiding misunderstandings and starting therapeutic work from the outset by a narration/story. Every stage requires a therapist's demeanor and knowledge of clinical practice and of the complex journey of migrants.

4.1. Professionals must take their cultural and trauma-related countertransference into account

Cultural countertransference (78) is the set of emotions, thoughts, and attitudes felt by the care provider toward the patient, related to the cultural similarities and differences of each. Professionals are influenced by their own identity, history, and knowledge, but also their prejudices and the conventional wisdom that constitutes implicit bias (79): their religion, education, life habits, name, etc.

The analysis and consideration of these attitudes of professionals supporting migrants in educational, therapeutic, academic, or legal settings are essential in transcultural work. These attitudes are objectified only by third-party references (supervisors, institutions, teams, theoretical references, etc.) (14).

When developed, the question of the cultural countertransference most often allows cultural misunderstandings to be resolved. Thus, team discussions make it possible to discriminate the portion of subjectivity, and to take the woman's own cultural representation into better account. Of course, the impact on interactions of postpartum depression, which is also more frequent in the migrant population, must not be underestimated. The point is to not induce pathological doubt in situations that do not fall in this category, and this requires reciprocal learning about cultural representations. To detect these attitudes, we propose that therapists engage in/commit to a process of decentering in relation to their own cultural affiliations. Clinicians must participate in the experience of cultural contacts, all the while creating methods to promote reflexivity concerning the intercultural dynamics that unfold during these contacts (14). The clinician's cultural subjectivity can be revealed by debriefing at the end of the consultation between the different therapists involved in it.

In situations regularly encountered with migrants (especially those who have traversed conflicts or wars and/or been subjected to trauma during their voyage, often intentional and repetitive), a traumatic dimension of countertransference can provide information about the impact of this trauma. Attention must also be paid to the risk of vicarious trauma (80) that can engender stupefaction, a blank mind. You do not have to tell yourself that this story is impossible because it is beyond comprehension; on the contrary, it may well signify real traumatic events. Moreover, psychotrauma can cause memory disorders and confusion in the patient's narrative: the description of dates and places can be complicated or inconsistent (81). Thus, knowledge of migrant journeys and elements of international politics/policy can be useful to therapists to avoid this pitfall of incredulity due to the unthinkable. Trauma implies precisely a form of insanity, in that it is defined by an experience of brutal and sudden confrontation with the reality of death (our own or that of others), stripped of all mediation of the signifying system that in ordinary life preserves the subject from the unexpected apprehension of nothingness (82).

While the patient's own rhythm must be respected in managing psychotrauma, professionals must not be inhibited in their approach to some elements if its clinical consequence or functional discomfort is important. Sometimes, avoidance-type counter-transference reactions by care providers who are not psychologists or psychiatrists also protect against not knowing how to use a traumatic narrative/story for therapeutic purposes/what to do with it on a therapeutic level. Subjects such as excision or children conceived by rape, for example, can cause blockages because of a feeling of lack of know-how due to the cultural distance (71).

Finally, some studies show that the analysis of therapists' countertransference allows a glimpse of what the babies may perceive through the mirror of their mothers (83), especially in traumatic dissociation.

4.2. Have detailed knowledge of the clinical aspects/presentation of complex trauma and of the cultural coding of symptoms of depression and anxiety

Numerous mothers and future mother present complex migration journeys, spattered with repeated traumatic events. The disorders observed often fit the clinical picture of a complex posttraumatic stress disorder (complex PTSD). Initially described by Herman (84), it is currently included in ICD 11 (11th revision of the World Health Organization's International Classification of Diseases, that is, WHO ICD-11). Herman stressed the dissociative symptoms, the somatic manifestations, and the negative changes in self-perception (guilt, shame, modification of identity). Suspicion/distrust accompanied by impairment/changes of systems of meaning (despair and loss of fundamental beliefs) can impair relationships with others, including professionals.

The clinical picture of migrants is sometimes atypical and far from the classic reactivation syndrome. Profound and lasting personality changes have been described. Some symptoms associated with distrust may be interpreted as delusions of persecution, which would generate diagnostic errors (ref). Because of other conceptual models of *fright* in some societies, patients can make it into a narrative with spirits and ancestors, with coded cultural events, or represent themselves as singular beings who have overcome recurrent trauma.

Similarly, the experience of depression varies between cultures, negative affects can present as guilt feelings more prevalently in Western cultures and as feelings of shame or persecution more often in so-called "traditional" societies (85). Somatic symptoms can also be in the foreground in the expression of psychological distress (86), such as massive anxiety or depression.

In some situations of isolation, the absence of local peers can jeopardize the accuracy of this diagnostic evaluation. Active creativity is then called for. Because of the stakes of a diagnosis and from an ethical perspective: get information from an interpreter, agree to see someone close to the extended family or designated by the patient, even though they are not part of the family, make telephone calls to the country of origin… And especially do not turn too rapidly toward a stigmatizing diagnosis and a potentially inappropriate treatment.

4.3. Alliance and refusal of care

Most of the requests from hospital teams due to a refusal of care for apparent cultural issues involve a way of reclaiming the performative character of what is spoken, that is, ensuring that the patient's discourse is heard and considered before any radical refusal is made.

Giving them a way to express their cultural representations can allow families to avoid western medical practices sometimes experienced as violent or immodest (delivery position, discarding the placenta, which can represents the baby's real double, showing a photo of a dead fetus …). We can encourage respect for traditional means of protection, which are most often compatible with the medical care needed. Working on maternal competence assumes/presupposes taking into account their know-how about the rhythm of breastfeeding, solid food introduction with food from their native country, the importance of playing with the child, and how to raise a child or make him strong. Adopting a moralizing position risks breaking one's bond with these patients, or even of undermining still more their sense of competence, already eroded by facing such different ways of doing things. The refusal of the cesarean described above is a good example of this. Among less extreme situations, let us mention the example of kaoline ingestion, frequent in West Africa: You can always inform women of the risk of anemia due to this consumption, consumption important to some women, because it is a common social practice in pregnancy. A refusal would most often reveal experience with a lack of consideration for what they have to say and with the other's subjectivity. Negotiations are most often possible if they are explained after understanding the performative character of the other's word. When a narrative can be developed, we often find an argument distant from the initial reason for refusal, more often associated with a singular aspect of the patient's individual or family history. Identifying where the reluctance really comes from makes it possible to work better and overcome fears (Table 1).

5. Conclusion

This article aims to help care providers to acquire/adopt transcultural skills, that is, an overall set of aptitudes and attitudes that allow them access to essential elements of their patient's cultural understanding and that will help their patient to create bridges between their health care customs "back there" and here.

Transcultural clinical practices often ask us to be creative and to abandon the rigidity of some standard frameworks of care. It implies for the clinician, to adjust his overall approach, including his attitude, his health examination, his therapeutic framework, in order to access the patient's cultural behaviors and mindsets. It also involves to overcome many relationships obstacles and to reduce the cultural distance which can both impact the therapeutic and diagnostic approach. Indeed, flexibility from health care workers promotes a thorough diagnostic evaluation and therefore a more effective therapeutic process, since the patient feeling more at ease and understood may be more inclined to share his distress, which can be a therapeutic process in itself by unlocking his normal thinking skills frozen by language barrier and cultural distance.

The transcultural approach is also a dynamic method that implies actively seeking levels of understanding, a general

TABLE 1 Summary: start working on cultural understanding early in the perinatal period- recommendations for health care workers (family doctors, psychologists, child psychiatrists, midwives, pediatricians, specialized child-care attendants, and social workers).

	Interview themes	Examples of content or questions
1	Start the interview by creating the basis for an appropriate relationship	Hospitality (Warmth and kindness) in the welcome Query the patient's cultural identity with precision Arrange for an interpreter as soon as necessary Explain clearly the professional's role and treatment objectives
2	Mitigate migration cleavage	Weave bridges between life there and life here Reestablish levels of continuity between here and there (religion, community, associative sectors)
3	Reconstruct the networks of belonging	What family remained there? Who is in France? How are they linked now (by telephone? how often?)? Did you announce the birth to them? Do you have friends, people from your cultural community nearby? Is the parent able sometimes to talk in her native language?
4	Help her to escape a feeling of powerlessness and facilitate her psychological reinvigoration	Help her capacities for resilience to emerge Show the heroic valence of the migration journey Help to coconstruct solutions: Even though she cannot do exactly as she would there, she can, for example, to perform a part of the ritual there. Or, do it here; it is equally functional even if it is not the day initially required.
5	Promote harmonious cultural métissage	Help her construct herself between the two cultural worlds she belongs to and take the creative richness of both (without it seeming inconciliable)
6	Construct a narrative of filiation	Historicize. include the child (or the future child) in the family history Ask about the baby's name, for example (Co-)construct a genogram
7	Put the family in the position of the expert: Let them express themselves, listen to them fully	What is done in your cultural groups to welcome a child? What are the rituals around birth?
8	Elaborate a meaning	State the problem: How would you name it in your country of origin? Is there a related/relevant cultural therapeutic logic?
9	Stress the parents' competence	Give yourself the means/resources (time, interpreter, team meetings) to avoid cultural misunderstandings. Welcome other ways of doing things Reinforce the parents' feelings of competence

culture—learning from our patients and getting help from colleagues. It also supposes a form of sincere opening to the other's culture and self-exposure, even while remaining professional. Every health care professional working in a cross-cultural world should not be scared of approaching cultural questions, by using an interpreter in situations where a language barrier exists, or by developing their openness and decentering skills. It will undoubtedly benefit the therapeutic alliance with the patient, particularly in the field of perinatal care in which cultural representations and native language play a crucial role.

Ethics statement

Ethics approval was given by The "Comité d'Evaluation de l'Ethique des projets de Recherche Biomédicale (CEERB) Paris Nord" (Institutional Review Board (IRB) of Paris NorthHospitals, Paris 7 University, APHP). All participants were adult and provided written informed consent to participate in the study. Written informed consent for publication of any potentially identifiable data was obtained from all participants.

Author contributions

RR, HC, JR, and MM made substantial contributions to the conception or design of the work, or the acquisition, analysis, or interpretation of data for the work. MB, HC, and AO drafted the work or revised it critically for important intellectual content. MR provided approval for publication of the content. All authors agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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Migrant mothers' experiences of Caesarean section: a transcultural qualitative study

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Objectives: Migrant women face an increased risk of adverse obstetrical outcome and Caesarean section. The psychological experience of Caesarean section combines physiological, social, and cultural dimensions. This qualitative study explores the subjective experiences of first-generation migrant women who gave birth by Caesarean section.

Methods: Seven qualitative, semi-directed interviews were conducted from January to March 2022 in a Paris maternity hospital, with women in their postpartum period who had had a scheduled or emergency Caesarean section and straightforward obstetrical outcomes. The presence of an interpreter-mediator was systematically offered. Thematic analysis of the interviews was carried out following the Interpretative Phenomenological Analysis (IPA) methodology.

Results: Four themes were identified in the thematic analysis relating to the women's experiences of Caesarean section: (1) The shock of the intervention combines disappointment, fear and early separation from the baby, (2) Pregnancy and delivery far from one's family aggravates the psychological suffering caused by isolation and loneliness related to migration, (3) The lack of cultural representations of Caesarean section leads to negative preconceptions and hinders mental preparation, in contrast with traditional or medicalised childbirth, and (4) The women's experiences of the medical follow-up highlights the importance of the continuity of care.

Discussion and conclusion: Caesarean section, which is a physical break, re-enacts the symbolic break (cultural, social, familial) that follows on from emigration. Improvements in care include the need for a better preparation for Caesarean section, active efforts for care continuity, and the development of early prevention interviews and groups in maternity units.

KEYWORDS

Caesarean section (C.S), emigrants and immigrants, migrant women, qualitative research and analysis, transcultural studies, obstetrics, maternity care

Background and introduction

Migrant women have a greater likelihood of risk-prone pregnancies, perinatal complications, premature childbirth, or Caesarean section (1–4). Higher rates of Caesarean section are observed among migrant women in large-scale studies. One Norwegian study in 2000, in an analysis of 17,000 births to non-European migrant women, found a prevalence of Caesarean section reaching 25%, while the average percentage for Norwegian women is 12%. Disparities according to cultural and geographical origin are considerable, with particularly high rates for women from the Philippines (25.8%) Brazil and Chile (24.3%) India and Sri Lanka (21.3%) and countries in the Horn of Africa (Somalia, Eritrea, Ethiopia) (20.5%) (5). A meta-analysis in 2013 found similar results from a synthesis of studies performed in 18 Western countries (Europe and North America). Among the 70 studies included, 69% evidenced different rates of Caesarean section between migrants and non-migrants. In particular, the rates are regularly higher among women from Sub-Saharan Africa, Somalia, and Southern Asia. In addition to this, the rates for emergency Caesarean section are higher among North African women, women from Western Asia, and Latin America. In the Paris region, a study by Linard using data from 2000–2002 found a proportion of 17% for Caesarean section among French women, versus 31% for women from Sub-Saharan Africa (6).

The factors suggested to explain this excess risk among migrant women are numerous. They include language and communication barriers, low socio-economic status, frail health of the mothers, high BMI, disproportion between uterus and foetus, previous uterine scarring, inadequate prenatal care, representations of pregnancy follow-up different from those of the country of origin, and isolation (1, 6–10). The precarious situations associated with migration are a considerable perinatal risk factor, as is isolation and absence of marital support, administrative status, and the existence or otherwise of health cover (3, 10). Command of the French language also appears a determining factor in the instatement of adequate pregnancy follow-up. Women who can neither read nor write French have a risk multiplied by three of not having the recommended pregnancy follow-up (10). For migrant mothers, fears associated with Caesarean section, often life-threatening in the country of origin, can combine with the experience of a cultural, religious or sexual transgression, and thus further compound the difficulties encountered for these parents starting a family far from their cultural landmarks and the support of their families (11). Anthropological studies on the perinatal period and obstetrics in traditional rural areas report negative cultural representations of Caesarean section (12–20). Women from Sub-Saharan Africa, West Africa or the Maghreb who have a Caesarean section run the risk of rejection by their husbands, or being branded as sterile, and are liable to be excluded from the social group if they can no longer bear children, or if they are held responsible for malformations in the baby (15, 16, 21, 22). Refusal to consent to Caesarean section is a frequent source of difficulties for caregivers in obstetric units, who argue compliance with medical decisions and the absolute need to save the lives of the mother and child, in the face of people who prefer to put their trust in divine will and who refuse the mutilation of Caesarean section (23). The intervention is most often dreaded or refused, since it signals to the community the woman's inability to give birth (12, 13).

Objectives of the study

This qualitative study aimed to explore the experiences of migrant women who have had a child by Caesarean section, so as to suggest lines for improvement in terms of psychological prevention and care provision for these vulnerable populations. The issue is approached from a transcultural and complementarist (24) point of view, so as to facilitate access to participants' inner experiences, while at the same time taking their anthropological and cultural belonging into account. This requirement has taken the form of an integration into the design of our interview guide of probe questions enabling women to express their cultural perceptions, and also of the availability of interpreters in the research interviews to enable participants to use their native language. The main aim of the research was to explore the subjective experience of women from diverse cultural origins who had given birth by Caesarean section. The secondary objective was to propose lines for improvement in terms of psychological prevention and mental care for this vulnerable population.

Materials and methods

Ethics approval

The research protocol was reviewed and approved by the Paris-Cité University ethics committee in 2021 (ethics committee registration number n° IRB: 00012021–109). All participants were adults and provided written informed consent to the study.

Methods

This was a single-centre, exploratory, qualitative study. The participants were recruited in the Port-Royal maternity unit, Cochin Hospital (APHP) in Paris during their post-natal stay. They had no previous links with the research team. Inclusions took place from January to March 2022 in the following manner: after an interview with the team of midwives and ascertainment of their eligibility, one researcher (JR) visited the rooms of the patients who were eligible to propose a face-to-face interview. The information letter and the consent form were given to the patients, who were allowed a period of reflection before agreeing to participate (see Annexe 1). Then an appointment was made, usually for the day after inclusion, according to patient preference and the availability of the ISM (Inter Services Migrants) interpreters. The participants could be accompanied by a person of their choice for the interview. Recruitment continued until theoretical saturation of the data, i.e. when the inclusion of new participants reaches a point of redundancy and all perspectives appear to have been explored (25, 26).

The inclusion criteria were as follows:

- Being a first-generation migrant mother, however long she has been in France
- Having recently given birth by Caesarean section, with straightforward sequellae for mother and child
- Caesarean section performed in emergency or planned.

The exclusion criteria were as follows:

- Mothers with a decompensated acute psychiatric disorder
- Mothers of new-born infants with serious neo-natal complications
- Mothers with serious obstetrical complications.

No financial remuneration was offered for participation in this research. The collection of informed consent and the perusal of the information letter were repeated before the interview and the data collection, with the help of the interpreter. An interpreter-mediator was systematically called on for the research interviews if the participant's mother tongue was not French, so as to facilitate access to the cultural concepts and representations of the participant, or to detail complex emotions is their own language. The participants' viewpoints were collected using a semi-structured interview guide specifically designed for this research. The interview guide included a limited number of open probe questions enabling the conversation to start on the research themes, while at the same time attempting to foster an interview that was as fluid and spontaneous as possible (Table 1). The duration of the interview was adapted to the state of fatigue of the participants, who were informed of the possibility of discontinuing the interview at any time. In case of manifestations of mental suffering in the course of the interviews, participants were asked if they consented to the transmission of this information to the health-care team in the maternity follow-up unit with a view to instatement of psychological care.

Data analysis

The interviews were recorded on a secure recorder in anonymous form, and were transcribed word-for-word. The analysis was performed using Nvivo (QSR International) qualitative analysis software. The analysis implemented Interpretative Phenomenological Analysis (IPA), which aims to provide an in-depth exploration of the ways in which subjects experience and give meaning to a phenomenon, from the study of their narratives on the subject (27, 28). The original interviews were coded line by line in full, using notations that were as close as possible to the original. Each unit of meaning (sentence, paragraph, verbatim) was translated into primary notations, and these were then grouped in codes, which in turn were grouped in themes and meta-themes. The thematic analysis grid was derived from the categories developed in inductive manner from the themes obtained. The analysis of the interviews was conducted by three researchers (JR, RR, MRM) (triangulation to ensure concordance between the analyses and the results).

Results

1. Description of the sample

TABLE 1 Interview guide.

1. You recently gave birth. How are you feeling today?
2. How did your pregnancy go? Can you tell me how things went and who accompanied and assisted you, medically or non-medically?
3. How did your close and broader family accompany you in this period of pregnancy and childbirth??
4. There are many ways to bring a child into the world. In your family, how is childbirth viewed? What were your own thoughts and expectations for your delivery?
5. Can you tell me about the delivery of your child? How was the decision to perform Caesarean section reached?
6. Tell me about your first encounter with your child. How did the first few days go?
7. What accompaniment did you have by professionals in the maternity ward following the Caesarean section?
8. Do you have anything to add about your experience of Caesarean section?

We interviewed seven women of various cultural origins: Australia, Spain, the Gambia (Soninke), Senegal (Wolof), Ivory Coast, Cameroon and Mali (Bambara). They had been in France for durations of 1 month to 5 years. The interviews were conducted from day 2 to day 4 after childbirth. Four of the Caesarean deliveries were emergency interventions, among which two followed on from a failed attempt to trigger labour. Two interviews were conducted without an interpreter, as none was available and the women were from African countries where French is commonly used. In line with our protocol, we interviewed only first-time migrant women.

Four of the seven participants were living with their partner and had their partner present during their stay in the maternity hospital. For those whose husbands were not present during the interview, this was due to situational factors (having to look after the eldest child, or the partner having gone to make the administrative declaration of birth) and not to a refusal on the part of the mother or to a lesser paternal investment.

As in any qualitative study, the small number of participants does not allow statistical conclusions to be drawn from the descriptive table of participants.

The interviews were conducted from January to March 2022 (Table 2). The duration of the interviews was 20 min to 1 hour, with an average of 53 min.

2. Thematic analysis

The thematic analysis detected 4 main themes articulating intimate, inter-subjective and cultural perceptions of Caesarean section with the experience of the care provided. We have named these themes (1) the shock of the Caesarean, (2) isolation and loneliness, (3) the absence of a cultural containing function, and (4) experiences of the medical follow-up.

The verbatims quoted as examples are intended to illustrate the themes and to help the understanding of the reader. The themes are to be considered as reflecting the experience of all the participants, except when the contrary is explicitly stated.

The shock of the caesarean

The first theme in the results is related to the intimate experience of Caesarean delivery in a setting of surprise, disappointment and pain. Although this particular theme does not apply specifically to migrant women, exemplifying an experience probably common to all women subjected to this experience, its complexity is greater in the context of migration. In almost all the situations here, the participant was unprepared for Caesarean delivery, which was at odds with her idealised projections of childbirth. One participant had the impression of having been deprived of the "magic of childbirth", while another said she had not had the dreamed-of delivery. Almost all participants said they had hoped for a "natural (or "physiological") delivery", or for a "delivery without epidural analgesia".

> MO2 – It was an experience completely different from what I had imagined. I wanted to give birth very naturally, in a natural setting, without epidural or anything else.
>
> MO3 – I wanted a physiological delivery, I had a plan for the birth of the child, I wanted to cut the umbilical cord with delayed clamping.

TABLE 2 Participant characteristics.

	Native language /country of origin	Marital status	Arrive in France	Parity	Reason for Caesarean	Psychological follow-up for:	People present during the research interview
M01	English (Australia)	With a partner	2 months	G1P1- 1st Caesarean	Caesarean during labour	–	Partner
M02	Spanish (Spain)	With a partner	Several years	G1P1- 1st Caesarean	Caesarean planned before labour	–	Partner
M03	Soninké/ Spanish (Gambia/Spain)	With a partner	2 and a half years	G1P1- 1st Caesarean	Caesarean during labour	Depression	Alone
M04	Wolof (Senegal)	With a partner	5 years	G2P2- 2nd Caesarean	Emergency Caesarean before labour	Denial of pregnancy	Alone
M05	French (Ivory Coast)	Single	5 years	G1P1- 1st Caesarean	Planned Caesarean before labour	Isolation	Alone
M06	Bafia (Cameroon)	Single	1 month	G1P2-1st Caesarean, twins.	Caesarean during labour	Traumatic experiences	Alone
M07	Bambara (Mali)	Single	1 month	G3P3- 3rd Caesarean, the first 2 in Mali	Emergency Caesarean before labour	–	A female friend

Only one woman said that she had not formed a childbirth plan, attributing this to lack of time and mental availability to settle down and dream. The context of migration and an atmosphere of health uncertainties relating to Covid-19 hampered maternal reveries.

> MO1 - It seems to me that with all the changes in recent years I have in a way got into the habit of not anticipating things […] It's just that our lives have changed so much… moving to France was an enormous life change, a new country, so many challenges.

The announcement and the decision to perform a Caesarean appears as an unwelcome surprise, too sudden, and received with disappointment by the participants who had never envisaged this mode of delivery. The preparation sessions for childbirth were of no help in the situations encountered here in preparing for Caesarean delivery.

> MO2 – It was nothing like what I expected it to be. It was very sudden, all the information came too fast, something you are not expecting at all.
>
> P (MO2) It was rather surgical, in fact, rather cold and surgical.

The corollary of this disappointment is the experience of a sort of failure, with feelings of incompetence and inability to bear children, going even as far as the expression of intense guilt. Caesarean delivery in this case is seen as deviant compared to what was systematically referred to as “normal” delivery.

> MO5 - I failed…
>
> MO2 - It made me feel like a bad mother, and a bad woman because I couldn't manage it properly - why? Did I do something wrong? Why can't I give birth normally, like other women?

This feeling of failure is stressed by several women, who compare themselves to other women around them, and most often with their own mother. The reference to other women is a constant observation in the interviews, characterising delivery as the life event that places a woman in her lineage and her social group.

> MO6 - In my family I am the only one to have had a Caesarean. Otherwise, everyone else has had a vaginal delivery.

The participants were interviewed just after delivery. Their post-operative physical state occupies considerable space in their discourse. Thus fatigue, or even exhaustion, and also pain and worries about recovering from surgery are present in all the interviews. The participants talked of worries about the healing of the scar on the uterus.

> MO1 – Everyone looks at the scar, asks me about pain, how things are going. But it's true that we never talk about what's going on inside in any detail. How will it heal?

The procedures encountered in the operating theatre cause distress, fear and anxiety. The lack of privacy on account of the numbers of professionals present, and the accumulation of strong stimuli (noise, lights, milling people) makes the visit to the operating theatre an experience that is a sensory aggression.

> MO7 - There they are very quick, there are too many people milling around! There are nurses, lights, bing! bang! You don't know what's going on. It's too much, far too much!
>
> MO4 – When I saw the people with masks, saying "It's alright, don't worry" I was flustered, I was in tears

Caesarean birth is sudden and violent, occurring in an immediacy that overturns psychic temporality of the mother who is thus deprived of the period of labour.

> MO2 – We were talking, and the moment we stopped talking they said "there we are, the baby is coming now" and a few seconds later we could hear him cry.

If we look at the words used to describe their Caesarean we note the words "hole", "big cut" or "they cut your belly open". Certain participants' narratives are in very lurid language, or else they give a wealth of details, which suggests that our interviews in the early aftermath had not left time for the experience to be integrated psychically. These raw narratives, without hierarchy in the detail, reflect the violence of the experience, and result from the great psychic vulnerability in which these mothers found themselves at the time of the interviews. They also provide insight into the traumatic potential of Caesarean section. The narratives are characterised by the lexical field of trauma, with the mention of a suspension of time, along with imposed passivity, giving the impression that the woman is placed in the position of undergoing (or enduring) her delivery, no longer an active party.

> MO4 – I went all day without eating. There were emergencies, they told me to wait in the other room. And I stayed there waiting till 4 in the morning. It was really hard, I was in pain […] Several times I asked for a drink of water and they said no, I wasn't to eat or drink … so I went more than 24 hours without eating, waiting for my turn.
>
> MO2 – It's as if someone said to you "You are going to die on such and such a day, you have two weeks, 18 days […] You can't compare waiting to die and having a baby, but there…

Caesarean section means an early separation of the mother from her baby, because of the time required to suture the wound and the time in post-operative care. This initial time of separation, lasting several hours, can cause a feeling of abandonment and loneliness for the mother, and the impression of a sudden switch to care of the baby rather than herself. The mother feels alone, abandoned in the recovery room and suddenly no longer the centre of attention of the healthcare team.

> MO1 – For me it was the recovery room that was the hardest. I was alone, unfortunately the nurse was not very friendly. I really felt as if I was alone on that floor. There really was no-one around […] I was in a sort of void, it's true. I was no longer the patient, and I was not yet the patient. Because, of course, it was the baby who mattered most […] I felt rather abandoned, in fact, once the baby was taken away.

Pregnancy and childbirth far from one's own people: an itinerary characterised by isolation and loneliness

Isolation, loneliness and mental distress

The women we interviewed were all suffering intensely from isolation as a result of migration. For those who had maintained contacts with their families, distant family support takes the form of frequent calls and exchanges, mostly several times a day, with the family remaining in the country of origin. When questioned about their experiences of pregnancy the participants all described a pregnancy that was difficult and stressful, with considerable vomiting throughout, the threat of premature birth, or mistaken announcements of miscarriage, and these ordeals were compounded by the pain of separation from the family.

> MO3 – In fact it's because I had the risk of a miscarriage. Very early on, at 15 weeks, and then a threatened delivery at 23 weeks. As a result I had to stay in bed, and in fact, as I'm not from here, well my family wasn't there either. So I spent my whole pregnancy alone with my husband.

This feeling of isolation leads to worries about what will happen after the Caesarean and the return home, and this is particularly marked for women with no companion.

> MO5 – I just know a few people here and there, but they can't be there for you all the time when you need them. Here everyone has his own occupations, people go to work… You can't call them and ask "can you come and help me".

One of the participants has a history of post-partum depression at the time of an earlier Caesarean delivery, which she links to her loneliness and solitude.

> MO7 – When I had my second son I had post partum depression, the real thing! Because overnight, I go to hospital, I have the baby, I come home, and I am alone! I don't even know how to bath the baby, I don't know how to do anything, I was shaking, it was horrible! Horrible – loneliness is … dreadful.

Solitude and distancing from the family as a result of migration were compounded by the closure of borders with the Covid-19 pandemic, and also by lockdown measures. Pregnant migrant women found themselves particularly vulnerable, in a situation of total dependence towards their husbands, who, when present, were their only resource.

> MO3 – Suddenly, no longer going to work, not being able to do anything, not being able to get out, I realised that apart from my husband I wasn't going to be able to do anything, or see anyone.

The husband, a needed but insufficient support

The husband, when present, has a major role in the pregnancy trajectory of these migrant women. Caesarean delivery imposes considerable requirements on the husband, since the mother in the recovery room is not available for prolonged skin-to-skin with the new-born baby, nor for the first baby care procedures. The two fathers who were present in the interviews (MO1 and M02) were particularly committed fathers, anxious to be present throughout their wife's time in the maternity unit. All the other participants were isolated and needed to care for the baby on their own. For them, the sadness of being alone, and also the worry of being temporarily incapacitated by the Caesarean and not physically fit to look after the new-born child, were particularly marked.

> MO5 – Its the time after the Caesarean [that worries me], because I have practically no-one here. How will I manage? Who will help me?

The mother, an omnipresent figure, is cruelly missed

The figure of their own mother appears omnipresent in the discourse of the participants, they miss her cruelly. The participants call on their mothers to accompany them from a distance, to reassure them.

> MO1 – My mother is a great person. I called her at one in the morning on three consecutive nights. In the depth of the night she told me "yes, yes I'm with you, it's nine in the morning here! Don't worry". That helped […] just knowing that someone was there. When it's night time you feel so alone […] so it does me good to be able to call my mother"

Several of our participants, especially those from Western African or Sub-Saharan countries express nostalgia for the maternal "portage" they would have had in their own country. The mother of the parturient woman has the role of providing psychological support, physical care and child care, to enable the young woman to regain strength.

> MO7 interpreter – Culturally, when the daughter gives birth she will spend one month with her mother, it depends on families, sometimes it's two weeks, sometimes 40 days.
>
> MO5 - So that you can eat well, you are given large portions of dishes at home. There you are given a big fish for you to recover!
>
> MO7 – You eat, you sleep, and you don't see your baby, they look after him.

In addition, the deliveries of their own mothers are systematically recalled by these women. The feeling of having had an abnormal experience is greater among the women whose mothers had vaginal deliveries, while those whose mother underwent Caesarean section identify more easily with that trajectory, which is welcome support.

> MO3 - For my mother too her pregnancies were complicated, she had a bad experience of the triggering of labour and Caesarean.

Trauma, secrecy, and disgrace

Migrants women can need to conceal their pregnancy from their families back home, which aggravates their isolation and loneliness in confronting these challenges. One of our participants (MO4) was initially in denial of her pregnancy and did not announce it to the family for fear of a negative, violent reaction from her father. For another participant, it was because of the worry of the disgrace she would bring on her family that made her prefer to keep her pregnancy secret.

> MO5 - If you aren't married, you can't have a baby… so I preferred not to tell them. As the father and I are no longer together, when things are that way, in the family it's – it's a sort of disgrace in the family. So I don't really want to talk about it, for that reason

For another of our participants (MO6) the pregnancy was the result of rape in the course of migration. The stunned state in which she found herself made the interview distressing, despite the participant's repeated request not to end it. The interview is punctuated by lengthy silences, and a virtual absence of spontaneous comments to restart the interview by the participant. Her obvious mental slowing, which could be attributed to both the fatigue from the recent Caesarean and to the presence of clear depressive symptoms, can also be seen as a state of dissociation and traumatic numbness.

The lack of a cultural containing function

A negative view of Caesarean section

Four of our participants from Sub-Saharan Africa reported negative cultural representations of Caesarean section. Caesarean delivery is perceived very negatively and there is heavy prejudice for the mother. Caesareans have a bad image first because they inspire fear: in countries with poor medical infrastructures the consequences of surgery – often fatal for earlier generations -are still feared. Caesarean delivery also means having fewer children, thus affecting the woman's fertility.

> MO4 – At that point I was afraid, because at home when we talk about Caesarean delivery, people are scared […] Because it was said that with a Caesarean you can only have three children, that's what they say in Senegal.

Beyond these worries of a medical nature, Caesarean delivery is viewed negatively as an absence of effort, preventing the woman from becoming "a real woman".

> MO7 – When I had the Caesarean, my grandmother said "As for you, you're just lazy, you couldn't even give birth! You had lots of help, didn't you! They open up your belly and they just give you the baby". For those women it's easy […] Yes, you should have vaginal delivery. That's it, you should… in fact you're not a real woman, not in their heads.

Caesarean delivery belongs to the medical world and makes traditional care impossible.

Caesarean delivery, by essence medicalised, calls into question the place and the presence of the medical profession in childbirth. For women from traditional societies, delivery should be natural and physiological, and excessive medical intervention is dreaded. One participant told about the experience of her own mother, who did not attend the appointment in the maternity unit for labour to be triggered.

> MO3 – In African culture, so much medical stuff is not really a good thing. For instance my mother, I know, for one of my brothers, they told her to come in so that could start the labour, and she didn't go, she waited till it started spontaneously.

The women systematically spoke of the traditional care provided for young parturient women having had a vaginal delivery to facilitate their recovery. This care is often described in detail, whether in terms of massages, the intake of certain plants or eating special foods suited to young mothers and favouring recovery. There is however no traditional care in case of Caesarean delivery. Caesareans belong to the world of medicine, and cuts women off from traditional care often provided by the mother or grandmother. This lack of cultural containing echoes with the loneliness and isolation mentioned earlier, which is compounded by the Caesarean delivery which excludes the woman from the traditional care needed for her full integration into the group of women and mothers.

> MO6 – I don't know about Caesarean, but with vaginal delivery when it's over you are cared for. Caesarean is a bit complicated. They would touch… it's still complicated. You shouldn't touch the belly, it should be left, you see…

Touch occupies an important place in the care of young parturient women. In intra-cultural situations, a woman who has not migrated and can receive the traditional care thus seems to deliver a second time with the expulsion of the "clots in the belly" (after-birth). Thus the delivery lasts several days, but in this case in the expert hands of women from the family. Everything then suggests that migrant women are dually distanced from their family group, by way of migration first of all, and by the medical profession *via* Caesarean section. The touch of another person, described as so important by the parturient women, is impossible, prevented.

> MO5 - When you have a vaginal delivery at home; you can be massaged, someone can massage your belly. It's the mother who does that, or an elderly person, with hot water. They say there are blood clots in the belly and they need to be expelled, so they massage you, they press on your belly, for several days.

The participants who have been able to share things with their family back home all have a particularly strong relationship with their mother, calling her on the phone several times a day and even in the night, and receiving a lot of advice and reassurance. While "our little hints" (MO4) can be used, the woman is nevertheless alone to massage her belly, or to carry out traditional care procedures or prepare special drinks to favour recovery.

> MO4 – My mother told me when I wake up in the morning to take a towel or a piece of cloth, dip it in hot water, and then put it on the wound and massage gently, not pressing too hard, but putting a little pressure, and the water should be quite warm even so. She also told me to take millet powder and make it into a porridge, to add palm oil and to drink it hot. That will clean out the belly, wash away the impurities, and all. That will help me have milk in my breasts so I can feed the baby.

In a setting of migration, threat to the rituals that welcome a baby

The importance of the rituals for the welcome of a baby are well known. They can take the form of physical, spiritual or symbolic care. The traditional practices intended to instate protection against evil spirits for the mother and her child can be difficult to set up in France. The advice proffered by the mother can contradict the rules imposed by the husband. For one participant in particular (MO5) this violent confrontation concerned traditional and cultural protection measures in contradiction with the religious requirements posed by the husband.

> MO4 – I tell my mother that I have done something, but I don't do it, because my husband does not want me to. Because he says to me "That's not written in the Koran, that's traditional stuff, not religious, so…" For him anything that is traditional and not religious, he doesn't do it

In addition, the lack of family and community support and "propping" in France can compromise the traditional rituals for welcoming the child. Among them, the Moslem baptism is practised in the first weeks after birth in Mali which is the country of origin of MO7. Even so, as a migrant mother having only recently arrived in France and with little access so far to other Malians, she does not envisage the baptism "here". It is a friend, also from Mali and present in the interviews, who acts as a mediator and proposes to introduce the mother and her baby to the local community, asking her own husband to organise the baptism.

> Friend of MO7 - In fact baptism within the week is compulsory. It's compulsory, it's religious, it's where you give the baby a first name, when the baby is shaved, they shave his hair. So we'll do it! Now I've seen it, I have my husband, they'll do it, it's compulsory. He'll do it.

The experience of medical follow-Up

Administrative difficulties and difficulties accessing care

Our participants have to cope with numerous administrative difficulties leading to difficulties in accessing care, this being true irrespective of socio-economic position or socio-professional category. Recently-arrived women all report difficulties in finding their way in the French healthcare system, notifying their pregnancy or initiating the monitoring of their pregnancy. They feel lost and in difficulty for access to midwives, gynaecologists and paediatricians in community practice to whom they are generally directed.

> MO1 - In fact I have an American friend who moved when she was eight months pregnant or thereabouts. She doesn't speak French, she has no Carte Vitale (personal identification for access to healthcare) of course, and that's difficult… When you are outside the system you are a bit lost.

These difficulties for access to health professionals in ambulatory care continue after childbirth, making the perspective of discharge with no outside support a worrying prospect. One participants expressed her anxiety about the imminent return home, although she had not been able to find a community midwife to visit her at home.

> MO3 – When I'm discharged, I need to find a midwife, as I had a Caesarean, a midwife who come to the home. But up to yesterday those that I have contacted are not available.

The language barrier is a considerable difficulty. For one of our participants, also a care professional, it is language that prevents the medical staff from providing medical information to the patients.

> MO3 – So they come here, they don't understand the language, nor the explanations. And several times at work I have seen that

> when the patients don't speak the language well, even the doctor gives fewer explanations because he knows she won't understand. It would be a good thing to always have interpreters in hospital.

Antenatal care, preparation for childbirth

There is a stark contrast within the interviews: while most of the participants are very satisfied generally with their stay in the maternity unit after the birth of the baby, the antenatal period is discontinuous and the sessions of preparation for childbirth appear unsuited to their situation. Indeed, all the participants mentioned their difficulty in finding their way in the French health system and finding a community midwife on their own to ensure follow-up of the pregnancy. They repeatedly mentioned not having been able to know who to turn to, having felt alone in organising the follow-up and having experienced considerable anxiety.

> MO3 – [In Spain] we have health centres where you find everything, you don't need to look for a midwife. You go there, make an appointment, you say you are pregnant and they organise everything. But here you have to look for a midwife yourself, or a paediatrician or a gynaecologist

Having friends and a network of acquaintances in France facilitates things. Almost counter-intuitively, our most vulnerable participants with regard to the administration and finance, housed in centres for migrants or social accommodation, were accompanied by the social services and were generally better supported in the follow-up of their pregnancy. Psychological care was also systematically offered, showing the considerable concern shown by staff for these difficult itineraries.

> MO5 – When I arrived at Port-Royal I was really well supported. I had the support of psychologists, social workers and a midwife. They were really there for me, they answered my questions and they listened to me. They did all they could. It really did me good, it brought me a lot.

Low-risk patients who were oriented towards externalised follow-up in ambulatory care were in contrast less satisfied with their prenatal surveillance. To the difficulties finding their way around the French system and in recruiting community health professionals can be added a feeling of discontinuity in the few appointments in the prenatal period in the maternity unit. When they meet different doctors and midwives at each consultation, the women are unable to share their concerns or worries. This lack of a stable reference is a central element that destabilises the participants, pregnant women and migrants already in a situation of vulnerability. The participants thus clearly expressed their need for caregiving continuity, not just referrals in medical files.

> MO2 – Sometimes you see doctors that are completely different, who have your file but that's all. I don't really feel I was properly followed, here, I don't feel really reassured with a person who you can really talk to and all that.

This absence of a frame of reference during pregnancy follow-up relates to a specific mode of organisation in the maternity unit at Port-Royal. We can however add that there are radical differences in care provision linked to differences in protocol between maternity units. One of our participants, who started her follow-up in another Paris maternity unit before transfer to Port-Royal reports a complete change in the mode of healthcare delivery.

> MO3 – The problem is that here they don't use at all the same protocol as the one I know. They placed a pessary which is like cervical cerclage, but here they don't use it and they removed it. It was a bit of a shock, because for me the pessary could help! It's a form of support! And I had medication, progesterone and all that, but they said that should go too. So it was a shock, right from the start, because they weren't doing the things I know about. I felt they weren't listening to me.

The sessions of preparation for childbirth appeared inadequate and unsuited to our participants who had ended up with a Caesarean. In all the interviews, we found this impression of sessions that had too little to say about the possibility of Caesarean delivery, and did not enable these women to anticipate this possibility. Sometimes, as for MO2, the preparation sessions were experienced as painful, entailing rivalry with other women, compounding their feelings of "abnormality" associated with Caesarean delivery.

> MO2 - I remember one day we were to prepare for childbirth with a community midwife, and we already knew we were programmed for Caesarean – having beside you couples who are saying things, like… "so we feel this and that" when I knew that I wouldn't feel anything at all…

Psychological care was offered to several participants during pregnancy, in the setting of pregnancy denial, depression sometimes compounded by isolation, or a history of trauma. This accompaniment proved to be particularly supportive for certain participants, while others had greater difficulty resorting to it, or regretted having been able to only broach very ordinary subjects in the sessions.

> MO3 – I talked with her, we got on well and all that, but she didn't go deep into my feelings. Sometimes I went there, we talked about everything and nothing, but not really about… the fact that I was sad, that I was crying, or things like that…

The delivery

The narratives of the participants mingle the difficulties linked to the Caesarean delivery, as seen in the first part of the results, and unanimous recognition of the professionalism and kindliness of the healthcare staff. No participant questioned the decision for Caesarean section despite their negative experience of it, and all agreed to the need for it. Likewise, none of them showed any anger or aggressiveness towards the maternity unit. The narratives are particularly positive and valorising, with the participants expressing their gratitude and satisfaction, stressing the kindliness of the staff and the quality of care in France.

> MO7 – They are so nice! I said to myself, these white people are so kind! […] If it was in Africa they would have been saying "Hey, you over there, get out! And who are you? Hurry up!"

However certain elements combine to make the experience of Caesarean delivery distressing. The premises, the organisation of the unit and the status of staff in the unit can be inadequately identified by the parents, compounding feelings of confusion generated by the decision to perform emergency Caesarean section.

> Father MO1 – At one moment they moved us to the day hospital, they took us to the pre-labour room or the birth room. There was a notice for a pre-labour room, but they had said birth room.

After the birth: the time in the maternity unit

The participants were unanimous in recognising the quality of the care provided by the maternity unit teams, describing the professionals as kind and supportive. The participants, and their husbands when present, expressed their satisfaction and even their gratitude.

> MO2 – From the moment we moved to the operating theatre, everyone was really professional, really reassuring and very kind.
>
> MO6 – They coddled me, they took care of me, they tried to see what was wrong. It's reassuring despite everything

The participants had a positive view of the staff visits to their room after the birth by staff they had seen before delivery, doctors, midwives or psychologists, stressing the importance of continuity in the therapeutic and human links with individuals who have been identified. This need for links could be exacerbated in setting of migration where links are often compromised.

> MO2 – She [the midwife who announced it] came to see me today, I was glad. To see how things were going
>
> MO5 - I have my psychologist, she came even yesterday, she keeps coming to see me, on Monday, yesterday […] Since the start of pregnancy, it has really supported me. She even told me, if I want, we can stay in contact, even if I see other psychologists out there, I can call her any time even so. She said so.

Discussion

One hypothesis can be derived from these results. The incision in the Caesarean procedure amplifies and updates the symbolic severance caused by migration. A migrant woman having had Caesarean delivery runs the risk of being excluded from the group of mothers for two reasons. Because of the isolation and loneliness linked to migration, first of all, she cannot be coddled and contained by the female accompaniment she would have had at home (this being independent from the modes and cultural variations of this accompaniment). Above all, it is the painful distancing from her own mother that makes the woman vulnerable, as the transmission of care gestures for parturient women (massages, food to regain strength) and for the care of the infant no longer occurs. These effects, frequently evidenced and well identified as vulnerability factors specific to migrant women in the perinatal period (29–31), are accentuated by the Caesarean. Caesarean section, a purely medical and surgical act, seen at least as having no cultural meaning when it is not viewed as a symbolic or even religious transgression (23), is also deprived of the appropriate care gestures that could provide this act with coherent symbolic and cultural meaning. And on top of that, the young parturient must avoid the traditional care and massages that could favour her recovery.

This study does not claim to be ethnological, nor does it describe how each culture perceives Caesarean section. Indeed, the participants were not interviewed as experts on their culture. The interview guide, while seeking to incorporate cultural elements, focused on the participants' subjective experiences; the cultural and familial elements that constitute their internalized cultural framework serving as a matrix for their subjective experiences. Thus, the thematic analysis aimed to identify factors of vulnerability linked to the transcultural situation and migration shared by all the participants.

These results casts light on the functions of childbirth and maternity which are specific neither to Caesarean delivery nor to migration. Here everything suggests that the transcultural context aligns with the situation of all women who are becoming mothers. How can one's integration into the group of adult women be envisaged if one has not been able to prove the ability to give birth? How can one learn to become a mother without the model and support of one's own mother? Migration, according to Claire Mestre, thus equates to "a quasi-experimental situation that enables us to explore how women need to call on different personal competences and do without the help of their mothers, in particular when they are from a society in which the place of the mother is essential for transmission" (30). Our results underline another important element: the atmosphere of uncertainty in which migrant women find themselves entails the risk of failure to invest fully in the birth of the child, a risk that then involves the child.

The parturient woman's mother occupies an essential place in the psychological elaboration of pregnancy, with the processes of identification, gratitude and idealisation [Bydlowski, quoted by 30]. The absence of this maternal figure can be partially compensated for by the adoption of a particularly maternal attitude by the husband, as seen in several narratives by our participants. This situation, frequent for migrant mothers distanced from their own mothers, (and more broadly for non-migrants suffering from isolation and loneliness) is accentuated by the physical injury of the Caesarean. The initial prolonged separation from the baby while the mother is in the recovery room, the first care procedures that the woman cannot perform – this makes the father particularly involved in the maternal care of the new-born infant. This is liable to accentuate the gulf with traditional maternal care in the home community and aggravate the mother's feelings of guilt and incompetence.

Our participants established a parallel between difficult experiences of pregnancy, different from what was expected, and the fact that the pregnancy ended up with a Caesarean. The narratives on these pregnancies show a predominance of the passive register (needing to rest, to stay in bed, vomiting, being restricted in activities, etc.). This passivity is also seen in the experience of Caesarean delivery, where the woman submits to the vicissitudes of protocol and medical decision. The distress relating to intrusion and upheaval linked to pregnancy also entails the necessary experience of passivity that the mother must endure to enable a new being to grow within her body (32). This theme of passivity, inherent in pregnancy and inherent in Caesarean delivery, echoes the situation of passivity to which the migrant woman is subjected, whereby she has to adapt, conform and accept an external cultural framework that differs from her interior cultural framework.

Indeed, the logics of healthcare, of life and death entertained by these women may not be in line with the issues and objectives of the medical care provision offered by healthcare teams here (23). This passivity resulting from both migrant status and pregnancy status is reinforced by Caesarean delivery, a painful episode that reinforces dependency towards caregivers and husbands, making the woman unable physically to take care of the baby on her own in the early days. In addition, the importance of pain during labour seems to be missed by certain women who feel they have been forced to be passive as a result of Caesarean delivery, losing control of their own delivery. The violent remarks by traditional grandmothers reported such as "you are just lazy" show the importance of the symbolic dimensions of distress, pain and suffering, seen as legitimising graduation to maternity.

Certain migrant women are particularly exposed to the risk of violence and traumatic events, as with one of our participants whose pregnancy was the result of rape during her travels. Some participants described their deliveries with a wealth of detail involving various sensory modes (visual, auditory or painful scenes) in some cases violent. These details, associated in counter-transference with an impression of diffuse unease, dejection or anger when listening to them, are challenging and of concern to us on account of the ultimate traumatic potential of the event (33, 34). Further to this, the literature outside the migratory setting identifies the risk of traumatic neurosis following a Caesarean delivery (35). We hypothesise that Caesarean delivery could compound the aftermath of the trauma of migration, in other words it could revive and update on the one hand real trauma experienced previously, and on the other the separations and bereavements of the past and the emptiness resulting from migration and distancing from one's own people. To be validated, this hypothesis requires a longitudinal exploration of a cohort of migrant women having had Caesarean deliveries. We can however also turn to research on trauma in transcultural settings, which can inform us on the modes of activation of experiences of traumatic migration following bodily alterations (36).

One result diverges from the literature and our expectations: none of the women expressed mistrust or contested medical decisions or care protocols. On the contrary, gratitude is unanimous, even when the feelings towards Caesarean delivery are negative. It can be hypothesised that the status of migrant fosters passive social and institutional attitudes (sometimes in extreme cases linked to the inability to understand and to be understood by caregivers). This interiorised passivity could generate narcissistic feelings of frailty and intense dependency towards caregivers and the institution, making expressions of discontent more hazardous.

It can be added that participants may have found it difficult to precisely identify the place of our research interviews in care provision in the institution. The participants indeed mentioned their difficulty in identifying and differentiating the different professionals in the unit, which leads us to think that visits to the patient's room by a researcher were possibly not sufficiently distinguished from the healthcare staff's visits, leading to possible confusion.

Proposals for the improvement of care

Pregnant migrant women need continuity and a healthcare reference in order to develop a feeling of containment throughout the pregnancy, from the start of follow-up until discharge from the maternity facility, and including the operating theatre. It appears fundamental to deploy efforts to reduce moments of isolation and loneliness inflicted on mothers finding themselves alone after their Caesarean, since these moments of isolation reactivate the woman's feelings of loneliness and abandonment. Likewise, organisational efforts should be deployed to restrict the period of separation between the mother and her baby after a Caesarean. As suggested by Goguikian Ratcliff (37), we also think it is important to search medical files for anamnestic data linked to vulnerability factors among these women, so that they can be taken into account by caregivers: time since arrival in the host country, fluency in the host country language, educational level and urban or rural origin.

All our participants felt insufficiently prepared for a Caesarean. Preparation for a Caesarean should be given more space in sessions preparing for childbirth, so as to reduce the experience of failure and comparisons with other women. Further to this, their own mothers have considerable importance for these migrant mothers. The presence of the patient's mother should be allowed and encouraged, wherever possible, to foster coherent cultural propping and support and to enable tradition care practices to be maintained. Care should be taken to avoid an ethno-centred stance allowing only the presence of the husband. In the absence of the mother or co-mothers, i.e. other women from the cultural group of origin (11, 38), caregivers, serving as substitute mothers, should endeavour to be "good enough" mothers. They can support motherhood by showing themselves to be sensitive to the cultural identity of these women.

Our results, and the excellent reception of our study by the participants, show that while the initial experience of Caesarean delivery is always difficult, migrant women draw considerable benefit from the opportunity to put words and give meaning to labour and childbirth, which is in favour of a re-appropriation and a return to an active posture after Caesarean delivery that has been experienced passively. We suggest the need to raise awareness among caregivers towards the difficulties of the post-natal period, and the need for systematic debriefing interviews on the birth of the child. In cases of emergency Caesarean, women draw considerable benefit from being able to see the team members present at the time.

In certain maternity units, there are groups of mothers which are open to mothers who have recently given birth. Given the frequency of Caesarean deliveries, it seems relevant to develop groups of women who have had a Caesarean, aiming to enable women to put their feelings of abnormality and guilt into perspective. There are also other systems, such as transcultural consultations, which, using a complementarist approach (Devereux) propose group consultations enabling an affective and cultural "envelope" to be formed for migrant patients with ambivalent representations of the practices encountered here and those of their home country (11, 31, 39).

Strengths and limitation of the present study

The interviews carried out with the presence of interpreters enabled women whose first language was not French to express their intimate thoughts in their native language. The presence of an interpreter, along with the interview guide specifically constructed to explore cultural perceptions of the subjects' experiences, were a major methodological concern, favouring the transcultural validity of the research.

The protocol, validated by the Ethics Committee, entails all due precautions to protect the participants in this very vulnerable period. Our objective was not to draw conclusions from experiences specific to one or other culture, but our results do cast light on vulnerabilities exacerbated by the difficulties of migration. Further to this, our exploratory methodology and the very early organisation of the interviews immediately after delivery was not suited to the exploration of phenomena that tend to appear in the later aftermath, such as traumatic neuroses.

A later study could include a larger number of mothers so as to have access to varied modes of care provision, or to explore the mental consequences at a distance from the event. Moreover, recent anthropological studies would be needed to collect cultural representations of C-section by cultural group, and their consequences on femininity and motherhood.

Conclusion

Caesarean section is a genuine public health challenge, since in France today one in five deliveries is a Caesarean delivery. Migrant women are particularly vulnerable in this respect. Caesarean birth is often experienced negatively and ill-prepared, and it accentuates the distress linked to isolation, feelings of loneliness and the difficulty of becoming a mother without the support of their own mother. The absence of a culturally coherent meaning allocated to Caesarean birth places the migrant women from traditional cultures in a position that devalorises them and causes feelings of guilt. We propose to deploy systematic interviews centred on the experience of Caesarean delivery in the post-natal period, as well as talking groups for women who have had a Caesarean. Migrant women need to be better prepared for this intervention in the course of sessions preparing for childbirth, involving the presence of interpreters. It is urgent to consider migrant women having had a Caesarean as dually vulnerable, and to actively accompany these women for a sufficiently long period before and after the intervention. Maternity unit professionals and mother and child protection centres should be made aware of these problems so that migrant women can develop protective strategies commensurate with their specific vulnerabilities.

Data availability statement

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author.

Ethics statement

The studies involving human participants were reviewed and approved by Comité d'Ethique de la Recherche de l'Université de Paris (CER U-Paris). The patients/participants provided their written informed consent to participate in this study. Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

Author contributions

JR, RR, and MM: conception of the study design, data analysis and interpretation, and final approval of the version to be published. JR: data collection and drafting the article. RR and MM: critical revision of the article. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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A 5 years' experience of a parent-baby day unit: impact on baby's development

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Introduction: Psychiatric Mother-Baby Units are well established in France, United Kingdom, and Australia, mostly in full-time hospitalization. Inpatient units are considered as best practice for improving outcomes for mothers and babies when the mother is experiencing severe mental illness and many studies have showed the effectiveness of care for the mother or the mother-infant relationship. Only a limited number of studies have focused on the day care setting or on the development of the baby. Our parent-baby day unit is the first day care unit in child psychiatry in Belgium. It offers specialized evaluation and therapeutic interventions focused on the baby and involves parents with mild or moderate psychiatric symptoms. The advantages of day care unit is to reduce the rupture with social and family living.

Aims: The objective of this study is to evaluate the effectiveness of parent-baby day unit in prevention of babies' developmental problems. First, we present the clinical characteristics of the population treated in the day-unit in comparison to the features presented in the literature review about mother-baby units, which usually receive full-time treatment. Then, we will identify the factors that might contribute to a positive evolution of the baby's development.

Materials and methods: In this study, we retrospectively analyze data of patients admitted between 2015 and 2020 in the day unit. Upon admission, the 3 pillars of perinatal care – babies, parents, and dyadic relationships – have systematically been investigated. All the families have received a standard perinatal medico-psycho-social anamnesis, including data on the pregnancy period. In this unit, all the babies are assessed at entry and at discharge using the diagnostic 0 to 5 scale, a clinical withdrawal risk, and a developmental assessment (Bayley). Parental psychopathology is assessed with the DSM5 diagnostic scale and the Edinburgh scale for depression. Parent–child interactions are categorized according to Axis II of the 0 to 5 scale. We have evaluated the improvement of children symptomatology, the child development and the mother–child relation between the entrance (T1) and the discharge (T2) and we have compared two groups of clinical situations: a group of patients with a successful evolution (considering baby's development and the alliance with the parents) and a group of unsuccessful evolution during hospitalization.

Statistical analysis: We use descriptive statistics to characterize our population. To compare the different groups of our cohort, we use the *T*-test and non-parametric tests for continue variables. For discrete variables, we used the Chi^2 test of Pearson.

Discussion: The clinical population of the day unit is comparable to the mother-baby units in terms of psychosocial fragility but the psychopathological profile of the parents entering the day unit shows more anxiety disorder and less post-partum psychosis. The babies' development quotient is in the average range at T1 and is maintained at T2. In the day unit, the number of symptoms as well as the relational withdrawal of the babies is reduced between T1 and T2. The quality of parent–child relationship is improved between T1 and T2. The children of the group of pejorative evolution had a lower developmental quotient at the T1 and an overrepresentation of traumatic life events.

Conclusion: These results indicate that parent-baby day unit lead to positive outcomes in clinical situations with anxio-depressive parents, relational withdrawal of the babies, functional problems of the babies but not when a significant impact on the development of the baby already exists. The results of this study can guide therapeutic approaches for the benefit of care in parent-baby day units, and improve the development of the child and of the dyadic relationships.

KEYWORDS

perinatality, mother-baby unit, parent-baby day unit, day treatment, child development, parent-child relation

Introduction

In western countries, perinatal mental disorders are associated with considerable maternal and fetal/child morbidity and mortality which remain one of the major problem for the child development and adaptability (1–3). Childhood toxic stress is defined as a severe, prolonged, or repetitive adversity, and a lack of the nurturance or support from a caregiver. It induces a disruption of the neuro-endocrine-immune response resulting in prolonged cortisol activation and a persistent inflammatory state. Children experiencing early life toxic stress are at risk of long-term adverse health effects including maladaptive coping skills, emotional, social and cognitive long-lasting effects, poor stress management, unhealthy lifestyles, mental illness, and physical disease (4–8). Some of the negative effects of early life toxic stress on child development are mediated by disruptions in the mother–child interaction, biological factors including cortisol secretion and epigenetic processes (3, 9–12).

Furthermore, in the context of early distorted parent–child interactions, number of studies have shown that treatment targeting only the mother is not sufficient to improve the development of the child (9, 13).

Additionally, the risk factors for parental psychiatric troubles are also risk factors for poor child development, suggesting that the relation between parental disorder and compromised early child development is multilevel and cumulative (14).

Regarding maternal depression, which is to date the most widely studied perinatal pathology, many authors conclude the need for care integrating as soon as possible the parent, the child and their emerging relationship to reduce the impact of cortisol secretion on the development of the child (15, 16).

Parent-baby units are an interesting perinatal care device recommended by the NICE (National institute for Health and Care Excellence) for women with acute postpartum mental disorders and their baby to facilitate mother-infant relationships (1). These units provide health care for mothers, care for the infant and sustain the mother-infant interactions (17). This type of unit has been established in several countries since the eighties (United Kingdom, France, and Australia) and more recently in New Zealand, Israel, India, Sri-Lanka, and in the US (18). In 2021 in France, following the publication of the "First 100 Days" report, additional budget was allocated for the opening of 10 new mother-baby units. This report also points out the difficulty to compare the different units, due to the heterogeneity of the existing devices (19, 20). First, the literature still refers to "mother-baby units," despite the fact that fathers are more frequently involved. Second, some parent-baby or mother-baby unit are day-night units, other are day units. Third, these units depend, based on their funding or their professional identity, either of adult psychiatry or of child psychiatry department.

Day units were created in parallel to day-night units (21). The choice of implementing a day structure has being often induced by economic necessity (22). These structures are positioned at an intermediate level for dyadic care, between outpatient care and full-time hospitalization. So, in case of parental psychopathology, it has to be stabilized and it should not compromise too much the parenting function at home.

Most studies on mother-baby units involve residential care setting. Very few have focused specifically on day units (18, 23–26). They describe the patient population and usually focus on the mother's clinical improvement (27–31) or on the parental satisfaction (32, 33). The mean maternal age at the time of admission ranges from 24 (34) to 33 years (28) with a mean child age ranging from 9 weeks (29) to 7 months (35). The average length of stay ranged from 7 days [US study by (36)] to 11 weeks (37). The most frequent diagnosis for the mothers were depressive disorder, schizophrenia, or other psychotic disorders.

The second focus of these studies concerns the parent–child relationship which is assessed through video observations, attachment scales, or assessments of parental competences. In the literature, a significant improvement in the parent–child bonding (30, 38, 39, 40) or in the feelings of parental competence (31) is often described.

Results are influenced by psychopathological and demographic factors (lower scores with a diagnosis of schizophrenia, personality disorder, lower social support or economic status) (29).

Few studies focus specifically on the well-being of the infants and on infant mental health (41). Other studies describe the symptomatology of the child at admission (29), the attachment type at discharge (42), or focus on the factors influencing the decision of child's placement after admission (43, 44). This specific aspect needs to be considered, as the largest European prospective longitudinal study (43) shows that 14.8% of children were separated from their mother at discharge from mother–child baby units. Different risk factors were brought forward: medical complications for the baby, severe psychiatric disorders for mothers, severe psychiatric disorders for fathers, bad social relationships of the mother, disability for the mothers and low-social economic status.

There is a lack of international consensus on how these units should be structured and equipped (19, 45). Early identification of potentially at risk situations with a low outcome for the baby is therefore a major challenge.

Our study aims at contributing to this debate. In comparison with the literature dealing with day-night units, we will describe the care trajectory of the population of a parent-baby day unit, and focus more specifically on the baby's status and on the parental psychopathology. The clinical evolution thanks to the day treatment, in terms of clinical improvement of the baby and the parent-baby relationship, will be evaluated at the time of discharge. The objective is to identify the specific variables related to a favorable or unfavorable outcome for the baby and to specify the clinical indications for day unit.

Methods

Description of the day unit

The Parent-Baby Day Hospitalization Unit (PBDH) opened its doors in 2015 within a tertiary pediatric hospital in Brussels. This innovative device is the first of its kind in the French-speaking part of Belgium. It is integrated in the child university psychiatry department and is in a great proximity with the pediatric department. It receives children from 0 to 2.5 years old, accompanied by their parents, on a part-time basis. The multidisciplinary team is composed of two child psychiatrists, a coordinator, two psychologists, a social worker, a psychomotrician, two pediatric nurses, three educators, two midwives and a secretary. A pediatrician and a psychiatrist are consultants at the request of the team.

The PBDH welcomes requests from parents or professionals regarding difficulties in the development of the child, functional disorders, difficulties in parenthood or in the establishment of early relationships. The capacity is 6 dyads/triads per day. Attendance can vary from 1 to 2 times per week. The Unit has a dual mandate of evaluation and therapeutic care. All admitted situations begin with an assessment period of 4 to 8 weeks.

The care is provided through group support and individual follow-up. During the day, different activities offer parental guidance, nursing assistance and a supportive environment for the child's development and the relationship with the parent. Therapeutic individual interventions are based on multiple models (attachment, systemic, psychodynamic, and behavioral theories). Practitioners always rely on the developmental needs of infants as the basis for their guidance and on mentalization work with parents (46). The use of video is an integral part of the treatment as it can support parent's reflection on their child and on their relationship with him/her. The unit tries to interact with the parents' network, and to systematically meet the close family in contact with the child.

Participants

All families admitted to the Parent-Baby Unit from its opening in 2015 to March 2020 were eligible for the study. Patients who attended the unit for only 1 day were excluded. All ethical measures regarding privacy, patient rights, and professional conduct were duly observed. This study was submitted to the HUDERF ethics committee, which gave its approval to start the study on 19/06/2020.

Data collection

Data were collected from the child psychiatric medical records of the hospitalized patients.

Hospitalizations resulting in a discharge plan jointly supported by the parents, the team and the network are considered as "successful." The hospitalization is considered "unsuccessful" when the child is placed or when developmental regression is observed (1 standard deviation below the mean for cognitive development between T1 and T2).

Tools

Child development and diagnosis

The Bayley Scale-III was used to **assess** child **development**. The *Bayley Scales of Infant and Toddler Development – BSID-III* assesses the development of young children between 1 and 42 months. The BSID-III is built around 5 scales: cognitive, language, motor, social–emotional, and behavioral. Only cognitive, language and motor scales were used here. The results for each scale are expressed as a composite score with a mean of 100 and a standard deviation of 15. We considered a score below 85 as a developmental delay.

The relational withdrawal was assessed with a clinical observation based on the ADBB (Alarm Distress Baby) scale developed by (47). The relational withdrawals were classified into 3 categories: no withdrawal, at risk of withdrawal and obvious withdrawal. Due to the low number of data collection, ADBB scores were not included in our study.

Regarding **the diagnostic assessment** of children, we have worked with the new edition of the Diagnostic Classification of Mental Health and Developmental Disorders in Early and Middle Childhood (DC: 0–5), (48). It is the updated and revised version of the DC: 0-3R classification.

Physical health of the child was evaluated by a pediatrician.

Parent–child interactions were evaluated using the Axis II (Relational Context) of the DC: 0–5 years old *scale (Zero to Three, 2005)* and more specifically the "Levels of adaptation of the parent–child relationship" scale. Four general levels of adjustment are described from level 1 (well-adjusted to satisfactory relationship) to level 4 (troubled to dangerous relationship). Parent–child interaction was assessed at T1 and T2 for each mother–child dyad.

Mental and physical health of the parent was considered either by the child psychiatrist in charge or by the consulting psychiatrist on the basis of the DSM V (49). Maternal depression was assessed using *The Edinburgh Postnatal Depression Scale (EPDS).* This scale is a 10-item self-administered questionnaire designed to assess the intensity of depressive symptoms experienced during the previous 7 days (50). A score of 12 or more is considered indicative of a risk of depression.

Statistical analysis

The data drawn from the sample of the patients are used for descriptive and frequency statistics. Then, for the comparison of the variables of the two groups (successful hospitalization versus unsuccessful), the statistical tests are adapted to the type of variable. For continuous variables, the underlying assumptions of the *T*-test are tested (homogeneity of variances using the Bartlett test of homogeneity of variances and normality of residuals using the Shapiro-Wilks test). If the underlying assumptions are met, a *T*-test is made and the mean ± standard deviation is presented. Finally, we use a non-parametric test to compare the groups: the Wilcoxon rank test (*median* and inter-quartile range [*Q25 – Q75*] are presented). For discrete variables, we do the comparison using Pearson's Chi^2 test. A value of p <0.05 is considered significant and < 0.001 is considered highly significant.

Results

Admission and socio-demographic data

Ninety-two situations were admitted at the PBDH between May 2015 and March 2020. Data related to admissions and socio-demographic characteristics of our population are presented in Tables 1, 2.

In the majority of cases, babies and their family were referred for therapeutic management (81.5%) while the remaining (18.5%) were referred for assessment.

The mean parental age at admission was 32.7 years for mothers and 36.3 years for fathers. The mean age of children at admission was 14.8 months. The age distribution of children at admission is bimodal, with two peaks of attendance, around 7.2 and 25.3 months. A bit more than half of the children are admitted after 12 months (52.1%). The average length of hospitalization is 29 weeks, or 6.65 months (ranges from 1 week to 24.8 months) (1.6 months for the evaluation group and 7.8 months for the therapy group), with an average family attendance rate of 69.6%.

The reasons for admission were analyzed from the perspective of the baby, the parents, and the relationship. Each of these poles can constitute the reason, separately or jointly, for referral to the UPBB. Regarding the child's reason for admission, there is a strong concern for the child himself (83%), the first reason being a developmental disorder (43.5%). Concerning the parent, in 77.2% of

TABLE 1 Admissions data.

	N	Mean (SD) or *N* (%)
Mission	92	
Evaluation group		18 (18.5)
Therapy group		74 (81.5)
Successful outcome		54 (73)
Unsuccessful outcome		20 (28)
Maternal age (years)		32.7 (6.3)
Paternal age (years)		36.3 (7.6)
Child age at admission (months)		14.8
0–6 months		31 (33.7)
6–12 months		12 (13)
12–24 months		26 (28.2)
>24 months		22 (23.9)
Child gender (percentage of girls)		40 (43.5)
Length of stay (week)		29
Evaluation group		7
Therapy group		34
Average frequentation	91	69.6
Reason for admission		
Concern for the child	92	83
None		9
Development delay		40 (43.5)
Withdrawal		16 (17.4)
Behavioral trouble		25 (27.2)
Somatic problem		2 (2.2)
Parent–child relational trouble	92	50 (54)
Parental difficulty	92	71 (77.2)
None		21 (22.8)
Parental skills		29 (31.5)
Acute psychiatric pathology		17 (18.5)
Chronic psychiatric pathology		25 (27.2)
Decision of the end of hospitalization (therapy group)		
Mutual decision		39
Parents		20
Medical staff		17
Partner support		
Involved in day care or appointments		18 (19.6)
Not present but supportive		27 (29.3)
Missing or lacking		47 (51.1)
Social or protectional service needed (yes)		30 (32.6)
Placement (yes)		16 (17.4)

the cases, a maternal problem is the reason for the admission. Finally, for slightly more than half of the dyads/triads (54.3%), the parent recognizes a relationship disorder at admission.

TABLE 2 Socio-demographic characteristics.

	N	Mean (SD) or *N* (%)
Mother cultural origins	92	
Belgian		23 (25)
Maghreb		43 (46.7)
Sub-Saharan Africa		11 (11.9)
Eastern Europe		10 (10.9)
Europe (rest)		1 (1.1)
Turquey		3 (4.2)
South America		1 (1.1)
Father cultural origins		
Belgian		17 (18)
Maghreb		46 (50)
Sub-Saharan Africa		13 (14)
Eastern Europe		8 (9)
Europe (rest)		2 (2)
Turquey		4 (4)
South America		1 (1)
Lifestyle	92	
Couple		63 (68.5)
Isolated		20 (21.7)
Extended family		6 (6.5)
Parental center		3 (3.2)
Paternal recognition (yes)	91	74 (81.3)
Socio-economic level (family)	89	
Low		46 (51.7)
Middle		39 (43.8)
High		5 (5.6)
Familial support		
No		33 (36)
Low		12 (13)
Enough		47 (51)
Parity		
1		43 (46.7)
2		21 (22.8)
3		17 (18.5)
4 or more		11 (12)
History of child placement in the family	48	9 (18.8)
History of intervention of social service	48	15 (31.2)
History of medical or psychological antecedent in the siblings	48	30 (62.5)

The majority of referrals were from intern second line health care (40.6%) and from extern second line health care (28.6%). A smaller number of patients came directly from the front line health care (16.4%), from social services related to youth care or protection (8.8%) or directly on the patient's initiative (5.5%). Nevertheless, 32.6% of the children had an open file with the youth care or youth protection (at the time of admission) and 8 children (8.7%) were in a placement situation (foster-care or intra-family).

The vast majority of admitted parents live together (68.8%). Most hosted families are of foreign cultural origin, with more than two languages spoken fluently at home (more than 75% in both cases), and half of them come from a disadvantaged socio-economic background. Only 3.4% of the mothers have an income as wage-earner, compared to 60% of the fathers.

Among families with more than one child, almost two-thirds have a history of medical or psychological follow-up for another child. One-fifth of families have had a court placement experience in their history, and one-third have had a social services intervention experience, whether it involves a parent or a child. 18.8% of the siblings had a history of placement and 31.2% had been followed by the youth protection services. The vast majority of families with more than a child (62.5%) had a psychological or somatic history for the children.

Concerning the involvement of fathers, he is absent half of the time (51.1%). He is not involved but supportive in 29.3% of cases and present in 19.6% of cases.

Pregnancy, neonatal, and child data

Pregnancy and Infant characteristics are presented in Table 3.

More than 25% of the children were born premature [statistical difference with the national average of 8% (51)] and just over a quarter of the children were born with low birth weight (26.1%). In comparison, the national low birth weight rate is 7.8% (value of $p = <0.001$) (CEPIP).

Concerning the antenatal period, two thirds reported stress factors during pregnancy and one third of the mothers had a history of miscarriage, a traumatic delivery and/or a hospitalization in the neonatal intensive care. One-fourth (23.3%) of the deliveries was made by cesarean section, this proportion is not significantly different from the Belgian national rate.

In terms of drug use, nearly one-fifth of the mothers smoked during their pregnancy, one-tenth used drugs and 6.5% were under psychiatric treatment.

Nearly half of the children admitted had a somatic or functional pathology at the time of admission, mainly gastroenterology pathology, sleep disorder (20%), and eating disorder (15%).

The diagnosis of the children at admission, according to the DC 0–5 scale (Axis I) shows that 26% of the children present a developmental disorder (global developmental delay, language delay, ASD), 19.6% present an anxiety disorder (generalized, separation anxiety) and 20.6% present symptoms related to a trauma.

The developmental scales show that more than a quarter of the children (26.1%) are admitted with a cognitive delay (scale score below 85), slightly less than a half (47.7%) with a language delay and 20% with a motor delay. Children also presented high risk (28.1%) or clinical withdrawal (39.3%) at entrance.

TABLE 3 Pregnancy and infant characteristics.

	N	Mean (SD) or *N* (%)
Pregnancy desire (yes)	92	79 (85)
Medically assisted procreation	92	6 (6.5)
Miscarriage antecedent	88	29 (32.9)
Stress during pregnancy	92	60 (65.2)
Tobacco during pregnancy	88	15 (17)
Drugs during pregnancy	87	9 (10.3)
Psychiatric treatment	92	6 (6.5)
Delivery mode	90	
Cesarean		21 (23.3)
Vaginal		69 (76.7)
Traumatism at delivery	81	25 (30.9)
Stay in neonatalogy	91	31 (34)
Breastfeeding	91	59 (64.8)
Weight < 2.5 kg at birth		24 (26.1)
Growth		
Normal		72 (78.3)
Upper the curve		8 (8.7)
Under the curve		12 (13)
Gestational age / prematurity		
Term		61 (66.3)
Late prematurity		20 (21.7)
Moderate		5 (5.4)
High		3 (3.3)
Extreme		2 (2.2)
Infant mental health diagnosis (DC 0–5)		
No diagnosis		23 (25)
Neuro-developmental disorder		24 (26)
Sensory processing disorder		2 (2)
Anxiety disorder		18 (19.6)
Mood disorder		9 (9.8)
Sleep disorder		18 (19.6)
Eating disorder		9 (9.8)
Related to trauma		19 (20.6)
Specific relation disorder		9 (9.8)
Somatic problems		42 (45.6)
Witness to domestic violence		35 (38)
Neglect		32 (34.8)
Maltreatment		13 (14.1)

The children witnessed domestic violence in 38% of the situations and have also experienced separation from their attachment figure or have been neglected. In 34.8% of cases, children experienced neglect and 14.1% abuse.

TABLE 4 Comparison of child development, withdrawal and parent–child interaction between entrance and discharge.

	Entrance	Discharge
Bayley (QD)	(*n* = 65)	(*n* = 26)
Cognitive	90.3 (26.1% <85)	97.3
Communication	85.9 (47.7% <85)	89.6
Motor	91.9 (20% <85)	97.5
Clinical withdrawal	(*n* = 89)	(*n* = 89)
No	29 (32.6%)	44 (49.4%)
At risk	25 (28.1%)	19 (21.3%)
withdrawal	35 (39.3%)	26 (29.3%)
Relational level	(*n* = 91)	(*n* = 91)
1	12 (13.2%)	28 (30.8%)
2	38 (41.8%)	26 (28.6%)
3	37 (40.6%)	32 (35.2%)
4	4 (4.4%)	5 (5.4%)

Child development and clinical improvement

Results at the Bayley and symptoms at entrance and discharge are presented in Table 4 and Figure 1. Sixty-five children received the developmental assessment at entrance and 26 where evaluated at the end of the intervention. An improvement in the developmental quotient between entry and discharge is observed but it does not reach statistical significance.

With respect to psychopathological symptoms, each child presents an average of 2.9 symptoms at admission, with an overrepresentation of symptoms of developmental disorder, sleep disorder, separation anxiety and relational withdrawal. At discharge, the children presented a symptom reduction with an average of 2.4 symptoms and a reduction of child's withdrawal but it does not reach statistical significance.

Parent–child interaction improvement

Nearly all patients have significant relational parent-baby relationship problems at admission. At entry, according to axis 2 of the DC 0–5, 41.8% of the dyads/tryads have type 2 relationships (strained to concerning) and 40.6% have type 3 relationships (compromised to disturbed). 13.2% have a relationship described as adapted appropriately and satisfactory (type 1). At discharge, 28.6% of patients had a type 2 relationship, 35.2% had a type 3 relationship and 30.8% had a type 1 relationship. These results are presented in Table 4.

Parental data

Mother and father clinical characteristics are presented in Table 5.

Among the mothers admitted to the PBDH, 53.8% have a psychologist or psychiatric follow-up.

The average score obtained for the Edinburgh Scale was 17, indicating the presence of depressive symptoms and a high risk of depression for these mothers. This result can be linked to the

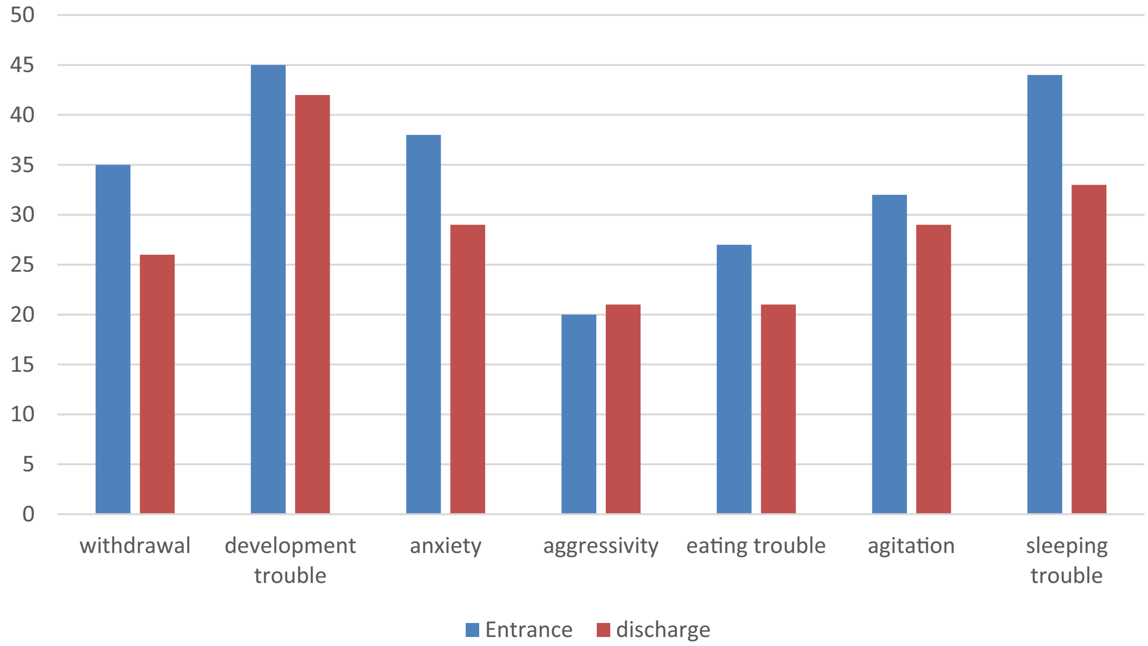

FIGURE 1
Clinical evolution between entrance and discharge.

mothers' diagnoses according to axis 1 of the DSM V: 52 have a depressive pathology (56.5%), 24 show anxiety (26%) and 24 have a borderline pathology (26%). The majority of the mothers in our care unit are suffering from anxiety and depression, with significant comorbidity (66.3% of the mothers have at least two diagnoses).

It should be noted that 41.3% have somatic problems and that more than one mother out of two (51.2%) have a traumatic past (history of psychological or physical abuse).

Concerning the fathers, 22.7% of them present a somatic pathology. 16.4% of the fathers have a traumatic past, as well as the same pathologies than the mothers (according to axis 1 of the DSM V) but to a lesser extent: depressive and borderline pathologies. This can be explained by a lack of comprehensive data collected from the fathers.

Factors contributing to a positive change in hospitalization

Table 6 examines the variables that could influence the outcome of the hospitalization. Regarding the impact of the family context, the mother's age at admission has a significant influence on the outcome of the hospitalization. It is not the case for fathers. When the hospitalization is successful, the mothers are on average 33.1 years old at admission (±6.16) against 29.4 years old (±6.21) when the hospitalization is not successful.

The age of the children at admission has no significant influence on the outcome of the hospitalization, following the Wilcoxon test.

Neither family support ($p = 0.30$) nor socioeconomic level ($p = 0.07$) seems to have a significant influence on the outcome of hospitalization.

Concerning the setting of the hospitalization, the Mc Nemar's Chi^2 test highlights that the reason for hospitalization on the mother's side significantly influences the outcome of the hospitalization ($\chi^2 = 10.62$, value of $p = 0.01$). On the contrary, the reason for admission on the child's side, the rate of attendance and the duration of hospitalization do not significantly influence the outcome of hospitalization.

Clinical situations in which mothers got support from their partners regarding the hospitalization ($\chi^2 = 6.40$, value of $p = 0.04$) without their partners being present in the care at the PBDH, have a 94% rate of success. This ratio drops to 54% when the partner is present and to 70.5% when they are absent and do not support the process. Absent partners represent 65% of the "unsuccessful" group.

Finally, the presence of a legal framework such as the youth care or youth protection at admission ($\chi^2 = 12.79$, value of $p = 0.0001$) significantly influences the outcome of the hospitalization. Hospitalizations made outside of a legal framework at admission have a positive outcome in 86.95% of the cases, whereas this ratio drops to 56% when children are admitted through a youth care and 42% through a youth protection service.

For variables grouping child-related data, those who witnessed domestic violence ($\chi^2 = 10.98$, value of $p < 0.001$), experienced neglect ($\chi^2 = 32.68$, value of $p < 0.001$) or abuse ($\chi^2 = 19.74$, value of $p < 0.001$) are more represented in the "unsuccessful hospitalization" group. There is therefore a significant influence on the outcome of hospitalization for these three variables.

Regarding child development, the mean scores at admission for the language scale and the motor scale have a significant influence on the evolution of the hospitalization. When hospitalization was successful, children had a mean score of 90.51 (±16.78) on the language scale, compared with 80.38 (±12.3) for those whose hospitalization was not successful. The children obtained an average

TABLE 5 Parental clinical characteristics.

	N	Mean (SD) or *N* (%)
Mothers characteristics		
Suicide attempt antecedent	91	17 (18.6)
Psychiatric follow-up at entrance	91	49 (53.8)
Somatic problem	90	38 (41.3)
History of physical or psychical trauma	86	44 (51.2)
Edinburgh scale	23	17
DSM V diagnostic	92	
No diagnostic		13 (14.1)
Schizophrenia/non affective psychosis		4 (4.3)
Depressive disorder		52 (56.5)
Anxiety disorder		24 (26.1)
Borderline personality disorder		24 (26.1)
Trauma disorder		6 (6.5)
Eating disorder		3 (3.2)
Substance use disorder		10 (10.9)
Bipolar disorder		1 (1.1)
Neurocognitive disorder		7 (7.6)
ASD		0
Conduct disorders		0
Comorbidity		61
Fathers characteristics		
Suicide attempt antecedent	65	2 (3.1)
Psychiatric follow-up at entrance	66	5 (7.5)
Somatic problem	66	15 (22.7)
History of physical or psychical trauma	61	10 (16.4)
Edinburgh scale		/
DSM V diagnostic	62	
No diagnostic		37 (59.7)
Schizophrenia/non affective psychosis		2 (3.2)
Depressive disorder		8 (12.9)
Anxiety disorder		3 (4.8)
Borderline personality disorder		7 (11.3)
Trauma disorder		5 (8.1)
Eating disorder		0
Substance use disorder		8 (12.9)
Bipolar disorder		0
Neurocognitive disorder		3 (4.8)
ASD		1 (1.6)
Conduct disorders		4 (6.4)

TABLE 6 Analysis of the variables influencing the outcome of the hospitalization.

Variable	Unsuccessful (*n*=20)	Successful (*n*=54)	value of *p*
Mother's age	29.4 ± 6.21	33.1 ± 6.16	<0.05
Father's age	33 [30.75–34.25]	33.5 [29–41]	0.44
Socio economic level			0.07
Low	15 (75%)	23 (42.59%)	
Middle	5 (25%)	25 (46.3%)	
High	0 (0%)	4 (7.41%)	
Unknown	0 (0%)	2 (3.7%)	
Familial support			0.30
No	10 (50%)	18 (33.33%)	
Low	1 (5%)	8 (14.81%)	
Enough	9 (45%)	28 (51.9%)	
Reason of admission			
Relational trouble	12 (60%)	35 (64.9%)	0.91
Parental difficulty			<0.05
Parental skills	6 (30%)	18 (33.33%)	
Chronic psych. Path.	10 (50%)	15 (27.8%)	
Acute psych. Path.	0 (0%)	17 (31.48%)	
None	4 (20%)	4 (7.41%)	
Concern for the child			0.07
Behavior	4 (20%)	17 (31.48%)	
Development delay	12 (60%)	15 (27.78%)	
Withdrawal	4 (20%)	12 (22.22%)	
Somatic problem	0 (0%)	2 (3.7%)	
None	0 (0%)	8 (14.81%)	
Average frequentation (d/w)	0.72 [0.55–0.86]	0.68 [0.56–0.85]	0.94
Length of stay (weeks)	27.5 [13.75–50.5]	26.5 [12.5–49]	0.83
Partner support			0.04
Involved	6 (30%)	7 (12.97%)	
Supportive	1 (5%)	16 (29.63%)	
Missing or lacking	13 (65%)	31 (57.41%)	
Youth care or protection entrance			<0.01
0	6 (30%)	40 (74.07%)	
Youth care	7 (35%)	9 (16.67%)	
Youth protection	7 (35%)	5 (9.26%)	
Low birth weight			0.9111
0	15 (75%)	43 (79.63%)	

(Continued)

TABLE 6 (Continued)

Variable	Unsuccessful ($n=20$)	Successful ($n=54$)	value of p
1	5 (25%)	11/ (20.37%)	
Witness to domestic violence			<0.01
0	4 (20%)	36 (66.67%)	
1	16 (80%)	18 (33.33%)	
Neglect			<0.001
0	1 (5%)	44 (81.48%)	
1	19 (95%)	10 (18.52%)	
Maltreatment			<0.001
0	10 (50%)	52 (96.3%)	
1	10 (50%)	2 (3.7%)	
ADBB score at entrance	10.75 ± 5.5	7.26 ± 5.11	0.31
Bayley score at entrance			
Cognitive score	89.06 ± 15.4	91.48 ± 16.8	0.61
Communication score	80.38 ± 12.3	90.51 ± 16.78	<0.05
Motricity score	85.25 ± 14.23	94.89 ± 15.68	<0.05
Child diagnostic at entrance			<0.05
0	2 (10%)	19 (35.19%)	
Sensory processing disorder	0 (0%)	2 (3.7%)	
Mood disorder	2 (10%)	5 (9.25%)	
Related to trauma	10 (50%)	4 (7.41%)	
Specific relational trouble	1 (5%)	1 (1.85%)	
Autism spectrum trouble	0 (0%)	2 (3.7%)	
Global developmental delay	3 (15%)	1 (1.85%)	
Language delay	1 (5%)	1 (1.85%)	
Other neuro-developmental	0 (0%)	1 (1.85%)	
Separation anxiety	0 (0%)	10 (18.52%)	
Generalized anxiety	1 (5%)	3 (5.56%)	
Sleeping trouble	0 (0%)	4 (7.41%)	
Eating trouble	0 (0%)	2 (3.7%)	
Relational level entrance (Axis II DC 0–5)			<0.01
1	0 (0%)	1 (1.89%)	
2	3 (15%)	32 (60.38%)	
3	15 (75%)	19 (35.85%)	
4	2 (10%)	1 (1.89%)	
Sibling placement history ($n = 10$)	6/10 (60%)	2/26 (7.7%)	<0.01
Maternal suicide attempt	6 (30%)	10 (18.5%)	0.4547
Father's suicide attempt	0	1 (1.8%)	1
Maternal trauma history	19/19 (100%)	23/49 (46.9%)	<0.001
Father trauma history	4/11 (36.3%)	5/27 (18.5%)	0.30
Maternal diagnostic (DSM V)			0.048
0	0/20 (0%)	3/53	
1	1/20 (5%)	3/53	
2	9/20 (45%)	27/53 (50.94%)	
3	0/20 (0%)	11/53 (20.75%)	
4	3/20 (15%)	5/53	
5	2/20 (10%)	2/53	
7	4/20 (20%)	2/53	
9	1/20 (5%)	0/53	
Paternal diagnostic			0.1077

(Continued)

score of 94.89 (±15.68) on the motor scale, compared with 85.25 (±14.23) for the children belonging to the "uncompleted hospitalization" group.

The child's diagnosis (according to DC: 0–5) also has a significant impact in the outcome of hospitalization ($\chi^2 = 33.68$, value of $p = 0.001$). Trauma-related disorders are more represented (50%) in the "unsuccessful hospitalization" group and account for 1 in 2 patients, whereas separation anxiety disorder is the most represented (18.52%) in the "successful hospitalization" group. Nevertheless, the absence of disorder is the most represented (35.19%) in the successful hospitalization group.

Relationship quality is closely linked to the outcome of hospitalization at PBDH ($\chi^2 = 13.71$, value of $p = 0.003$). The vast majority of patients present a type 3 relationship (compromised to disturbed) in the "unsuccessful hospitalization" group, whereas the type 2 relationship (strained to concerning) represent 60% of the "successful hospitalization" group. Patients with the latter type of relationship have 91.4% (32/32 + 3) chances to have a successful hospitalization. This ratio drops to 55.9% (19/19 + 15) for type 3 relationships. Finally, the Wilcoxon test suggests that there is a significant improvement in the parent-baby relationship between entry and discharge when the hospitalization is successful ($p < 0.001$).

Regarding siblings, a history of placement significantly influences the outcome of hospitalization. When the hospitalized child's siblings have been placed, the hospitalization has a 25% probability to succeed. The number of siblings with a history of placement is more represented in the "unsuccessful hospitalization" group (60%).

With respect to parental history variables, there was significantly ($\chi^2 = 14.15$, value of $p<0.001$) more traumatic history among mothers in the "hospitalization not completed" group (100%). This feature is not found among the fathers. The maternal pathology significantly influences the outcome of the hospitalization. Depressive disorders (2; 50.94%) and anxiety disorders (3; 20.75%) are more represented in the "completed hospitalization" group. Depressive disorders are also found in 45% of cases in the "not completed" group and are the most common disorder in this latter group.

Discussion

This retrospective study is the first major study to focus specifically on the developmental and clinical improvement of the baby in a PBDH. It is also the first survey to highlight significant differences between day and day/night units, both in the population data and in the characteristics of admission in a parent-baby unit. Strengths and limitations will be discussed.

Admission data

One of the first findings is that the length of stay in day units (29 weeks) is much longer than in day/night facilities. Based on the existing literature, the durations of the stay in the later vary from 5.93 days for a study in the US (36) to 11.6 weeks in Israel (37). It is even more obvious when considering only therapeutic care (34 weeks). These differences for day/night units seem to partly result from the different nature and purpose of MBUs in different countries and regions. Nevertheless, the possibility of receiving intensive care in a day unit, avoiding the break in contact inherent in a day/night hospitalization, undoubtedly offers a longer-term perspective of therapeutic management. Care is also longer for chronic pathologies and personality disorders (22), yet our sample includes a high rate of mothers with borderline personality disorder (26%). Based on the type of psychopathology we receive (including a high rate of parents who have experienced traumas weakening the construction of the bond with the other), the high duration of the care seems necessary to accompany these families properly and respect their temporality. It is also necessary to consider that the hospitalization framework, putting the baby at the very heart of the care, has an influence on the duration of hospitalization. Clearly, the outcome changes if the objective of hospitalization is the clinical improvement of the parent or of the baby.

The average age of care in day units differs from that of full-time units, where most admissions take place very early (before 8 weeks of age). The greater psychiatric morbidity of mothers admitted to residential units probably explains the earlier age of admission (52). The high percentage of mothers suffering from depression is a factor that probably contributes to a later age of admission (average age 15 months). It might indeed literally immobilize the mother (interest in home care), and present constraints related to traveling with a young child. These results should encourage the development of psychiatric services offering postpartum home care. Furthermore, the positioning of university hospitals as third-line hospitals may also explain the later move of families who already benefit from outpatient care before being reoriented to day hospitalization. Finally, in many day/night units, the age of admission for babies is sometimes limited to the first year of life, whereas our system welcomes them until they start kindergarten.

Unlike the situation in French MBUs, where the number of voluntary admissions reaches 90%, in our PBDH a significant number of admissions are supported or imposed by the youth care or the youth protection. In our study, 32.6% of the families are supervised by one of these two services. This percentage is similar to the one established during the Marcé research in Belgium between 2001 and 2007 (34%) (53). The ways in which medical and social services work together differ significantly in the two countries.

Examination of the grounds for admission shows a significant difference from what is described in the literature for day/night units. The results of the Marcé study show that in 80% of cases, "children are generally well" on admission. In our study, this proportion is reversed, since in 90.2% of cases, difficulties for the baby are observed on admission (all areas of development combined). Once again, it seems that one explanation for this situation is the one of the positioning of the facility, which the workers identify as a "care structure at the baby's departure," whereas the day/night mother-baby units rather welcome mother populations identified as being at risk, in an earlier way at the end of the maternity ward when the baby's psychopathology is not yet at the forefront.

The high rate (17.4%) of referral of the baby to a care facility at the end of the hospitalization deserves attention, since it is similar to or exceeds certain percentages found in day/night studies in Europe [4 out of 23 placements, i.e., 17.4% in the study by (54); 14.8% in the study by (43)]. It should be taken into consideration that some of the children ($n = 7$) were already in institutional care (nursery or hospital) and that 5 of them remained there after hospitalization. The higher average age of our population probably partly explains this high rate since a decision for separation is sometimes taken when successive therapeutic proposals fail. It is interesting to note that in other recent studies, including an Australian study examining the orientation of patients at discharge from hospital (55), the return of children to their home with their parent (s) is almost systematic (only 2.4% separation), raising questions involving the health and child protection policies in place in each country.

Social vulnerabilities, partner involving, and familial support

As we expected, the context of vulnerability is similar in our population and in the day/night units. The vast majority of parents have poor financial resources, lack family or extra-familial assistance and have a family history of immigration. These data are difficult to compare with the literature given the heterogeneity of the measures used.

Nevertheless, it is imperative to consider the seriousness of this vulnerability, since it will be added, for some parents, to psychopathology and/or traumas experienced in childhood. The interplay between socioeconomic factors, migration history and perinatal health is widely described in the literature (56, 57). However, unlike what has been shown in previous studies (58), none of the economic or social variables considered individually, is associated with a pejorative outcome of hospitalization in our study. Only the mother's age seems to have an influence on the outcome of the hospitalization. Indeed, early motherhood has been widely shown to be associated with poorer child development and young mothers are at increased risk for postpartum depression (59, 60). Multivariate statistical analyses could help measuring the risk associated with cumulative factors. We have also studied data related to the lifestyle of the families in care. As shown in other studies on MBUs, the majority of families attending the PBDH live together as couples [68.5% in our study vs. 68–90% according to the studies reviewed, (24)], yet we expected to receive a majority of single parents since support by the group is an indication for referral to our service.

The place of fathers in mother-baby hospitalization and perinatal care facilities has evolved considerably since the units first opened (61), but this topic is hardly addressed in the research literature (62, 63). Nowadays, some residential units have small studios or rooms set up to accommodate inpatient couples. The name "Parent-Baby Day Unit" implies, *de facto*, the inclusion of the partner in the care. In our study, the isolation of a large number of women in the exercise of their parental functions was found (absence of the father or of his support in 51% of the cases). The comparison of the two populations shows that hospitalization is most likely to be successful when the father is supportive, without necessarily being present at the hospital. A more detailed analysis of these data helps understanding this paradox since paternal psychopathology concerned a higher number of fathers when the father was the caregiver involved with the baby during hospitalization (15 of 18). These results should be considered with caution, since a considerable amount of data concerning paternal psychopathology is missing. In addition, it was easier to assess parental psychopathology for fathers who spent full days in the unit. In any case, these data support the point of view of some authors who consider the father's refusal of any contact with the hospitalization services as a poor prognosis factor for the evolution of the dyad (44).

Focus on the child and the pregnancy

The analysis of the perinatal risk factors in our population illustrates the importance of antenatal prevention: most of the families attending our unit have a history of complicated pregnancy or immediate postpartum. The high proportion of children who stayed in the neonatal unit (34%) and the number of children with low birth weight (26.1%) clearly highlights it. This latter rate is statistically significantly higher than the data in the Belgian population, and the literature has highlighted its correlation with socioeconomic risk factors like in our population (64). The study by Wright et al. (41) similarly found an abnormally high rate of children with low birth weight (20%) among children admitted in a MBU, while the percentage of prematurity did not differ from their national average. As a reminder, many epidemiological studies show a link between major psycho-emotional stress during pregnancy or low birth weight and subsequent outcomes in terms of psychopathology but also cardiovascular diseases (65, 66).

The major results of this research show an improvement in the developmental quotient of the child between entry and discharge. There is also an overall decrease in the symptomatology present at entry. None of these results is however statistically significant and it must therefore be interpreted with caution given the inherent bias of the retrospective aspect of this study. A major weakness of this work was the difficulty to collect quantitative data. Developmental scales could only be collected at discharge for 26 children (compared to 65 at entry). There are three possible reasons for this loss of data. The main one is that the duration of intervention for many children (9) did not allow for re-administration of the test (the test is administered at 6-month intervals to avoid learning bias). The second reason is linked to an abrupt interruption in the care process with no test at the end (generally decided by the parents) in 9 situations. Finally, the last explanation is the difficulty of collecting quantitative data from a research perspective while engaged in a clinical process (e.g., systematizing the use of exit scales even if the patient is clinically better) (9 situations). Equally, the improvement in scores cognition, language could have been better studied if the developmental assessment had been done in relatively older age groups (the mean age of admission of the infant was 14.8 months, one third of the population was below 6 months and around 50% was less than one year). This is given to lower scores of reliability of Bayley scale in younger age groups (67). Similarly, for showing a visible change in scores in Bayley scale with any intervention a longer period of interval (T2−T1) is preferable. In this study, the average period was 6.65 months (range from 1 week to 24.8 months).

There is a lack of use of a validated grid to collect symptoms. The age range of the infants at admission was lower in the use of Pediatric behavioral assessment scales or check lists though some infants in the present study were referred with behavioral trouble. Using an appropriate scale for sensory integration profile could be an important parameter for assessment in infants who have suffered from stress disorders, since around 30% infants were found to have suffered from sleep and eating disorders in the present study. But, at this time, the most significant test (Infant/Toddler sensory profile instrument) is still not translated and validated in French for this age group (68).

Nevertheless, this is the first study to systematically examine the development of children in a day parent-baby unit using a standardized developmental scale. Only two other studies conducted on parent-baby day-units look at the psychopathology and development of the child at entry (29, 41). The first used the Marcé questionnaire and reported the following figures (child's condition at entry): 76% cases children in good health, 11% psychomotor problems, 9% emotional problems and 5% somatic problems. These results are far from ours, which reveal an alarming picture of the children's condition at entry in our unit (45.6% somatic problems, 26.1% cognitive delay, 47% communication delay and 20% motor delay). Concern for the child's condition is even greater in the unfavorable outcome group (significative statistical difference for communication and motricity assessment). The second study (41) used the Ages & Stages Questionnaires-3 to screen children's development, and found the same trends than us, with greater difficulties in the socio-emotional areas than in the motor and cognitive areas. The strong representation of somatic problems, and in particular gastro-alimentary problems, should encourage the team to propose more targeted therapeutic treatments for this group of pathologies (sensory workshops around food, more individual care during meals, cooking workshops with parents, integration of a speech therapist in the therapeutic team, etc.)

Another study looks at the early relational withdrawal of children from mothers hospitalized in MBU. The study boosts it around 35% (69). We find in the PBDH the same proportion, 39.3% of children with marked relational withdrawal, to which can be added those who are considered at risk of withdrawal (28.1%). Despite the fact that number of children admitted to the unit for developmental assessment show withdrawal and communication disorders, this proportion remains significant. In the general population, a high prevalence (up to 75%) is found among infants whose mothers met criteria for a major depressive episode at 6 months postpartum (70).

The child population at PBDH is often impacted by trauma and the results show that these life events significantly influence the hospitalization negatively. The rates of neglect and the number of children witnessing domestic violence are very high (34.8 and 38% of cases respectively), just as the number of children presenting disorders related to mistreatment and overrepresented in the pejorative group. In comparison, data from the literature estimate neglect at 1–15%, and witnessing domestic violence at 10–20% (71).

Dyadic relation and parental psychopathology

The assessment of the relationship through the Axis II of the 0–5 years scale shows an improvement of the relationship between the partners of the mother–child dyad at the end of the care process. This improvement is also statistically higher in the successful group and the dyadic relation was worser in pejorative group, as expected. Although the 0–3 scale is recognized for the value of its multiaxial assessment, (72) point out the lack of a quantitative criteria between pathological and non-pathological, as well as the lack of information related to the degree of intensity of the symptom and explanation of its meaning. Consequently, the parent–child interaction should be investigated through other tools at PBDH. It would be interesting to know the relational attachment patterns of parents and children.

Compared to the populations in MBUs, the psychopathology of the parents appears to be less severe in the PBDH unit. The maternal population is mainly represented by depressive disorders (37%) and anxiety disorders (26%), whereas a systematic review of MBUs identifies depressive disorders (50%), psychotic disorders (25%), and bipolar disorders (10%) as the most frequent pathologies. (24). Anxiety disorders are therefore much more represented in PBDH than in UMBs, which had been highlighted by the study of a day unit (73). High anxiety levels are known to be possible risk factor for the development of disorders in parenting (74). The high rate of trauma experienced by the patients was predictable, given the psychiatric fragility of the sample population, and the prevalence of borderline personality disorder (29%), as shown in previous studies of MBU's (75). The appropriate management of this pathology is even more important given that a study identified it as one of the first factors associated with placement in a population attending a mother-baby unit (76). Our results did not show a significant influence of this pathology on the outcome, but maternal trauma history is over-represented in our pejorative evolution group. For the management of this disorder, scientists generally advocate a therapeutic setting that combines flexibility, stability, and availability (77), which characterizes daytime settings over a day/night structure.

Interestingly, our study shows statistical differences in the repartition of psychopathological pathology between the two groups. Mothers with an anxious diagnosis are all included in the successful group. This suggests that this profile of patients benefits particularly from our care.

Parents' somatic complaints are seldom studied in the literature. Our study shows that this aspect of parental symptomatology is significant (39% of mothers, 26% of fathers show a current somatic pathology) and must be addressed simultaneously within the care to the families (collaboration with a treating physician, relay to the proximity care network, workshops or body therapies) because it can sometimes be the only witness of a parent's psychic suffering (psycho-somatic disorder). The need to work hand in hand with a psychiatrist and a pediatrician is obvious in the perinatal care system, but the aspect of the parents' physical health is insufficiently addressed.

Conclusion and perspectives

This study contributes to giving the baby its central place in perinatal settings, particularly in mother-baby units where concern for the mother was historically the primary focus of care. The strength of our research lies in the precise description of the physical and developmental state of the baby at admission to a parent-baby day unit. Developmental, behavioral, and relational improvement was observed at discharge but the sustainable effect of intervention needs prospective long term follow up studies.

The differences observed in our study between the day/night population and the daytime population should contribute to improve the care pathway for families. In our study, the system seemed to be more effective for anxiety disorders of the child and the parent. It also underlines the vigilance necessary for the implementation of care which could be inadequate for the child when his clinical presentation is already worrying at the time of admission.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving human participants were reviewed and approved by Comité d'éthique de l'hôpital universitaire des enfants Reine Fabiola. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

Author contributions

VD conceptualized the study, and wrote the grant funding proposition, together with AM, LC, and CG. AM prepared the first draft and subsequent versions of the protocol and this manuscript, wrote the ethical approvals documents, and selected the parental scales. AM and LC collected research data and analyzed them. LC and CG are the psychologists responsible for the developmental and relational assessment tools. PB was statistician who led the design of statistical analysis model. All authors contributed to the article and approved the submitted version.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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The polyfactorial model of autism and the question of causality

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After recalling the different pediatric, psychopathological and child psychiatric models of mental disorders in children and adolescents, the author presents in detail the so-called polyfactorial model, which includes primary, secondary, and mixed factors. This model is the epistemological heir of the Freudian concept of "complementary series." The example of autism is then explored as a paradigm of the usefulness of this polyfactorial model. Finally, we reflect on the notion of causality, from Aristotelian causality to epigenetic causality, which could 1day re-legitimize psychoanalysis and the impact of the relationship on genome expression.

KEYWORDS

autism, Neuroscience, Psychoanalysis, epistemology, Model, epigenetic

Introduction

The question of epistemological models is important in child psychiatry, on the one hand to think about the question of causality and on the other hand to guide the choice of therapeutic strategies.

Growth and psychological maturation correspond to eminently complex processes and the same is true for developmental disorders.

It is an oversimplification to try to make people believe otherwise, and therefore the management of mental disorders in children and adolescents must carefully consider its reference model(s) (1).

In this work, autism will be taken as the paradigm for reflection, but many of the reflections presented in this respect can, in our opinion, be extended to other child psychiatric disorders.

The pediatric, psychopathological and child psychiatric models

The question is not whether one model is more valid than another, but to emphasize that each discipline, depending on its practice and objectives, refers to models that are specific to it and therefore useful.

Pediatrics refers to a medical model—like all somatic disciplines—which is rather monofactorial (a single cause is supposed to account for the pathological situation), deductive (based on univocal cause-and-effect relationships) and referring to a linear type of temporality (organized according to the arrow of time in the usual sense of the term).

Psychoanalysis (and psychodynamic psychopathology in general) refers to a different model, of a polyfactorial nature (as S. Freud had proposed it in 1915/17 with his concept of "complementary series") (2), inferential (proceeding by associations of thoughts and not by deduction), and based on a circular temporality integrating the so-called after effects, according to which the past partly accounts for the present, but the present also allowing, permanently, to retell, rewrite and re-construct the past history.

It should be noted that the deductive somatic model aims for rapid efficiency, whereas the inferential psychopathological model cannot claim to master the tempo of understanding, elaboration and decision.

In any case, the child psychiatric model seeks its place and its identity in relation to these two models.

Depending on the country and the time, the psychiatric model is more or less close to one of the two previous models.

Today, in Anglo-Saxon countries, the child psychiatric model is very close to the medical model (3), whereas in France, it remains, in a way, at equal distance from the medical model and the psychopathological model, still quite strongly impregnated by the psychoanalytical references which presided over the birth of child psychiatry in our country.

It is therefore clear that pediatricians on the one hand, and child psychiatrists, psychologists and child psychoanalysts on the other, do not refer to the same model.

This does not mean that part of the conceptual path cannot be travelled jointly, even if fundamental differences exist. But it does underline the fact that this common path requires mutual esteem and respect between the representatives of the somatic and psychic disciplines.

The difference in tempo often acts as a seed of possible dissension, and in this respect, it is important to say something about the concept of "negative capacity" that Bion (4) developed from the work of the romantic poet J. Keats (5).

This is the clinician's ability to tolerate ignorance for a certain period of time, not to want to understand everything immediately, to allow himself to be impregnated by the clinical situation, to allow himself to be deeply affected at the level of his emotions, and finally to know how to give time to time so that elaborations, interpretations and conclusions do not have the value of theorizing and defensive force.

This aptitude is necessary to the caretakers of the psyche whereas it is not expected nor even sometimes necessary for caretakers of the body: hence, sometimes, a certain number of possible misunderstandings and specially within the framework of what is called liaison child psychiatry.

The polyfactorial model

The polyfactorial model probably applies to a number of somatic pathologies (6, 7), but in the field of child and adolescent mental disorders, it seems to us that the most plausible model of the etiology of autistic pathology at present is a polyfactorial model, which is the only one capable of articulating physical causality, interactive causality and epigenetic causality, thereby imposing on us an integrated multidimensional management of this pathology.

In this perspective, autistic functioning would then be a sort of "common final pathway" of a whole series of etiopathogenic configurations within which endogenous and exogenous factors would always be present, but in a variable proportion according to each child.

What is important to specify is the double level of the polyfactorial dimension that can be invoked both for the primary factors of vulnerability, which are always multiple (exogenous and endogenous), and for the secondary factors of fixation and maintenance, among which the deferred action is essential, because the meanings that the first interactive dysfunctions of their child may have for such and such parents can then function autonomously as secondary causal factors, as we shall return to.

This being said, primary factors are only risk factors, whereas secondary factors are factors that fix a psychopathology that is sometimes partially reversible.

It is likely that the years to come will see the emergence and development of the concept of "epigenetic causality" (8) which will finally make it possible to articulate, in a dialectical and singular manner for each patient, the role of internal determinants (the personal part of each person with his or her genetic, neurobiological, cognitive equipment and so on …) and external determinants (the part of the environment in all its components, ecological, dietary, social, cultural, family and so on…) as to the origin of his or her difficulties or developmental disorders.

Primary and secondary factors

*The Freudian concept of "complementary series" (Freud, 1915/1917) mentioned above represents, in a way, the epistemological ancestor of our current polyfactorial model.

This concept constituted, at the time, a real epistemological revolution insofar as it was a profound break with the prevailing medical vision, which was then closely linked to the perspectives developed by Claude Bernard (9).

Freud proposed a sort of power grab by making the hypothesis that the constitution of any neurotic organization could only be conceived as the result of the joint influence of endogenous and exogenous factors.

Among the endogenous factors, he had in mind the fixation points that he had put forward as part of his scheme of psycho-affective development: an oral fixation point predisposed, according to him, to the organization of a hysterical neurosis, an anal fixation point to the organization of an obsessive neurosis, and a phallic or urethro-phallic fixation point to the organization of a phobic or hysterical neurosis.

But, he specified, these fixation points could not be considered as causes in the linear sense of the term, because they were, according to him, only predisposing factors, not determining ones.

In this model, exogenous factors had to precipitate and decompensate things, and among these exogenous factors, he insisted, in a central way, on the question of sexual frustration.

He added that in this model, there was a sort of 'sliding scale' between endogenous and exogenous factors, the more important the one being, the less important the other needing to be, but the presence of both being indispensable in each subject to account for his or her psychopathology, according to a pathogenic equation that was therefore strictly specific and individual.

This conception of psychopathological etiology must undoubtedly be clarified; it must however be emphasized that it was extremely innovative at the time, and that our current polyfactorial model derives quite directly from it, even if it has since become much more complex.

* Today, in fact, we consider that any psychopathological situation is the result of the interplay of primary and secondary factors, and that these two lines of factors are each fundamentally polyfactorial in

nature. Both primary and secondary factors can, moreover, be of an endogenous nature (personal part of the child) or of an exogenous nature (environment).

Primary factors are only predisposing factors (or vulnerability factors): they are not sufficient, but they are necessary, they do not create psychopathology, but they increase the risk of it.

Secondary factors are decompensating (triggering) factors for psychopathology in subjects who have primary factors that make them vulnerable.

A paradigmatic example: childhood autism?

By way of illustration, we will now take the example of childhood autism and pervasive developmental disorders, while making it clear that in our opinion, these considerations on autism and autism spectrum disorders have conceptual implications that extend well beyond these various pathologies.

(1) Examples of primary (vulnerability) factors

– *Genetic factors*

These are undeniable, but they are now to be understood from the point of view of genetics of vulnerability, and not causal genetics in the classical sense of the term (10). It is known, for example, that Fragile X syndrome is a predisposing factor for childhood autism, since although in the population of autistic children there is a clear excess of children carrying this anomaly (about 7%) compared to the frequency observed in the general population, conversely not all Fragile X children are autistic.

Furthermore, Kanner's (11) genetic model of autism as a specified pervasive developmental disorder is currently oriented towards a constellation of alleles involved in so-called "candidate" genes, spread over all the chromosomes and whose joint state would underlie the autistic predisposition. Only this model makes it possible to account for the fact that around an autistic proposer, nearly 1% of other cases are found among first-degree relatives, whereas from the second degree of kinship, the frequency falls back to that observed in the general population (10).

No Mendelian model is capable of explaining this phenomenon, and we are therefore in the perspective of a susceptibility genetics referring to the question of the heritability of complex traits and to epistatic interaction processes (12), which now invite us to classify genetic factors among the so-called primary factors of the polyfactorial model.

Even the recent discoveries (13) on neuroligins and neurexins, which are said to occur in less than 1% of autism cases, should probably be interpreted only in terms of vulnerability.

– *Neurological factors*

The same reasoning as for Fragile X syndrome can be applied to Bourneville Tuberous Sclerosis (BTS): among autistic children, there is a clear increase in the frequency of this condition compared to the general population (A. (14)), but the fact that 25–60% of children affected by this particular encephalopathy are also autistic makes BTS a primary—and only primary - predisposing factor, *via* mutation of the TSC1 (Hamartine) and TSC2 (Tuberine) genes.

The complex problem of epilepsies associated with approximately 30% of autism cases and appearing at some point in the course of the disease must also be mentioned in this section (15). It is still difficult, at the present time, to really specify the intimate mechanisms of this association, which probably differ according to each type of epilepsy, but as far as West syndrome is concerned, it seems that it can be considered as a primary risk factor: it is known that 20–25% of children with West syndrome become autistic in the first 2 years of life, without it being possible to predict in advance which ones, and on the basis of a profound interference with early interactions by certain motor stereotypies of the upper limbs, as Ouss et al. (16) have been able to begin to show in the framework of our research program "PILE" (International Program on Child Language).

As for the anomalies revealed by functional MRI of the superior temporal lobes in children with autism (17), if in some cases they are not only the consequence of autistic-like functioning, they can perhaps be understood as reflecting an important etiopathogenic link, but here we are no longer quite in the framework of primary factors in the strict sense.

– *Sensory factors*

Unlike blindness, deafness appears to be an undeniable risk factor for infantile autism, to the extent that it was thought, at one time, that it was important to identify a specific syndrome associating autism and deafness. Today, while auditory and/or visual cortical hypersensitivity has been demonstrated in the pathogenesis of autism (18), it is thought that deafness plays a primary, non-specific role *via* the partial relational isolation it causes in a large number of cases.

– Infectious factors

Congenital rubella was once shown to occur more frequently in the histories of children with autism than in the general population. The anti-rubella vaccination has somewhat relegated this question to the background, but it remains that the neurological and sensory disorders linked to this embryo-fetopathy could undoubtedly function as primary risk factors.

– Environmental factors, finally

The whole question of interactive dysfunctions and maternal depressions is at stake here. This question must be treated with caution. It is important to consider that these environmental factors can only be considered as primary risk factors within a truly polyfactorial model, i.e., considering that they probably need to be associated with other primary factors (genetic, in particular) to have a real impact. With this proviso, which is essential, we can then imagine that they can play a role as risk factors to be integrated into the "autising process" proposed by Hochmann (19).

This list is by no means exhaustive, and developments in child psychiatric research will very probably lead to new discoveries in this field in the years to come, perhaps with the identification of primary environmental factors in the broad sense (dietary or ecological, for example).

(2) Examples of secondary (triggering) factors

As child psychiatry is still relatively young, these secondary factors are far from being fully identified.

As we have just said with regard to primary factors, the future will certainly allow us to specify, in a wide variety of fields, secondary factors for triggering or decompensating autism in vulnerable children (environmental, nutritional, socio-familial, cultural, ecological, anthropological factors and so on…)

It is now thought that certain perinatal factors, and in particular maternal stress, could play a role in triggering autistic organization in certain babies who are vulnerable in terms of their primary factors.

Certain modifications of the maternal lifestyle as well as perinatal maternal stress as well as sleep disorders and maternal melatonin metabolism could also be invoked (20, 21).

To which we can also add that abnormalities in the general movements of the fetus and the baby during the first months of life (extension reflex and "fidgety" movements in particular) could testify to a neurodevelopmental fragility and interfere with the system of early interactions (A. Beaulieu) while also offering the conditions for an early prevention of the emergence of an autistic organization.

For the time being, the secondary factors that we know best, and on which we can act the most, are the relational factors, i.e., the child's encounter with the psyche of others, within the framework of the "autising process" mentioned above.

It is not at all a question of reopening the debate on the guilt of families in the genesis of infantile autism, a debate which we know the ravages to which it has given rise (22, 23).

It is clear that parents are in no way responsible or guilty for their child's autism, which can now only be understood from a polyfactorial perspective.

However, within the framework of a polyfactorial model such as the one presented here, it must be possible to question the status of possible early interactive anomalies as primary or secondary factors, and possibly take them into account as such, either from a prevention (secondary) perspective, or from a care perspective.

(3) Thus, what is fascinating in child psychiatry is that certain factors can play as primary or secondary factors depending on the case, and that it is the role of psychopathological analysis to be able to specify things as finely as possible.

Let us take the case of quantitative or qualitative deficiencies, which have been the subject of particular attention for a number of years.

In some cases, deficiency factors act as primary factors of fragility, and we know, for example, that some children presenting an anaclitic depression, in the sense of Spitz (24), can perfectly well become autistic if the deficiency situation persists and the experience of the orphanages in Romania has again recently provided sad confirmation of this (25–27).

On the other hand, it seems, according to F. Tustin (28), that autistic pathologies are, moreover, quite often the result of the encounter of vulnerable children with an environment that is deficient or insufficiently available, at the interactive level, for this or that reason.

A case-by-case analysis is therefore essential.

All the more so as the frequency of cases of autism seems to be increasing, currently within families in great precariousness (29) and that, among these, the number of migrant families is undoubtedly significant.

As a result, we should not allow ourselves to think, as some authors have hastily done, that migrant families bring with them autism genes; we can see the unjustifiable shortcut that this reasoning makes, with obvious political implications.

The reality is more complex than that and, without doubt, we should rather take into consideration the fact that socio-familial precariousness may well, as a secondary factor, decompensate children who are vulnerable in terms of their primary factors (and in particular genetic factors) within the framework of an epigenetic causality; these children would probably never have become autistic without the encounter with this particular and painful sociological reality.

Taking into account, on the one hand, the intertwining of primary and secondary factors and, on the other hand, the fact that the same factor may intervene, depending on the case, as a primary or secondary factor, seems to us at present the only way to allow room for freedom and a place to the effects of encounters in psychopathological etiopathogeny, which would otherwise risk being reduced to a linear and reductive schema of a strictly endogenous and neurodevelopmental type.

From Aristotelian causality To epigenetic causality

The central epistemological problem posed by this reductionist view, which is belied by the clinic, paradoxically seems to be often overlooked.

The different types of causality

Without detailing here the four types of causes that Aristotle (384-322 BC) defines in his Nicomachean Ethics (30) (the material cause, the formal cause, the driving cause and the final cause), we know that from the point of view of a dynamic psychology or psychopathology, it would undoubtedly be interesting to distinguish between the causes that drive (in reference to the drive system) and the causes that attract (in reference to the goal representations).

The drive itself includes its more or less specific goal which pushes the subject to action and the goal-representations (in the Freudian sense of the term) have an effect of attraction which mobilizes the action procedures of this same subject.

But, once again, the essential thing is undoubtedly to resolutely consider a polyfactorial model, both for the approach of development and of developmental disorders.

Within the framework of this polyfactorial model, the notion of interactive causality emerges, involving dialectical effects between the personal part of the subject (i.e., his or her genetic, neurobiological, somatic equipment and so on..) and the role of the environment in all its components (ecological, biological, nutritional, social, family, cultural and so on…). The relational component (i.e., the subject's encounter with the psychic work of others) has a particularly important function within the environment, and is essential for the disciplines devoted to the functioning of the psyche.

Taking this relational component into account within a polyfactorial model does not mean a return to a purely external causality if one clearly maintains the framework of a polyfactorial model.

In this perspective, there would probably be no pure psychological causality insofar as the impact of the environment depends fundamentally on the personal part of the subject.

Even in the case of traumatic pathology, the study of Post Traumatic Stress Disorders (PTSD) must today consider the subject's 'temperament' for neuroscientists (which corresponds to what psychoanalysts call history and deferred action) in order to understand the different effects of the same trauma on different subjects.

The reign of an exclusive psychogenesis thus appears today to be definitively outdated.

In fact, we would like to insist here on the notion of epigenetic causality, which represents one of the dimensions–probably the most recently discovered—of this interactive causality, a dimension linked to the concept of epigenesis.

Epigenetic causality In general and In relation To autistic organizations

Psychological development and the disorders of this development are played out at the exact intersection of a certain number of endogenous factors and a certain number of exogenous factors.

There is no room for any kind of division and psychopathology—whether psychoanalytical, attachmentist, systemic, cognitive or developmental—must imperatively integrate this fundamental and founding dialectic between internal and external determinants.

On this basis, many developmental disorders can probably be conceptualized as the result of a mixed epigenetic causality.

What does this term mean?

Usually, when we talk about epigenetic causality, we refer to the influence of the external environment on the expression of the genome.

However, the problem of autistic ontogenesis perhaps also invites us to consider the influence of experiences, feelings and bodily experiences themselves on the expression of the genetic determinants of this epigenesis.

In other words, if it is plausible to imagine that the nature of the child's early interactions with the adults who take care of them (parents and/or professionals) can have an impact on the expression and regulation of the genetic part that underlies the setting up of an autistic type of organization (we could speak here of external epigenetics), however it is not impossible to think that the bodily and sensory experiences linked to autistic functioning itself may also have an impact on the expression and regulation of the parts of the genome that govern the genetic level of autistic susceptibility (here we could talk about internal epigenetics).

It is the intertwining of these two facets of epigenetics that leads us to propose the term mixed epigenetic causality.

The causality of autistic pathologies or when The consequences of first causes become second causes

However, as psychoanalyst as one may be, we can affirm today that there is no pure psychogenesis of autism.

Not just anyone becomes autistic, as the study of extreme situations such as the one studied in Romania during the opening of nurseries and orphanages at the end of Ceausescu's political era has clearly shown.

All the children discovered there were certainly severely deficient in the sense of R. Spitz's hospitalism (24), but only 30% among them were truly autistic (even if there are areas of symptomatic overlap between deficiency pathology and autistic pathology).

The autistic causality is most likely, as we have seen, polyfactorial, interactive and epigenetic.

This being so, things are even more complex insofar as in the framework of the "autising process" described by Hochmann (19), the consequences of the very first dysfunctions (whether they occur in the baby or in the adult) can then assume the status of second causes and thus turn on a sort of interactive spiral that is dangerously pathogenic and destined to rapidly self-aggravate.

Can epigenetic causality re-legitimize psychoanalysis?

Lebovici often said (unpublished oral communication) that psychoanalysis had nothing to fear from the current spectacular advances in neuroscience, and that it was even looking forward to them, as they would certainly give us new doors of entry into a polyfactorial model which it is important for us to stand firm, in the field of psychopathology.

It is in this perspective that the following lines are written.

Whatever the future of this hypothesis of a mixed epigenetic causality, which is of course only valid as a conceptual proposal at the present time, we wish to emphasize that psychopathology today can no longer ignore the body, this body which is in essence at the interface of the relationship with the external environment and our internal perceptions forming the basis of our sensoriality, our sensuality and hence our sexuality.

The future of psychoanalysis in a sense is at stake here, because in view of current neurobiological positions, the requalification of speech will probably only be possible through the demonstration (still to come) of its epigenetic effects, which are already plausible.

Conclusion

Epistemology in psychoanalysis and psychiatry is still insufficiently developed (31); it is nevertheless essential for us to specify the models to which we refer implicitly and/or explicitly, to think about the question of causality and to choose our therapeutic strategies.

We have taken the example of autism, which is known to be a controversial subject, but this epistemological reflection concerns all mental disorders in children and adolescents and can only be effectively conducted in a resolutely transdisciplinary spirit.

Data availability statement

The original contributions presented in the study are included in the article/Supplementary material, further inquiries can be directed to the corresponding author.

Author contributions

The author confirms being the sole contributor of this work and has approved it for publication.

Conflict of interest

The author declares that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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The genesis of perinatal clinical psychology and its contemporary issues

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The main aim of French clinical psychology is to explain the psychic processes of transformation, to which the subject is central. In this context, transformations in the perinatal period open an innovating field in perinatal clinical psychology focused on the conscious/subconscious, subjective/inter-subjective psychic reality of a subject who is in the process of becoming (or becoming once again) a parent and being born a human.

KEYWORDS

clinical psychology, perinatality, parenthood, psychic reality, perinatal clinical psychology

1. Introduction: clinical psychology

Clinical psychology is defined by its two-sided object:

- An exploration of the conscious/subconscious, subjective/inter-subjective psychic reality of the subjects' day-to-day life in situation, in its individual/group, and normal/pathological forms.
- The implementation of formats of inter-subjective encounters favourable to the containing observation of verbal/non-verbal, focal/free associative processuality, and the implementation of the subjectifying meaning of its phenomenology and its conflicting inhibiting and dissociative obstacles.

The clinical methodology for this psychic reality is inseparable from its implementation across all participants involved: it is the inter-subjective encounter and its reciprocal effects that enables the exploration of virtual associations and a meaningful update of their contents.

Of course, this definition of clinical psychology is a skeleton which, to take flesh, must feed on constant ethical reflection and a multitude of modes (diagnosis, results, orientation, individual and group psychotherapies, supervision, expertise, etc.), belonging to the main classic domains of intervention by clinical psychologists: medicine, psychiatry, education and training, employment, justice, institutions, etc. inseparable from their specific cultural context.

Nonetheless, this attempt at a definition aspires to three decisive qualities:

- Highlighting clearly cardinal epistemological 'values' of clinical psychology (1–12).
- Being intrinsically open to inter-disciplinarity, so that the different specialties of care can appropriate this definition of clinical psychology and enrich it with their singularities and critical experience. If clinical psychology is the corporate monopoly of clinical psychologists alone, it betrays the foundations of its mixed identity, its true 'mixed blood'; indeed, the permanent effort towards transition in practises and the multiplication of theories contributes to its history.

- Offering an appropriate framework to engage in its full application in the inter-disciplinary field of perinatal clinical psychology.

2. Perinatal clinical psychology

As an echo to this definition of clinical psychology, the perinatal period can be seen as an open window on the conscious/subconscious, subjective/inter-subjective psychic reality of the subject in a situation of becoming a parent and being born a human.

The process of perinatal parenthood corresponds to a particular amplification of bio-psychic transformations and the inherent processes of associativity/inhibition-dissociation. This parental metamorphosis corresponds, in extreme situations, to a potentially two-faceted crisis: traumatic and/or cathartic.

For better or for worse, parenthood, in transitory manner, highlights the typical elements of the structure and content of the subject's psychic reality. Bydlowski (13), one of the first French psychoanalysts to have focused on perinatal clinical practise after Deutsch, Bibring, and Benedek, speaks of the pregnant woman's 'psychic transparency'.

However this 'psychic transparency', an element clinically and technically central to our definition of perinatal clinical psychology, is no doubt only virtual. The redeployment of its meaning[1] is only a potential that may or may not be confirmed. The greater associativity inherent in the perinatal experience will only provide subectifying opportunities insofar as the internal and external characteristics of the subject's psychic apparatus, the conjugal, familial, and professional inter-subjective spaces, along with the perinatal medical, psychological and social follow-up (maternity, neonatology, paediatrics, perinatal networks, etc.) are or are not favourable to its elaborative containing function. The constraints of this situation will be compounded in case of medically 'pathological' pregnancies and births, and/or in case of parenthood disorders and early parent/foetus/baby disharmony.

The first facet of the object perinatal clinical psychology will thus be an exploration of subjects' subjective/inter-subjective, conscious/subconscious daily psychic reality in a situation of becoming a parent (or becoming a parent once more), of being born human and becoming carers, in all forms—normal/pathological, as an individual, a couple or a group.

The second facet of the object of perinatal clinical psychology is attention towards the scope for parents and caregivers to draw benefit from private, institutional surroundings favouring the metabolisation of verbal and non-verbal associative processuality (somatic, in particular), inherent in the bio-psychic transformations of parenthood and its professional care.

This containing clinical observation of what exists among the different participants and the varied stimulations deriving from the metabolising potential of verbal/non-verbal associativity corresponds to the strategies of primary medical, psychological and social prevention in perinatal clinical psychology. It has meaning and actual existence only in the setting of collegiate collaboration.

Possessing essential common grounds with primary prevention, secondary prevention concerns the offer of modes of inter-subjective encounters targeting parents and caregivers harbouring dissociative-inhibiting obstacles, thus hampering the metabolisation of verbal/non-verbal associative processuality and the attribution of meaning to subjective aspects of its phenomenology.

The inclusion of the 'normal' and the 'psychopathological' in clinical psychology, and thereby primary and secondary prevention, is essential to its perinatal version. As a result, as we will see, the history of perinatal clinical psychology demonstrates, in caricatural mode, how dissatisfactory and distressing it is for parents and their foetuses/babies to see attention directed solely to 'noisy' cases, which will merely receive one-off psychiatric responses.

The extension of the perinatal psychiatry and perinatal clinical psychology to the wide variations in perinatal setting in what is 'normal', and the extension to interdisciplinary caring attitudes more broadly (primary prevention), has been a source of humanisation of births and recognition of masked, mute or visible pathologies (secondary prevention).

Indeed, the 'ordinary' signs of suffering in the perinatal world are liable to be ignored or trivialised, and to evolve underground: the recurrence, still underestimated in the perinatal world, of anxiety, mood and psychosomatic disorders, family relationship disharmony, infant psychosomatic disorders, etc. easily demonstrates this cultural blind spot.

Denouncing the mirage of the conformist medical 'normality' of parenthood (i.e., defensively idealised and a cause of violent exclusions), primary prevention in the perinatal world is justified by a great individual variability in the nature, the content and the chronology of the parental anticipatory maturation in attitudes towards the foetus/baby (14). In response to the singularities of this 'spontaneous' parental prevention, institutional prevention aims to be 'made to measure'. It thus adopts a humanistic approach by offering an interdisciplinary ritual framework, promoting recognition, shared anticipation and explanation for the potential shockwaves of this crisis, oscillating between vulnerability and creativity.

2.1. The epistemological foundations of perinatal clinical psychology

Perinatal clinical psychology is thus centred on the many psychological and psycho-pathological avatars of the encounter of 'being born a human', of 'becoming a parent' and of 'being a caregiver'.

It unites perinatal psychology and perinatal psycho-pathology. The first studies the diversity of the tempered (but broad) formalisations of the process of attunement between the genesis of a subject and the accompanying process of parenthood. Perinatal psycho-pathology distinguishes the pathological patterns that perinatal psychology— in the strict sense—sets out to address in terms of prevention and care. Perinatal ethno-psychiatry (15) ponders about the cultural valency of the disharmony at hand, and its place in an appropriate therapeutic response.

Current perinatal clinical psychology was recently born out of interdisciplinary clinical practise, where psychiatry in the post-partum

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[1] The term 'transparency' could well suggest the illusion of total transparency. This notion would however neglect the irreducible unconscious of both the woman giving birth and those around her.

period plays an essential role. Its history is the best guide to understand the current issues in perinatal clinical psychology.

2.1.1. Genesis

With (16), followed by his pupil (17), French psychiatry was initially innovative in the affirmation of the psychic specificity of the post-partum period (18). But it is only recently that perinatal psychiatry, essentially in Britain in the 1950s, appeared as an institutional practise (19). After a few pioneering experiments, the first mother-baby hospitalisation unit was opened in 1959 at Banstead Hospital in the United Kingdom, on the initiative of adult psychiatrists. This institutional formalisation was motivated by an immediate desire to not separate mothers from their newly born babies. In a population of psychotic mothers, Baker and his team, as early as 1961, reported encouraging retrospective results on the outcome of non-separated dyads from birth, compared to separated dyads. Since then, the dynamics of British perinatal psychiatry have never been refuted. To be convinced of this fact, one only has to mention the activities of the International Marcé Society for perinatal mental health, the first lectureship in perinatal psychiatry in London created by Kumar, or the work by Cox and Holden (20), a Scottish author, famous for a scale designed for the detection of maternal post-partum depression (the EPDS manual), which has been translated into French (21).

In France, it was Racamier who first organised mother-infant hospitalisations at Prémontré psychiatric hospital in northern France. Its concept of motherhood generated reflection in France on the psychic process of 'becoming a mother'. However, it was not until 1979 that the first mother-baby hospitalisation unit was opened at Créteil inter-communal hospital. There are currently 20 full-time hospitalisation units and 11 day-units.[2] The first national meeting of the different psychiatric teams offering joint hospitalisations was held in Créteil on January 29 and 30th 1993.

The contributions by Sutter and Bourgeois (19) and Dugnat (22, 23) provide a good overview of the environment of these units. They were, to start with, defined to host dyads, in which mothers either presented an overt, acute pathology of the post-partum period with its manic/depressive bipolarity (10–15% of parturient women), or an earlier psychiatric pathology or serious personality disorders. Young women with no psychiatric history were also hospitalised with their babies, because of difficulties building their identity as mothers and establishing an attachment favourable to their infant's development.

Perinatal psychiatry was forged around the clinical care of psychotic, depressive and anxiety disorders and suicide risk among pregnant women and during the post-partum period. Today, whilst remaining faithful to this original line of approach, it is in particular extending its spectrum to a wider prevention perspective. Perinatal psychiatry can no longer merely apprehend overt, 'noisy' psycho-pathological disorders. It is opening up to the diversity of parenting dysfunctions—from the most masked to the most obvious, from the mildest to the most severe. This extension is particularly the result of incessant interaction between child and adult psychiatrists and psychologists involved in interdisciplinary clinical perinatal activities.

Perinatal clinical psychopathology is a co-construction between somatic and psychic specialists. For the latter, experience shared on a daily basis between psychiatrists and clinical psychologists who are involved in implementing primary and secondary prevention strategies in perinatal units, plays an essential role. On this point, it is interesting to observe how perinatal clinical practise is an area of great mutual creativity between psychiatrists (child and adult) and psychologists, in stark contrast with other sectors of mental health!

2.1.2. The theoretical foundations of perinatal clinical psychology

In France, the psychology and psychopathology of very young children have been central in the claim for a specific identity of this new domain. On the interface between psychoanalytical and experimental observation, clinical practise in early infancy is progressively opening up to the complexity of the emergence of the psyche and inter-subjectivity. The light cast by theory of attachment of Bowlby (24–26) and by his successors, the reconsideration of the notion of self/other non-differentiation by new-born babies (27) and the essential realisation of the interactive dimension in babies' postnatal bio-psychic development, have all led to innovating questioning on perinatal epigenetics.

In this line of thought, current perinatal psychopathology in France, combines, within an original referential space, contributions from 'developmental psychoanalysis' among infants (28) and research on the process of parenthood and its disorders.

a. Developmental psychoanalysis is situated at the crossroads between studies on genesis and dysfunctions in relation to:

- the skills of the foetus/baby (29) and their interactive epigenetics (30);
- object relationships and attachment (31, 32);
- the emergence of the self and inter-subjectivity (Stern, 1989) (33–35);
- fantasised parent-baby interactions (36, 37) and inter/trans-generational transmission (38).
- triadification (39); and
- the psychosomatic functioning of the infant (40).

b. Following on from theoretical founding research by English-language authors Deutsh (41), Benedek (42), Bibring (43), and Winnicott (44), the psychoanalytical approach to parenthood has a central role (14).

2.1.3. The influence of a few pioneers

French perinatal clinical psychology is greatly indebted to certain clinicians, who were at first isolated, and whose innovating actions opened new paths that have since become emblematic. As a result, the current identity of this practise is to be situated in the wake of the first psychoanalysts first involved in paediatrics and then in maternity and neonatology.

Raimbault inaugurated this approach in the 60s. Royer, who managed the paediatric nephrology unit in the Hôpital des Enfants Malades (Paris), asked him to join his team. This collaboration, which was radically original in those days, led to the creation of the first research unit to include psychoanalysts and sociologists. The '*Clinique du réel*' (45) (real-life clinical practise) was born from this encounter between psychoanalysis and medical paediatrics.

It is clear that the theme of child death here forms an institutional diagonal and an epistemological dynamic. It was the premature

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[2] http://www.marce-francophone.fr

death of children with leukaemia that led Royer to invite Raimbault (46) to his unit. Death was also often decisive in initiating interdisciplinary psychological reflection in the areas of maternity and paediatrics.

In neonatology in the 80s, Soulé (47) at the Institut de Puériculture, explored 'the death-wish in new-born paediatrics'. In the maternity ward, realisation of the little-known devastating psycho-social effects of mourning for a child triggered a slow but definite change in practises from the 90s onwards (48).

However, the propounding of a psychodynamic clinical reflection on motherhood started in France some 10 years earlier, particularly with the inter-disciplinary work by the psychoanalyst Bydlowski in Clamart, in Papiernik's team (49, 50). As for the psychoanalyst Druon (51) working in Relier's neonatal medical unit, she adapted Bick's method of observation (52) to neonatology.

Finally, the local success of these innovating practises and the dissemination of this work *via* publications and congresses provided strong anchor points in favour of triggering awareness of the need for reflection in France on a coherent form of perinatal clinical psychology.

2.1.4. Identity, boundaries and intersections: interdisciplinary, networked and preventive practise

Perinatal clinical psychology is inseparable from its interdisciplinary nature: adult and child psychiatry, gynaecology-obstetrics, neonatology, paediatrics, general medicine, facilities for the care of infants, and the social services constitute the many branches of a network for which the common goal of coherence is its identity.

As has been well demonstrated in its history, perinatal clinical psychology was born from collaboration among a variety of specialists attending parents and foetuses/infants. It was not elaborated by specialists of the psyche alone (psychiatrists and psychologists), but by a panel of professional participants with distinct training qualifications, as illustrated by the reference list of this manual.

Perinatal anamnesis, in the presence of the mother, father and foetus/baby, apprehends human beings in their wholeness, at the cost of the need to move beyond dogmatic divides among caregivers: psychic/somatic, normal/pathological, gynaecologist-obstetricians/paediatricians, prenatal/post-natal caregivers, etc.

The interdisciplinary approach—often fruitful in its conflicting aspects (53)—is certainly very assertively dynamic in the continuum of perinatal activities. The 'somatic' specialists/psychologists-psychiatrists collaboration in teams (including private practise) offer a promise of unity if a common preventive orientation can be materialised. This collaboration in no way means outsourcing from the 'somatic' specialists to the psychologists-psychiatrists all that relates to relationships, affects and trauma. However, this preventive axis can prove fruitful if there is common ground, where each one of the participants can shed light, reflecting their qualification, their history and their subjectivity.

This shared investment will generate first and foremost indirect collaboration with psychiatrists-psychologists. Corridor conversations, exchanges in the staff room, liaising in offices, and meetings (of the Balint group type) will be the basis for the daily metabolisation of the shockwaves of the vulnerability observed. By suggesting, but without imposing, a more or less formalised questioning on caregivers' experiences, this multidisciplinary sharing helps give meaning and combats the defensive operational inertia of the body/mind duality, which has been fed by ambient scientism and biassed training courses. As a result, awareness, which always needs to be re-conquered, of the interactive unity of the patient/carer 'system' is assuredly the basic substrate enabling the efficacy of the containing function of professionals. The more discreet are the disorders in presence and the more central to interpersonal relationship for patients, the more the caregivers' perception of this interaction will be a determining factor.

The direct interventions by psychiatrists-psychologists with families will not replace or compensate for failure of the care initiated. They will follow on from a multidisciplinary reflection that is often worth explaining to parents and babies by caregivers, to prepare for mediation. The psychologists' direct action will therefore be initiated by harvesting a substantial amount of information from professionals who have been made aware of perinatal psychology and psychopathology in parenthood. In return, direct action by the specialist will find its place, as with their colleagues, in constant reference to the common project in the facility. This mutual collaboration in the field is probably a more dynamic argument in favour of a preventive approach than many petitions on principle.

3. The ethics of care and prevention

In the best of cases, medical, psychological and social strategies in perinatal units will therefore follow an open epistemological path, resolutely interdisciplinary, rendered dynamic by research, escaping the insistent medical threat of a 'ready-made' protocol and any predictive, alienating, scientistic, preventive logic, and will take root in a broad ethical community debate on the meaning of the transmission of our paradoxical humanity, a simultaneous source of fecundity and tragic conflict.

The subject at hand is sensitive because the public health strategies of a state-based society to pre-empt and provide support for bio-psychic risks in key periods of the life of its members are certainly an indicator of its political maturity. This is particularly true of the perinatal period, where the meaning and content of the measures adopted in favour of preventing suffering inherent in 'becoming a parent', 'being born a human' and to 'being a caregiver' are particularly subtle political issues.

It is therefore not surprising that the preventive discourse in the perinatal field encounters ideological issues and favours Manichean caricature. It is constantly open to manipulation by idealists ('prevention is going to eradicate all suffering') and to pessimism ('the apple does not fall far from the tree'). Between the utopia of total control over nature and the fatalism of an evil repetition across generations, the creative impetus in perinatality and prevention faithfully reflects the paradoxical human blend of Eros and Thanatos.

3.1. The birth of early prevention

According to pioneers Soulé et al. (54), prevention work with families 'is based on three key ideas: the notion of early instatement, the consideration of the rules of infant mental hygiene, and trans-disciplinary[3] aspects, i.e., the mode of participation of all medical and psychosocial professionals who intervene with families for whatever reason'.

In line with this, prevention is said to be early if it is situated before the child's birth, at the crossroads between primary prevention (reducing the incidence of an illness) and secondary prevention (decreasing its prevalence).

These basic notions are inseparable from those of risk and vulnerability. Risks relate to 'the uncertainty of the outcome of a child's confrontation with environmental or interior stressors' (55)—and to vulnerability: 'faced with the same risks, ultimately not all children present the same disorders (…) personal factors play a considerable role and thus lead to greater or lesser vulnerability'.

The notion of vulnerability (56) illustrates the fact that different people faced with a given risk do not present the same disorders. Vulnerability highlights personal factors. The same blow to three dolls made of glass, plastic and steel will not have the same effect. With the concept of resilience popularised today, this concept merits attentive critical analysis (57–59).

However, more than anything, it is the confrontation with the notions of risk indicators and the emergence of hidden demands that could be the most fruitful and plead in favour of a natural, convergent perspective between social, psychological and medical prevention and provision of support for parents or parental substitutes and children. 'Behind temporary needs', explained Soulé and Noël, there are situations or behavioural modes among parents or children whose experience shows that the children's entourage has difficulty in integrating at once the demands of the child, their own needs and social constraints. These are in fact inadequate, conventional or ill-adapted behaviours. This is why they can be 'warning lights' (55). The perception of these explicit or implicit warning lights starts with the recognition of the multiplicity of expressions of psychosocial distress.

Finally, as these authors stress, when we envisage risk indicators, 'warning lights' and hidden needs, it is important to remember that they are possible signs of distress and in no way obvious symptoms of a recognised pathology. The borderline between well-tempered prevention and an alienating and suspecting investigation is narrow. The challenge for consultants and the whole of the team is to be simultaneously respectful of each person's freedom and potential, and to be able to hear, not only spoken demands for help, but hidden demands too.

According to Soulé and Noël, the difficulties encountered by the caregivers to reach and maintain this balance is an insistent reminder of how 'a certain number of motivations supporting us in this fine idea of prevention are also based on the myth of superpower and man's total control over nature'. If it is not individually and institutionally elaborated, this ideological position 'becomes harmful if, in the name of health, it introduces rigid regulations for education and behaviour' (55).

On a larger scale, social control by the state or by a dominant class, the correction of deviance, normalisation, and the totalitarian deprivation of individual freedom are considered by these pioneers as theoretical and excessive arguments against preventive action, but it is primordial not to set them aside, as they have the advantage of pointing the finger 'at the dangers of doctrines or policies that seek to apply prevention but without assessing the risks and potential excesses' (55).

3.2. Criticism of preventive reasoning: from suspicion to mutually enlightened watchfulness

In a book on perinatal prevention, which showed great maturity in its theoretical, clinical and ethical reflection, Molenat (60) defends a point of view that enables an assessment of the journey undertaken since these pioneering propositions to reach the current state today.

'Classic risk factors are a first level of vigilance, although carrying their own poison: detecting what is psychopathological without necessarily seeking the patient's subjectivity, which means that ready-made responses are needed'. Assessment or screening grids, 'potentially useful in a period of raising awareness, (…) are reused in sometimes unfortunate ways, while a state of mind based on respect for others should prevail'.

According to F. Molénat, if perinatal prevention is not accompanied in the various networks of professionals by a common ethical goal of respecting parental creative subjectivity, it will be synonymous with institutional abuse, thus favouring the deleterious repetitions that it claims to combat. The subjects concerned by this prevention exclusively dedicated to screening for the psychopathological are, in the long run, 'defendants' deprived of attention characterised by 'consideration' and 'solicitude'. Experiments and assessments of prevention programmes show that the essential driving force seems to be the identification aid that a professional at ease in his/her role provides for the families he/she cares for. There is also the particular interest caregivers have in their work, which enables them to mobilise their energy in durable manner.

It is the caregivers' non-elaborated vulnerability, their uneasiness in inter-professional communication and internal power struggles that deprive professionals of a coherent collective containing function that only a network of clinical reflection can allow. If the main question of 'how can a professional, without knowledge of it, repeat what distressed parents have experienced in their own construction?' is not addressed, professional collaboration will keep on building towers of Babel.

By radically drawing away from a culture of repair and opening up towards the anticipation of parental competence, early prevention can find its place in the perinatal world. This objective has a *sine qua non* condition: the professionals' elaborative support, which alone can break off the fatality of repetition of suffering.

Furthermore, it is by being particularly careful with the usual terms of social and medical policies, that prevention will gain its ethical legitimacy: 'This approach evidently discards the usual medical

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[3] We prefer the term interdisciplinarity. As with generational transmission, clinically described by Grangeon as trangenerational or intergenerational, the collaboration expressed by 'inter-' refers generally to mutual exchanges where the unavoidable risk of conflict will lead to debate that will generate symbols and attunement, while collaboration of the 'trans-' type, conversely, will suggest dissymmetrical relationships with little or no elaboration of the antagonisms on its boundaries.

and social policy terms. It is no longer a question of screening for distress or depression in the post-partum period, but about re-introducing sufficient humanity and rigour into practises so that everyone finds their place'.

These relevant suggestions, enriched through the years by day-to-day clinical work, active research and inter-professional training, are directed towards perinatal mental health[4], placing 'the user at the centre of a system to be renovated'.[5]

Finally, the decidedly necessary criticism of preventive reasoning does not justify its outright rejection or the exclusion of its promoters in the name of some potential deviance. On the other hand, it leads us to consider preventive action as inseparable from an on-going, ethical, clinical reflection on caregiving. The encounter of prevention issues (now classic) with, more recently, informed consent for care provision, currently offers innovative and promising food for thought (61).

At the end of a century marked by the triumph of medicalisation, synonymous in France with a decrease in maternal and infantile mortality, with the transfer of birth to a medical environment and to increased social protection, we find ourselves in a period of mutation in familial functioning.

On the one hand, changes in conjugal relationships, the sharp increase in the number of 'recomposed' families, changes in familial and filial rights, and on the other hand, a relative decrease in birth rates, a spacing between births, the frequency in late pregnancies, and the abundance of medically assisted procreation procedures are so many facets of the emergent part of the iceberg, amounting to a relative de-institutionalisation of the family, the classic profile of which has been transformed.

This new dynamic is more globally part of an on-going social mutation, where the shift of power from families and religion to medicine triggers a thinning-out of the former community networks and thus affects the symbolic efficacy of the habitual rites of passage attendant on birth and filiation. In this period of metamorphosis of beliefs and social rites, one of the aspects that is likely to be weakened is that of the establishment of parental identity.

To establish a preventive strategy on maternity wards therefore requires first of all to address the issue of what determines the conditions of existence of symbolic reciprocity in secular rites offered by institutions. Is medical follow-up of pregnancies, births and post-partum the psychic organiser of the parenthood process or, on the contrary, the iatrogenic mediator of alienating scientism? Are there rites of passage that enable—individually and collectively—the violence inherent in this transition to be confronted? Are the usual procedures in maternity wards the object of genuinely informed consent, the guarantee of symbolic efficacy (62)?

4. Perinatal clinical psychology: promising heuristic potential!

To date, the legitimacy of perinatal clinical psychology has been globally secured in the field in France, even if some worrying geographical and institutional discrepancies persist across the country. Even though increased means have been granted to medical and psychosocial perinatal prevention, it could benefit from more assertive adherence by politicians and the public to ensure durable public funding, given the collective stakes involved.

In the future, the medical and psychosocial strategies in maternity wards will need to pursue an open epistemological journey, resolutely interdisciplinary and rendered dynamic by research, sheltering from the insistent medical threat of a 'ready-made' protocol, from a scientistic, predictive, alienating, and preventive logic, and taking root in a broad ethical community debate on the meaning of the transmission of our paradoxical humanity, at once a source of fecundity and of tragic conflict. Quite a challenge!…where clinicians calling on individual or group psychoanalysis have a major role to play in the theoretical and clinical field, remarkable for its heuristic potential.

In fact, in the last 2 decades, innovative clinical experiments in the perinatal period and in early infancy, and the original theories that they have produced, prove extremely fruitful for psychic care in general and psychoanalysis in particular.

As a culmination of this contribution, I would like to focus on the roots of this heuristic force of clinical practise in the area of our origins. Paradoxically, it draws its dynamism from the constancy and ruthlessness of its internal tensions, which call for insistent cross-sectional questioning. Here is an introduction to its main themes:

- Conflict in inter-disciplinary work between 'somatic' and 'psychic' specialists. It is an inexhaustible fountain of this clinical and epistemological prodigality. The oppositions between the various 'somatic' specialists (the frontline between obstetricians and paediatricians and also between 'somatic specialists' of the prenatal and postnatal periods) are less often mentioned, but should nevertheless not be underestimated.

Currently, the growing emergence of care and prevention networking strategies is widening the issues of this dynamic towards the intra- and extra-hospital interfaces—public/private, institutional/liberal. The fruitfulness of these confrontations to smooth over the dividing lines is inseparable from the modes of expression and elaboration of the conflicts in presence. In this context, clinical reappraisals, research-action and perinatal training are fertile laboratories favouring an authentic multi-disciplinary culture:

- A permanent dialectic between the discovery of the amplitude of psychological variations and the psycho-pathological forms encountered. In fact, the 1,001 intermingled metamorphoses of becoming a mother, becoming a father and being born a human are a constant recall of the need to put the nosographic categories into perspective, as they are too geometrical.
- An evolving dynamic of the bodily and psychic boundaries of the processes of profound perinatal transformation: they generate elective attention to the issues of limits and the entanglement of the somatic and the psyche, of the self and the other, of the individual and the group, familial, social, etc.

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[4] Ministère de l'Emploi et de la Solidarité, Ministère délégué à la Santé, report by Piel and Roelandt: 'De la psychiatrie vers la santé mentale'. Submitted July 2001.

[5] Ministère de l'Emploi et de la Solidarité, Ministère délégué à la Santé, Plan santé mentale 'L'usager au centre d'un dispositif à rénover'. Report submitted December 2001.

- A reliving of the conflicts of separation punctuating our lives. ‘Psychic transparency’ is too often idealised: it is, at one extreme, a source of morbid and catastrophic repetitions and at the other a source of maturing radical transformations, but in both cases, always synonymous with crisis.
- A re-edition of the *Oedipus complex*, but first and foremost a confrontation with the reminiscence of the classic archaism described by psychoanalysis, and even more, the archaic archaism that I have attempted to describe with my suggestion of the virtual object relationship in intra-uterine life characterised by the issues of the relationship container/content (53).
- A trivial face-to-face with the limits of our symbolic efficacy confronted with the violence of the suspense, the uncertainty of human genesis in real time around embryos/foetuses/babies, and afterwards in the adults around them. This generational threat is by essence potentially traumatic. It concerns any perinatal journey from the most trivial in appearance, in obstetrical and/or psychological terms, to the most blatantly pathological on somatic and/or psychic level.
- Awareness—not without moral violence—of the determination of the many layers conditioning the quality of the societal, institutional and familial containing functions attendant on these metamorphoses of becoming a parent, being born a human and being a caregiver.
- Finally, perinatal clinical practise is confronted with the origins of a human being, in a given culture, both individual and group-based. The subject belongs to a genealogy that pre-exists and is a singular appearance serving the transmission.

The potential creativity of the perinatal world is incarnated in this bitter, often violent dialectic tension between these two poles of origin and originality. This individual specificity, to express itself, will have to impose itself as a branch on the tree of life where the trunk is the filiation, itself rooted in the Russian-doll succession of generations.

The perinatal world is inhabited by this dual origin of human beings: that of the links—whether clear or unclear—with their genetic matrix, their filiation, their collective heritage, and their culture, and that of the uniqueness of the epigenetics of their being, of their potential originality.

In the end, to defend the preventive potentiality of this period is to seek to promote parental and professional originality and at the same time to underline the insistent threat of repetition, sometimes deleterious, always violent, of the origins.

In this setting gathering users and caregivers, there is a strong convergence between the group formalisation of institutional functioning and what (63) called the ‘group psychic apparatus’ which ‘accomplishes a particular psychic task: that of producing and treating the psychic reality of and in the group’. In fact, any situation in perinatal care belongs to this dialectic uniting the sphere of relationships between the members of a group, and the sphere of the relationships of each member in relation to the group. Indeed, the maternity ward as an institution, the unity of time and place and action, unites all the participants present, users and caregivers. This space–time corresponds to the interweaving of affects and representations of and in the user and caregiver group.

The unity present on the scene of clinical care around our origins is the classic scene of tragedy, unitarian in its temporality, its geography and its action. Given the potential of tragedy to ‘inspire terror or pity’ (definition: Petit Robert) it appears that one of our most spontaneous defence mechanisms as members of a maternity ward team or as researchers is that of isolation, which is achieved by centring only on the reactions of the receivers of messages without taking account of the attitudes and experiences of the senders, i.e., ourselves. The challenge in gaining awareness of this protective positioning is essential, since it enables the establishment of the foundations of an interactive, empathetic common territory between the protagonists, all actors in their particular places in a single tragedy, a ‘situation in which a human becomes painfully aware of a destiny or fatality weighing on his or her life, nature and very condition (definition: Petit Robert)’. Whether destiny or fatality, these are indeed the possible issues to address in this collective representation, which will lead either to a predictive oracle condemning a powerless audience to an inexorable morbid fate, or in contrast to a revelation synonymous with narrative and project (destiny), bringing expectation and life.

The maternity institution, a horn of plenty on the throne of cultural propaganda, finally gives birth to as much darkness as light. Between its celestial showroom and the trivial back-of-the-shop of daily drama (infertility, abortion, miscarriage, perinatal accidents, neonate separation from the mother, prematurity, announcement of a disability, institutional violence, psychosocial distress etc.) the contrast can be enormous (14).

At the centre of the interweaving of Eros and Thanatos, perinatal clinical practise spectacularly stages the encounter between our resilience and our creativity and the dizzying heights of our vulnerability. Perinatal clinical psychology meets the challenge of exploring the somatic and psychic reality of this individual and collective scene, supporting its symbolisation by way of relevant approaches. Quite a challenge indeed!

Author contributions

The author confirms being the sole contributor of this work and has approved it for publication.

Conflict of interest

The author declares that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Three-level containment model of hospitalized adolescents with borderline pathology: a holistic therapeutic perspective

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Borderline personality disorders account for 50% of adolescent hospitalization cases in psychiatry. The severity and psychopathological complexity of these symptoms indicate the need for inclusive models of understanding. Adopting a holistic approach allows for the consideration of not only the patient's environment, but also their position within that environment and their life history. In this article, a model based on the concept of therapeutic containment at three levels is presented. Global containment refers to the mindset and organization of the institution that provides care, which is itself a part of society at a specific time. Local containment focuses on understanding and therapeutic interventions within the immediate social environment of the individual. Lastly, individual containment encompasses the development of independent processes during the course of care. These three levels are integrated in the hospital treatment of borderline personality disorders, forming a trans-theoretical approach.

KEYWORDS

adolescents, hospitalization, holistic model, thee-level containing, borderline personality

Introduction

Adolescent psychiatry is characterized by its richness, variability, and the complexity of its symptoms. Models of understanding cannot afford to be exclusive due to the multifaceted nature of the adolescent experience. The relational function of symptoms prevents us from perceiving the adolescent as an isolated or independent entity. Acts and transitions to action challenge the notion of the adolescent as a purely thinking subject. The development of reflexive faculties at this age contradicts the idea of an all-acting subject. Frequent somatizations remind us that the mind cannot be detached from its bodily anchorage. The need for medication highlights the importance of the relational aspect. Furthermore, the acuteness of family conflicts and traumas underscores the group and systemic dimension of symptoms. The practice of adolescent psychiatry goes beyond integration or holism, as it encompasses the environment and the overall journey of the individual, utilizing various theoretical tools.

Borderline Personality Disorder (BPD) is closely intertwined with the environment. Its etiology is complex and influenced by multiple factors. The biopsychosocial model has gained consensus, emphasizing the continuous interaction between the individual and the environment in the development of BPD (1). Epigenetics research supports this view, particularly stress-diathesis models that consider the interplay of genetic vulnerability and the effects of life experiences (2, 3). In addition to genetic, neurobiological, and temperamental factors (4), the occurrence of adverse events is well-documented in borderline pathology (5–7). From its inception in the late nineteenth century to current classifications, BPD has been characterized by its intricate relationship with life events, under the concept of psychogenicity. The clinical description of borderline pathology establishes a strong connection between patients' symptoms, their environment, and the relational events that shape their turbulent journey. Borderline individuals are in a constant state of interaction with their environment, which significantly influences them. The inherent fragility of the self makes them porous to every element in their environment, almost like mirrors. Additionally, healthy adolescents and patients (adolescents or adults) with BPD share specific characteristics in terms of psychic functioning and dynamic conflict movements, such as issues with autonomy and dependence, increased sensitivity to intrusion and abandonment, cognitive functioning with a Manichean tendency, and self and relationship instability, among others. Adults with BPD seem to extend an unresolved adolescent process indefinitely (8). Thus, there exists a continuum between adolescence, borderline psychopathology, and society. Our clinical work aims to establish a continuous process of differentiation and gradation, taking into account these various aspects in a holistic manner.

BPD is a complex psychiatric condition characterized by a wide range of symptoms, all revolving around the theme of instability and oscillation. This includes oscillation between anxieties of intrusion and abandonment, instability of the self and relationships, impulsivity, feelings of rage and emptiness, as well as anxious and mood symptoms. Clinical presentations of BPD vary, but are often marked by acute episodes that may involve anxiety, self-mutilation, suicidal attempts, dissociative episodes, and more. These moments of clinical severity, with their potential prognostic implications, are the primary reasons for hospitalization in psychiatry, accounting for 30 to 50% of hospitalized adolescents (9). The coexistence of these noisy but episodic clinical moments with underlying and enduring psychopathological processes necessitates a two-level analysis: nosographic and psychopathological, or categorical and dimensional. In this regard, it remains useful to distinguish between BPD as defined by DSM criteria (characterized by overt symptoms such as self-destructive behaviors, uncontrollable anger, impulsivity, etc.) and borderline functioning in the psychoanalytic psychopathological sense, encompassing a borderline state/personality organization (involving defense mechanisms such as splitting and projective identification, anaclitic dependency relationships, ego porosity with intra-psychic conflicts between intrusion and abandonment, etc.) (10). It is possible to experience one without the other, and often, but not always, adolescence reveals a borderline crisis with symptoms that have been evolving since childhood. Thus, we refer to an "adolescent borderline crisis," which may complicate an earlier borderline functioning (11), reserving the term "BPD" for situations that confirm a categorically observable functioning persisting into adulthood. The therapeutic perspectives outlined in this article apply to the entire field of borderline pathology.

These moments of clinical and prognostic severity, where the vital prognosis is at stake, often lead to the hospitalization of patients with borderline symptoms. Self-destruction, which is a challenging symptomatic dimension to address in psychiatric consultation or psychotherapy (12), frequently necessitates hospitalization. For adolescents with borderline symptoms, the duration of hospital care can be divided into three distinct periods, with some overlap in timeframes, each lasting approximately one week.

The first period of hospitalization focuses on global containment. This involves providing reassurance to the adolescent and limiting their internal or externalized agitation, anxieties, and resulting disorganization. Achieving global containment in the hospital requires various measures, including architectural considerations (department structure), organizational aspects (care planning), and institutional factors (team mindset and ensuring team safety, based on the clinical assessment of the patient).

- Once global containment has been established, the second phase of care involves local containment. During this stage, the focus is on the adolescent and their immediate relational environment, with the goal of understanding the factors contributing to the deterioration of their psychological state. This includes identifying the psychological events that have led to the current situation, which may have been challenging and unchanging for weeks, months, or even years. Additionally, the general context of the adolescent and their typical level of functioning is evaluated. This phase of the work entails exploring emotions and representations, analyzing and mentalizing experiences, and fostering individual and collective sharing.

- Finally, the last phase of care, individual containment, combines a gradual increase in patient autonomy to prepare for discharge and the development of a more active role in their own care. Therefore, the objective is to facilitate the patient's departure from the hospital by finding a satisfactory compromise that considers the needs and desires of all parties involved, including caregivers and parents. This is the time when the young person reconnects with friends and teachers, organizes their return to school, and coordinates their ongoing psychiatric care. The separation from the care team at the time of discharge is smoother when the focus of the therapeutic exchanges has shifted from symptoms to the sharing of positive and constructive experiences.

These three periods of care correspond to three distinct levels of therapeutic intervention, drawing from various theoretical perspectives that complement each other. They are organized according to the global, local, and individual levels, ranging from the broader containment to more focused interventions. Given that psychotherapy models may not be accessible in all geographical areas and that patients with severe disorders may have difficulty engaging in regular psychotherapy, our model aims to integrate different therapeutic approaches, which can be particularly beneficial for these challenging cases. These approaches serve as the foundation for achieving the three therapeutic objectives in BPD that align with the three levels of care organization: 1. stopping self-destruction, 2. identifying and addressing the factors contributing to the crisis, and 3. recognizing and connecting emotions to their triggering events to regulate them through bodily, psychological, and relational processes.

Global therapeutic containment

To prevent self-destruction and facilitate the patient's personal growth and interpersonal connections, a crucial resource is the supportive institution. In order to fulfill this role effectively, the institution must possess specific characteristics, as outlined by the theoretical framework of institutional psychotherapy.

A. Institutional care mindset

Over the last 70 years, theorists of psychiatric care have discovered, with institutional psychotherapy, the extent to which the state of mind and the application of an active and organized collective life is a fundamental institutional prerequisite for receiving and caring for a patient (13). Although these practices were thought of with adult patients suffering from psychotic disorders in mind, certain principles are completely relevant in adolescent psychiatry, particularly with patients with borderline symptoms (14).

In the hospital, death distress is the daily life of the psychiatrist who treats borderline pathology. These workers are the ones to whom adolescents convey their suicidal behaviors, their negative feelings toward their entourage, and toward the caregivers who are in contact with them. The psychological work often revolves around understanding the stakes of the individual's situation and their family history concerning death. It always deals with a group, in this case a family, and the way in which death distress is dealt with within this group. A quarter of adolescents (24.6%) with DSM-5 criteria of BPD have a parent who has attempted suicide (15). The psychiatric work is also linked to the analysis of the history, the narration, and the re-appropriation of the individual and collective parts of the fear of separation and death. The group device constitutes a therapeutic space in the spirit of "transitionality", as developed by Winnicott. It is a space in which patients who are in a state of crisis are able to express the most archaic and stormy strata of their thoughts and it can open or reopen "the way to a sufficiently existential foundation" (16), especially when the singular therapeutic relation between the patient and his psychiatrist remains unsuccessful for a real "psychic accommodation" (17). However, before all this work intending to give the patient's story a narrative can begin, first, one must consider protecting the patient against acts of self-destruction, and thus ensuring their survival. At the very beginning of the care lies the necessity to stop the escalation of suicidal behaviors and the escalation of danger and fear.

The psychiatrist overseeing the patient's treatment must effectively manage the adolescent's behavior to ensure their safety and wellbeing. Initially, measures such as restricting the patient's freedom of movement, removing dangerous objects, and ensuring a secure environment are necessary steps in providing care. However, it is crucial to recognize the limitations of these restrictive measures and to determine when it is appropriate to gradually reduce them, allowing the patient to regain their autonomy and sense of responsibility. As the patient's clinical condition improves, typically within a few hours or days, the care team can work toward preserving the patient's privacy, freedom of movement, and encouraging their active participation in decision-making. It is important to strike a balance between maintaining safety and avoiding excessive control, as an excessive focus on absolute safety and eliminating all risks can actually be counterproductive and reinforce suicidal symptoms. By excessively exerting control, caregivers inadvertently reinforce the patient's pre-existing feelings of powerlessness and contribute to a sense of intrusion that further intensifies their fears. Therefore, it is essential for caregivers, in addition to the psychiatrist, to navigate the concept of risk and help adolescents gradually overcome their risk-avoidant thinking (often characterized by all-or-nothing reasoning) that is inherent in borderline psychopathology.

B. Organization of institutional care

In the tradition of F. Tosquelles and H. Simon, the institutional organization of care aims to address three threats to patients' wellbeing: inaction, an unfavorable hospital atmosphere, and the prejudice of patient irresponsibility (13). Hospitalized patients engage in various mediation activities such as visual arts, rock climbing, relaxation exercises, meditation, singing, and circus arts. These activities provide patients with an active role within a group, fostering interdependent relationships and collective creativity. The focus shifts from the patient's illness to their strengths, aligning with Jean-Baptiste Pussin's approach in the eighteenth century. These workshops offer patients an opportunity to explore aspects of their personality that provide personal pleasure, fulfillment, and unique experiences (18). During these experiences, patients are both differentiated from others and connected to them, facing the complex challenge of maintaining individuality while remaining connected. The mediated group setting facilitates symbolic expression without the therapeutic relationship becoming overly threatening. The use of artistic media allows patients to "play and give form" to their relationship with the clinician (19). As caregivers also participate in workshops outside their expertise, these mediation sessions promote a more horizontal caregiver-patient relationship. It encourages a co-construction of care rather than a hierarchical medical approach that can be perceived as powerful and intimidating. The attachment insecurity often observed in patients with borderline pathology complicates the provision of help, as they may simultaneously seek and reject it. Moreover, this defense mechanism evolves in response to the actions and words of others, creating an idealized or demonized image of oneself and others, further amplifying the perceived power of medicine and caregivers (the power to heal magically or harm diabolically). Therapeutic efforts are more effective when adapted to include the patient as an active participant in their healing process, thereby alleviating the overwhelming sense of powerlessness experienced by patients who have endured numerous traumatic experiences.

A weekly multidisciplinary synthesis meeting is organized to gather insights from various caregivers, serving three fundamental functions. Firstly, it facilitates the exchange of information about the patient that goes beyond their symptoms and daily care, including details the clinical significance of which may not yet be fully understood. Secondly, it provides an opportunity for caregivers to express their emotions and delve deeper into the emotions evoked and projected by the patient. Lastly, it serves as a collective platform for developing the care project. The fragmented nature of borderline identity presents challenges in providing care, as different fragments of the patient's personality are projected onto different caregivers throughout the course of hospitalization, referred to as the "transferential constellation" (20–22). Establishing trust in care is difficult for caregivers, and for the patient, investing in a relationship with a care team is less intimidating than investing in a relationship with a single individual. Similarly, caregivers quickly encounter their own powerlessness in providing "good care," and teamwork serves as a support system, enabling shared experiences and the ability to navigate this feeling without jeopardizing the therapeutic relationship. By identifying the nature of exchanges, emotions, and psychological conflicts that unfold during the synthesis meeting, the care project begins to take shape. The collective unconscious plays a role, and the sharing of representations fosters the emergence of new ideas. Collective intelligence promotes the appearance of these emerging processes, which unfold in unpredictable ways, even though the therapeutic setting influences their probability of occurrence (13). These processes confirm that the representations generated by the group are greater than the sum of their parts. To facilitate the emergence of the patient's true self from their symptoms, the fragmented elements must first be gathered, re-associated, and reorganized. Cleavages and projections are the challenges that compromise coherence and continuity, and caregivers work to address them. Through the synthesis of the elements of the transferential constellation, they aim to give it shape and assist the patient in constructing a coherent sense of self (10). This work creates a space for the patient to acquire or reclaim the subjective position that may have been lost beneath the symptoms (13).

C. Recognizing the societal factors influencing self-destructive behaviors

We have observed that clinicians have long established connections between borderline symptoms and the environment. However, the significant increase in suicidal behavior among adolescents since 2020 has driven research to identify psychopathological correlations between societal factors and the psychological wellbeing of adolescents. While it is not to say that society creates and solely explains psychological disorders, it can be considered that society influences the expression, development, resolution, and acceptability of these disorders to varying degrees. Psychological disorders can manifest differently depending on the societal context in which they arise, and they may also occur more frequently if society lacks the protective barriers that typically contain fear, anger, and sadness. In this sense, if we conceptualize institutional containment as that of society, the state, a group, or collective organization, it enables proactive intervention in the best interest of individuals. Furthermore, gaining a comprehensive understanding of the relationship between these societal factors and patients' symptoms is a crucial prerequisite for any therapy. On one hand, recognizing these phenomena at the collective level allows for their discussion with the patient and their family, who will experience them in a unique but not isolated manner. On the other hand, this holistic understanding of the environmental effects enables the establishment of prevention policies that have individual-level consequences and, if effective, reduce the need for individualized care. Therefore, preventing suicidal behaviors in adolescents, including those related to borderline crises, requires an understanding of these mechanisms that encompass both the adolescent themselves and the institutions responsible for their care. It is also important to avoid interpreting all problematic phenomena in adolescents solely from a medical perspective, as this would underestimate other crucial causal factors that are societal rather than medical in nature. Examples of such factors include social inequalities, territorial policies, child protection, and the impact of screen usage on cognitive functions. Hence, it is the responsibility of caregivers to bear witness to the various factors contributing to their patients' disorders, with the goal of addressing them collectively at a societal level in parallel with individual treatment.

Among the various societal factors, the COVID-19 pandemic has been identified as an immediate explanation for the significant increase in suicidal behaviors among adolescents. The restrictions imposed during the pandemic had a profound impact on the global population, particularly hindering adolescents who have a strong need for exploration beyond the family environment. This period of withdrawal and fear of others not only led to an increase in suicidal issues but also manifested in symptoms related to fear and control, such as phobias, anorexia, and somatization. It is important to note that even before the pandemic, the isolation of young people was recognized as a societal problem (23). However, the quality of family support and social integration plays a significant role as a prognostic factor in suicidal problems. The fragility of such support, which varies from country to country, contributes to the sense of isolation in young people who rely more on virtual relationships rather than in-person connections. Since 2020, alongside the reinforcement of social distancing measures, adolescents have experienced a notable increase in symptoms of "derealization" and dissociation, which are characteristic symptoms of borderline personality disorder (24). These symptoms reflect an overwhelming feeling of detachment from oneself and reality, where individuals no longer feel present in their own bodies. While these symptoms are severe, they can be alleviated through the use of words, self-care, in-person relationships, and shared activities. Many solutions exist outside the realm of medicine to address these issues. In some countries, the suffering experienced by adolescents and the timing of their symptoms, when correlated with the school calendar, reveal a significant amount of school-related pressure. This pressure, combined with frequent cases of bullying in the school environment, contributes to the adolescents' sense of inadequacy. It highlights the challenges they face in hierarchical and competitive systems that prioritize conformity over personal development and lack sufficient opportunities for encounters and cooperation. Additionally, instances of neglect and abuse, both within and outside the family, exist and are often inadequately addressed in terms of prevention and care. These situations are now highly medicalized in several countries, and the lack of medical resources to address the increasing rates of suicidal acts further hinders caregivers from fulfilling their role as a last line of defense against suicide. Providing a welcoming, reassuring, and present presence over time and ensuring continuity of support are crucial aspects of this role, but are often impeded by resource limitations.

At a family level, the deployment of parenthood is affected by professional and personal stress, which is further exacerbated by the lack of adequate social and community support. This hinders the collective effort in raising a child, leaving isolated and inadequately supported adults to shoulder the responsibility alone. However, isn't it said that "it takes a village to raise a child"? The pervasive experience of death and distress during the pandemic has underscored the crucial importance of psychological resilience and the need for a collective social project that goes beyond mere survival. In times of adversity, and in life in general, the psychological suffering arising from a situation is often more closely tied to the absence of meaning (shared values and objectives) rather than the situation itself. Without a sense of purpose, any stress-inducing reality can become traumatic. Young people are acutely aware of the stakes involved in climate change and the preservation of life, and their sense of isolation grows when adults fail to protect the planet and the natural world. This sense of immobilization and denial creates a feeling of dissociation between the recognition of a major threat and the failure of adults to safeguard the future. Furthermore, the actions of young people are sometimes criticized and belittled. However, engaging in civic activism is one of the most effective ways to foster social integration and combat anxiety and powerlessness. For society, youth represents a wealth of fresh perspectives, an impetus for action and creativity, and the primary transformative potential, provided they are given the necessary opportunities to fulfill their role.

Outlined here are the societal issues that can influence the psychopathology of destructive behaviors, prompting us to consider collective action for the prevention of mental health problems. Such efforts would significantly support institutions in maintaining their therapeutic role in alleviating patients' suffering. However, it is crucial not to oversimplify borderline pathology as a purely psychic or purely social phenomenon, but rather to recognize its multifaceted nature. Examining the immediate relational environment of these individuals helps us comprehend how the interaction between the individual and society unfolds, with the ultimate goal of informing therapeutic interventions.

Local therapeutic containment

A. The assessment of the relational environment

Given the chaotic environment typically experienced by adolescents with borderline symptoms, the therapeutic management of these patients requires a comprehensive assessment of their relational history. It is crucial to address environmental challenges in addition to therapeutic interventions, often involving medical and child protection measures, as solely focusing on therapy may prove insufficient or ineffective. During hospital treatment, significant time is dedicated to conducting medical interviews with both parents present. These interviews serve to explore the family and patient's history, including psychiatric, medical, and legal aspects, as well as assess the difficulties and level of parental distress. Therapists actively encourage the psychological and relational engagement already initiated by the psychiatric crisis and hospitalization. In this way, they initiate treatment of borderline symptoms by incorporating the patient's environment and evaluate the family's capacity to adapt to change, engage in self-reflection and questioning, and collaborate with professionals, all of which are important prognostic factors for adolescent borderline pathology.

Early adversity in borderline pathology has a significant impact on neural and cognitive functioning associated with the stress axis through epigenetic mechanisms (25–27). It also disrupts attachment patterns, which in turn contribute to the development and perpetuation of borderline symptoms (2, 28). Research has shown that severe childhood maltreatment is prevalent among adult patients with BPD, and similar findings have been observed in adolescents (29). This maltreatment includes traumatic life events, abuse, and dysfunctional parenting. Examples of traumatic events include early separations, childhood loss, bereavement, and severe threats of loss such as parental suicide attempts. Maltreatment often involves emotional abuse, including child denigration, as well as sexual abuse (found in 30–90% of cases and 28.6% of borderline adolescents) (15, 30), and neglect. A significant number of hospitalized young people face challenges in their immediate environment, where their basic needs beyond material necessities are often overlooked. These needs include shared time, attention, emotional care, consistent rules, and support.

Parenting dysfunctions are characterized by controlling behaviors and a decrease in the level of emotional care. Problematic interactions, such as emotional invalidation, suicidal threats in front of the child, repeated conflicts revolving around the child, and loyalty issues between parents, are also common (1, 15). These adversity factors play various roles in the psychogenesis of borderline pathology, with their accumulation associated with the severity and early onset of symptoms (30). Furthermore, their variable combinations contribute to distinct symptom patterns and clinical presentations (15). The cumulative effects of these factors contribute to the complex trauma experienced by adolescents with borderline pathology. Such trauma involves early and prolonged experiences that often involve attachment figures. Like other forms of complex trauma, the consequences include not only dissociative trauma symptoms but also profound identity shifts and attachment disorders (31).

Hospitalization frequently provides an opportunity to identify or observe evidence of ongoing or past abuse. Caregivers play a fundamental role in establishing an educational framework during hospitalization, while recognizing the importance of involving educational services regularly used during treatment. The aim is to create conditions that promote a calmer and more supportive parental environment for the adolescent. This approach includes supporting both the parents and the adolescent in addressing their limitations, while respecting each party's subjectivity and needs. In most clinical situations, caregivers emphasize the importance of meeting the adolescent's emotional needs in an age-appropriate manner. This includes providing a reliable and predictable environment, supporting autonomy in a gradual manner, setting parental expectations based on the child's abilities, and ensuring shared time, attention, emotional care, support, and a structured framework with clear rules. Observing these parental difficulties often prompts clinicians to establish close collaborations with child protection services, which may vary in difficulty depending on the country. Such collaborations are necessary to break the cycle of abuse or neglect. In addition to the responsibilities of child protection services, psychiatric professionals systematically consider and address these relational factors to establish an environment that promotes the adolescent's recovery.

B. Considering the systemic dimension of symptoms

The presence of adversity should be considered at both the individual and family levels (32). Exploring the family history in the presence of the adolescent can be therapeutic from the beginning. Admission to the hospital often unveils a suicide attempt by a parent or other family member during the medical interview. Conflicts between the adolescent and their parents are frequently linked to relationships between the parents and members of their family of origin, such as siblings or parents. Parents often describe complex relational issues with siblings or parents that have led to major conflicts and unresolved life-and-death matters. In these interviews, the adolescent realizes with relief that they are not the root cause of the problem as they had believed, but rather that their symptoms represent a variation of it. Traumatic relational elements leave their mark differently across generations and time, and the adolescent's symptoms provide a gateway to accessing these hidden crypts and secrets that are passed down (33, 34). The adolescent, who carries the symptoms, becomes the person who enables the family to access care and is valued by the care team for this role.

Working with family conflicts is akin to working with trauma. Patients and parents recount life situations that have remained unresolved, painful, and immobilizing. During these narratives, emotions emerge and become associated with the words and shared representations. Expressing emotions, connecting them to representations, and sharing them with others are the essential steps to move beyond a traumatic state. Conducting three to five one-hour interviews with the patient and their parents together often results in a significant change in family dynamics. The crisis situation, with its deep family imbalance, simultaneously encourages mobilization and resilience. Although hospitalization may be a moment of suffering and imbalance within the family unit, it also opens up possibilities for a new and better-adjusted equilibrium.

It is widely accepted that the therapeutic management of BPD in adolescence requires significant involvement of the family (35, 36). The exploration of family dynamics begins with the adolescent's symptoms. The family is encouraged to establish hypotheses about factors that are somehow related to the symptoms or the adolescent's overall distress. Starting from the symptoms, an associative thread is woven, leading back to the history, the "pre-history" of the adolescent, and various family members, particularly addressing the knots, conflicts, and impasses within this collective history. The suicide attempt, a central symptom of borderline pathology, represents not only a refusal to live but also a message directed at emotional support, indicating a resignation to jointly find a solution to pre-existing distress. It signifies the loss of hope in the relationship and a desire to detach oneself from others during this overwhelming distress to regain some control that has been completely lost and to preempt an unbearable fear of abandonment. The suicide attempt also reflects an anger that cannot be directed toward someone who would not hear or support it. Borderline anger represents an inward turn of aggressiveness, and the role of the psychiatrist is to explore with the patient and their parents which feelings could not be identified and expressed to their family and friends.

In some cases, a suicide attempt can also reflect an intense feeling of self-disappointment, often associated with an excessively demanding ego-ideal. The adolescent may feel that they are not living up to the expectations placed upon them by their family, such as achieving academic success. This self-disappointment is a sign of introjection, where the adolescent internalizes the demands of society, often conveyed through their parents. Adolescent psychopathology shows that what we commonly refer to as "school failure" becomes a matter of life or death when it is accompanied by a sense of rejection. Unlike a healthy individual who may respond to the failure of their ideal with modesty, patients with borderline pathology experience it with depression. The externalized ego-ideal is gratified through action (37). The adolescent perceives that their failures could bring shame to their family and friends, or that society excludes them. Consequently, they contemplate death as a means of protecting themselves. What the adolescent truly seeks is not merely a diploma, but rather the unconditional love and acceptance of their parents, as well as a sense of belonging in society. They long for the certainty of being loved regardless of their abilities and for gaining collective esteem regardless of their social status. They shoulder the immense weight associated with these expectations, to the extent that it becomes a matter of life or death, particularly when there are no supportive structures in place. This leaves the patient trapped in their anxieties without the possibility of overcoming the issue of "success" and developing self-actualization (38). Adolescents who struggle academically and contemplate death are expressing that their need for emotional safety outweighs their need for scholarly success.

Family relationship work is highly beneficial in the care of adolescents, but it often evokes feelings of guilt that need to be identified and addressed. This guilt stems from the awareness of the affective power of interpersonal relationships on oneself and others, which patients may alternately minimize or exaggerate through a process of splitting. One way to alleviate overwhelming guilt is for parents and doctors to sometimes seek genetic or biological explanations for the pervasive nature of the disorder. However, parents are relieved when they overcome the difficulties associated with guilt and are able to identify the unresolved wounds from their own history that resonate with and contribute to their adolescent child's difficulties. They appreciate the opportunity to comprehend the emotional and transgenerational issues that have hindered their relationship with their child, as it empowers them to influence the relationship at both an individual and family level. Medical interviews with parents involve psychoeducational work on the diagnosis, characteristics, and evolution of the disorder, which also helps alleviate parental guilt (39). Multifamily groups and individual therapy can be utilized to teach parents how to overcome an invalidating parenting style (40). Additionally, siblings may be included in consultations as well, as borderline symptoms affect the entire family, and siblings often remain silent to avoid exacerbating the problem.

Moreover, the process of finding meaning and understanding the functions of symptoms is not contradictory to the search for a biological or genetic cause, or the implementation of solutions that may include medication. It involves attributing complexity to situations and moving beyond binary reasoning—cause and effect—to embrace circular and systemic reasoning. After a suicide attempt, patients often identify one or two causes for their actions, while their parents identify two or three causes, which may not necessarily align. However, after a few weeks of hospitalization, caregivers can deconstruct a dozen causes or, rather, conditions that contribute to the emergence of suicidal thoughts or behaviors (38). No single factor alone determines the outcome, but the convergence of these factors creates the possibility of a suicidal event or symptom. Once these conditions have been identified, caregivers work collaboratively with patients and families to develop solutions, whether they are psychological, physical, or involve adjustments to the adolescent's daily life (e.g., treatment approaches, living arrangements, school accommodations, etc.). However, caregivers often encounter resistance if they propose quick-fix solutions. The concern or distress may shift from one issue to another, and simple adjustments may not be suitable until the underlying relational needs are understood and the solutions are co-constructed. The stability of the family system can challenge the therapeutic alliance, and this reality is openly addressed with families as a process linked to the complexities of therapeutic responses. The conditions that contribute to the emergence of symptoms reflect a range of unmet needs that could not be expressed in any other way, but are now being explored to address them proactively before they manifest as symptoms. The adolescent assumes the role of the person who enables the identification of these dysfunctions and is valued for fulfilling this role. The more the family can establish themselves in this process, shifting their focus from the adolescent's symptoms and reorganizing their functioning, the better the prognosis for the patient. This family work occurs on two levels: the internal reality level, which involves addressing individual and group unconscious conflicts, and the external reality level, which involves concrete interventions to adapt the educational framework to meet the adolescent's daily needs.

Systemic family therapies are commonly prescribed for the majority of families following hospitalization, when there is a capacity for psychological mobilization and the therapeutic work is deemed significant. However, not all families of adolescents with borderline pathology possess the minimum degree of cohesion required for this type of therapy. Additionally, a highly projective group dynamic can render this work unfeasible. Other specific therapeutic interventions have been developed to support parents or the family, focusing on particular areas such as psychoeducation based on Dialectical Behavioral Therapy, mentalization processing, or trauma-focused approaches (39, 41–46). Family interviews conducted during hospitalization can already address various dimensions, including reducing educational inconsistencies, reviewing parental and adolescent expectations, identifying and interrupting role-reversal dynamics, reestablishing protective roles, decreasing levels of control and criticism, promoting emotional validation of the adolescent by their parents, identifying issues related to relational distance (separation or intrusion), recognizing paradoxical communications, addressing conflicts, and highlighting transgenerational repetitions.

C. Psychoeducation

At this stage, the issue of diagnosis arises, either at the request of the parents or with the aim of clarification, therapy, and psychoeducation. While diagnosis is not a routine practice in adolescents, mainly due to the risk of pathologizing identity formation during this critical developmental period, it can still be valuable in many cases. Its utility should be evaluated on a case-by-case basis. Diagnosis can guide the treatment team during and after hospitalization, such as determining the need for a day hospital program or requesting disability recognition. However, its primary benefit lies in helping the patient and their parents organize their responses to the symptoms with a prognostic perspective. Experiencing a significant stressor like a suicide attempt or self-injury in a child necessitates that parents project themselves into the future and avoid succumbing to despair. However, the diagnosis of BPD inherently implies a prognosis extending into adulthood, which may not align with the duration of the most overt and disruptive BPD symptoms. Therefore, when diagnosing BPD in adolescence, it is crucial to clarify the precise nature of the observed symptoms, including emotional, relational, behavioral, and identity-related instability. Additionally, underlying psychopathological elements should be addressed, such as increased dependence, excessive reliance on relationships at the expense of one's own needs, and diminished self-esteem and confidence in one's abilities. It is important to explain to parents the diagnostic challenges in balancing the concept of a personality disorder with the scientific predictive validity of the disorder. Statistically, approximately three-quarters of individuals with BPD experience significant symptom remission within 2 years. This has been supported by studies conducted by Bernstein et al. (47), Levy et al. (48), and Miller et al. (49). Furthermore, the proximity of certain features of adolescent crisis to BPD symptoms may have led to an overestimation of the disorder's prevalence, which is actually estimated to be around 3% instead of the previously suggested range of 6% to 18% (47, 50–52). Borderline behavioral symptoms can overlap with symptoms present in other disorders, such as eating disorders, substance use disorders, or adolescent depression. The expression of these symptoms may be more prominent at the physical level during this developmental stage, including self-injury and impulsive suicide attempts (47, 53). The co-occurrence of these symptoms may have contributed to the overestimation of BPD. In such cases, the duration of borderline-type symptoms may be shorter than that of the co-occurring disorder, leading the concept of a personality disorder to hold less relevance for the patient, parents, and healthcare professionals.

For these reasons, it is more appropriate and cautious to use the term "borderline crisis in adolescence" (11, 54), which captures the symptomatic behavioral presentation while incorporating the scientific elements of predictive validity for the disorder at this age. This approach allows for differentiation and compatibility between categorical and dimensional perspectives, both of which are necessary for medical evaluation. The term "borderline crisis" describes the presence or absence of symptoms according to international nosology, while "borderline personality functioning" describes the underlying traits that should be targeted in long-term therapeutic interventions. This diagnosis also enables the naming of the clinical state in real-time without having to wait for a year or more (as required by DSM criteria for diagnosing BPD in adolescence), thus facilitating specific and prompt interventions during crises. Furthermore, in certain situations, it is particularly useful to inform parents that their adolescent does not have a psychiatric diagnosis or does not meet the criteria for BPD, especially when the environment's perception of illness undermines the patient's speech, desires, or choices. This requires clinical and therapeutic sensitivity to reassure the family and help them identify precise transgenerational traumas in order to establish more realistic expectations for their child's future. Regardless of the diagnosis, it is crucial to avoid exclusively focusing on the adolescent with BPD (36). They should be seen as part of the solution rather than the problem, and the entire family system needs to modify roles and behaviors to foster cohesion. Psychoeducation can be incorporated alongside symptom defocusing, even after a diagnosis has been made, to ensure the importance of transgenerational work is not overlooked despite the need for psychoeducation.

D. Restoring a balanced relationship with others

Attachment theories, such as the strange situation paradigm, have taught us that one-year-old infants respond to brief separations from their primary attachment figure in specific ways (55–57). When an adolescent is hospitalized, they encounter strangers and experience multiple separations from and reunions with their parents. From an attachment perspective, this situation is also relevant. Although the temporality differs, it allows for comparisons, and the precise analysis of the patients' attachment patterns (secure, ambivalent, avoidant, or disorganized) is essential, given that attachment insecurity characterizes 96% of hospitalized adolescents (58). Thus, psychiatric hospitalization at this age becomes both an opportunity to assess attachment security and a therapeutic tool to experiment with separation in a controlled environment, with familiar reference points and with outcomes that differ from experiencing "fear without a solution," as described by Main and Hesse (58).

Hospitalization also provides an opportunity to mobilize Internal Working Models [IWM, (59, 60)], which are mental representations of the relationships established with attachment figures. These models incorporate beliefs about oneself as worthy of existence and deserving of help, as well as beliefs about others as competent and available when help is needed. Many hospitalized adolescents report a tendency to cope with major life difficulties on their own. In reality, their symptoms maintain a "forced" relational closeness with attachment figures. A central objective of hospitalization is to enable them to integrate the possibility of creating new relational modalities, including seeking support from others. This can be facilitated through interviews, art therapy, medication, shared meals, or informal exchanges with the treatment team. The idea is to help the patient recognize that their current relational patterns, which may have been adaptive in the past, have become dysfunctional. For instance, adolescents with predominantly avoidant attachment are encouraged to reach out to caregivers when they have even the slightest need or difficulty, despite their inclination to minimize and rely on themselves. They are asked to make 5 to 10 relational attempts, such as requesting treatment as needed, asking for time to talk, or discussing their day. These attempts are systematically addressed and analyzed in individual interviews with the doctor and a nurse. Through the gradual re-establishment of secure connections in a safe environment, patients with avoidant attachment can regain or develop the capacity to reflect on their internal states, identify their emotions, and take care of themselves. This process often requires evoking multiple situations in interviews to construct secure representations, as patients initially resist due to their typical avoidant belief of "No one can help you better than yourself" (61).

From a complementary psychoanalytic perspective, listening to and observing hospitalized patients, even at a young age, reveals significant repetitions that unfold in the transference toward the caregivers. These repetitions reflect the patients' past experiences with others. Each patient guides the caregivers toward a specific relational position, which is important to identify, name, and share. This process involves accepting identification with this position and then shifting away from it, opening up creative possibilities for care instead of sterile repetitions of destructive relational patterns. Similar to the role of psychoanalysis in institutional psychotherapy, understanding the psychoanalytic dynamics of the patient's case is not characterized by silence, neutral listening, or verbal interpretation (21, 22). It allows for the elaboration of institutional responses, prescriptions, actions, attitudes, and everything that contributes to the therapeutic context and the patient's care project. However, in adolescence, these relational mechanisms become more complex as transferential processes unfold directly with caregivers and indirectly through parents. These dynamics need to be analyzed, particularly in terms of areas of splitting and projection. These therapeutic procedures resemble Transference Focused Therapy adapted for adolescents (62, 63), which aims to identify internalized dyads of idealized and persecutory object relationships during psychotherapy. These dyads are characterized by oscillating, incompatible, and unconscious self-other representations. The therapist seeks to track their activation and alternation by identifying with whom these movements are "staged" or conveyed (63). These processes need to be analyzed and clarified within the relationships in which they occur (with parents, team members, or peers) before or alongside their direct exploration in the transference itself. Negative therapeutic reactions (64) may manifest in patients with borderline pathology, either as a temporary deterioration of their clinical state during hospitalization or as resistance to leaving the hospital after a few weeks of care. It is the team's responsibility to analyze the relational dynamics and psychic modifications involved in the therapeutic relationship, considering the patient's past experiences and their implications for meeting and separation. With proper attention, the discharge process can become a represented and surmountable separation.

Finally, the transferential aspects within the institution cannot be understood without considering the transference-countertransference dynamics between patients and the institution itself. This work occurs during weekly caregiver-client meetings involving all patients and the care team. In line with the tradition of institutional psychotherapy, individuals can only be effectively treated if the institution itself is also treated (13, 18, 20, 65). These therapeutic moments provide an opportunity to question the framework and purpose of care, as well as the patients' and the group's appropriation of them. They also serve as a therapeutic moment for the institution, allowing for ongoing reflection. The agenda of these meetings is determined in real-time, based on the group's and caregivers' current issues. Improvisation creates space for the emergence of affects and primary processes, which are the foundation of collective work. With borderline symptoms, the psychic space spills over into reality (66, 67), and the patient group becomes a privileged context for the projection movements of each patient. These moments enable the gathering and containment of projective identification, persecution, and disorganization arising from daily events in the unit. Moreover, the use of peer identification is a powerful therapeutic process in adolescence. Treatment within the group setting provides an opportunity to develop reflexivity and mentalization (68, 69). It also facilitates the collective construction and clarification of treatment objectives, stages, and encountered difficulties. Collective elaboration and containment work occur during group sessions and in post-group discussions among caregivers.

In summary, this work at the interface between the patient and their relational environment aims to address the relational aspects of borderline symptoms and enable the patient to differentiate between projection and reality in their daily interactions with others. This process of differentiation allows for a re-anchoring of these patients in reality.

Individual therapeutic containment

This work on different levels of containment is crucial for the patient to establish a sense of individuality within the complex system of care that evolves and adapts over time.

A. (Re)discovering a differentiated identity

During hospitalization, individual medical interviews are conducted regularly (though not necessarily on a daily basis). These interviews involve a nurse caregiver who has a closer connection to the adolescent's daily emotional experiences compared to the doctor. The nurse caregiver shares mediation times, meal times, and informal moments with the patient, in addition to the interview sessions. The main objective of these interviews is to restore the connection between emotions and mental representations, addressing the limited capacity for mentalization often observed in patients with borderline pathology (70). Adolescents with borderline pathology frequently struggle to make sense of the emotions they experience and the sudden changes in their emotional states. During the interviews, concrete relational situations are analyzed to understand how daily events impact the patient's emotions. Borderline affective instability is characterized by an exaggerated reactivity to seemingly insignificant triggers. However, these patients often have a history of significant emotional disruptions from a young age, and their emotional turmoil during adolescence is often linked to situations that hold more significance than they initially appear. These situations may include the fear of separation, relationship difficulties, or the feeling that their needs are not being met. Patients tend to anticipate negative events and perceive the world as threatening, which can lead them to generalize their experiences and interpret even minor situations as highly significant (28). During the interview, a thorough analysis of these relational situations allows the patient's conscious experiences to be compared with their unexpressed emotional and bodily reactions. This process aims to construct new shared representations and promote a deeper understanding of the patient's crisis. It is crucial to look beyond environmental factors and identify the underlying psychic events that triggered the patient's crisis. While the patient's environment may contribute to potential risks, understanding how the patient experienced specific relational events provides valuable insights into the mechanisms of the crisis and helps outline the stages of psychological mobilization.

The individual interviews also serve to promote the subjectivation of the adolescent in the days following family interactions. During these interviews, adolescents have the opportunity to express their perceptions, understanding, and feelings regarding their parents' stories and emotions. This process helps enhance their perceptual abilities by incorporating the perspectives of the caregivers themselves. When these shared perceptions align, they are grounded in a common reality and reinforce the patient's confidence in their own perceptual capabilities. However, when perceptions differ, it opens up a discussion about relational reference points and potential causes underlying the emotions of family members, thus exploring the family history. This allows the patient to practice inferring their own and others' mental states and develop mentalizing abilities, which are key targets in mentalization-based therapies (69–72). Furthermore, the development of insight and reflexivity abilities, which involve self-reflection, is an important aspect of the patient's prognosis. By engaging in representative work on internal and interpersonal psychological processes, the patient can subjectively appropriate their experiences and consolidate their sense of self. The therapist tactfully points out paradoxes, contradictions, oppositions, and inconsistencies in the patient's discourse, allowing the adolescent to become aware of the internal conflicts at play and to develop independent thinking. The analysis of daily aspects in the here and now serves as a starting point for reflecting on past and recurring experiences. These therapeutic techniques share similarities with aspects of Transference-Focused Therapy, which focuses on therapeutic affective movements, and Dialectical Behavioral Therapy, which addresses Manichean functioning (63, 73, 74). During hospitalization, the level of support in the here and now is intensified for patients with more fragile selves. For patients with less severe borderline symptoms, the work conducted in these interviews enables them to acquire the psychological tools necessary to establish a new relationship with the world based on an understanding of the factors that contributed to the crisis and its emotional manifestations. However, for other patients, whose capacity to connect emotions and representations is too overwhelming, the therapeutic work will focus more intensely on reactivated past relational experiences in the therapeutic relationship with the caregivers during hospitalization. In this context, the meaning and explicitness of daily therapeutic interventions become crucial in facilitating this process of psychological connection.

Medical interviews provide a crucial space for discussing traumatic events, their causes, and their consequences, allowing patients to share their experiences with another human being. This opportunity for emotional sharing is supported by empathetic caregivers. Adolescents often experience isolated traumatic events, such as episodes of physical violence. However, the complex nature of trauma in borderline pathology makes it difficult for patients to recognize the abnormality of their experiences. Caregivers play a vital role in guiding patients regarding social norms (e.g., the abnormality and illegality of sexual intercourse between a 13-year-old and an adult) and addressing the emotional needs of young individuals as they develop (emotions, needs, constraints, desires, projects, etc.). Through the sharing of the patient's experiences, a sense of temporality is established within their narrative. This temporal framework is essential for addressing traumatic symptoms, as individuals often experience a loss of temporal landmarks. Distinguishing between the patient's individual story and the stories of their parents or family members also enables temporal and spatial differentiation. Through discussing life experiences with a compassionate listener, subjective appropriation of these experiences occurs, allowing for differentiation from the experiences of relatives. This psychological work contributes to reducing dissociative phenomena and establishing a stronger grounding in reality. The clinician assumes the role of a secondary environmental response, providing representation and validation that was lacking in the primary response. This identification, influenced by Ferenczi's concept of a primitive failure of the object's response to the subject's experiences, highlights the importance of later inscription of experiences that were not adequately processed. As Green (75) suggests, "the response through countertransference is the one that should have taken place on the part of the object". Consequently, the psychological work surrounding these experiences helps reduce the risks of repetitive traumatic patterns and psychotic disintegration, aiming to restore the patient's relationship with reality (76). Crisis hospitalization also serves as a significant moment for the transformation of childhood images. The borderline crisis presents an opportunity to confront the images constructed since childhood with the reality of the parents as they are during treatment. It allows the adolescent to engage in a seesaw process, as described by Kestemberg (77), involving a transition from disappointment to self-conquest.

B. Body therapies

After the initial phase of hospitalization, which may be characterized by a "honeymoon" period, patients often enter into conflictual therapeutic relationships with their caregivers, feeling that they are not receiving sufficient help in managing their symptoms. Additionally, due to decreased mentalization and insight capacities in patients with borderline pathology, their symptoms tend to manifest behaviorally and bodily, with self-aggressive behaviors, such as self-injury, being common. These self-injurious behaviors serve as visible expressions of underlying instability, which the patients themselves often struggle to identify. This presents another opportunity for shared representation work, which is conducted on a daily basis with the nurses and discussed in meetings with the doctors. Nurses propose hypotheses and draw connections between internal emotional experiences and events the patients have encountered during the day or week. To aid in this process, nurses utilize various tools to help patients identify and regulate their emotions. One such tool is the establishment of a written emotional report, which assists patients in labeling the fluctuations of their physical sensations and emotions throughout the week.

In the development of patients with borderline pathology, attention may shift toward both parents and one of them as a means of self-protection and/or protecting others. Early experiences of concern for a parent's compromised health, fear of separation, or other sources of worry can lead to heightened observational abilities directed toward the environment, while self-observation capacities may be diminished. Green (78) described how these early modalities, through the paradigm of the dead mother complex, affect the child's personality, resulting in difficulties perceiving and interpreting affects within oneself and the development of alexithymia. Bowlby (60) also proposed that early experiences of relational withdrawal from attachment figures have an impact on development, a finding supported by several studies linking insecure attachment and borderline symptoms (79–81). Adverse experiences can lead to distinct perceptual modifications, with relational withdrawal experiences correlating with hypersensitivity in facial emotion recognition and intrusion experiences associated with decreased emotional perceptual accuracy (82). In all cases, attention to the internal world is diminished, likely due to difficulties accessing early parental mirroring functions (83). During hospital treatment, the emotional regulation work conducted with caregivers enables patients to shift their attention to their internal world. Caregivers provide them with tools to apprehend and regulate their emotions once they are identified and understood. While putting emotions into words is an important tool, it is not the only one. Breathing exercises, such as abdominal breathing, cardiac coherence exercises, and mindfulness, are fundamental. Caregivers, nurses, or body therapists may also offer massages or other body treatments that promote a sense of calm. In extreme cases, patients may find comfort in being wrapped in blankets or sheets, lying in a hammock, or resting on cushions in a soothing environment. Body care is also provided through various activities, such as sports, yoga, dance therapy, relaxation exercises, circus arts, or socio-aesthetic care, offered on a daily basis.

Caregivers and therapists who organize these activities serve as conduits for accessing affective and bodily integration, aiming to construct psychic representations from threatening, absent, or dissociated bodily perceptions. This "space outside-the-self" can only be accessible to the psyche if it can be represented, and pre-representations registered in the body can only be expressed through interaction (84). The experience of affect arises through the shaping of a relational schema. The development of a sense of self, which is lacking in patients, emerges from interactions with others and involves both psychological relationships (gaining meaning, sharing representations) and bodily aspects (tonic support, affective holding, handling). Similar to how an infant relies on the reflexive ability of their early environment, adolescents depend on the capacity of their environment to engage relationally, affectively, and bodily to support their tonicity (85). The internalization of bodily and relational elements, connected through processes of symbolization, allows adolescents to form an envelope that contains psychic, affective, and sensory contents, similar to Anzieu's (86) concept of the Skin-Ego. Just as infants, during the separation that inaugurates mother-child differentiation, maintain their unity by symbolically linking perceptions and sensations to psychically survive the absence, adolescents also rely on this process to navigate separation without exhausting themselves.

In addition to body therapies, alleviating borderline symptoms may involve pharmacological treatments, although their effectiveness is somewhat limited according to randomized controlled trials (87, 88). Medications are primarily prescribed based on the identified clinical dimensions present in borderline pathology or its numerous comorbidities, such as anxiety, self-aggression, major mood variations, and insomnia. Second-generation antipsychotics with anxiolytic aims are often preferred for a limited duration of a few weeks or months (89). The choice of medication depends on the patient's psycho-corporal profile, considering the specificities of adolescent metabolism that may result in more side effects compared to adults receiving the same dosage, particularly metabolic syndrome (90, 91). The appropriate timing for medication prescription is also considered, with the need to establish a therapeutic alliance and address any resistance or concerns regarding medication, as the fantasy of dependence can be particularly significant for patients and parents. Therefore, addressing this dimension in parallel is crucial to improve compliance. Additionally, the presence of various addictions often requires regular consultations with specialized addiction professionals during and/or after hospitalization.

C. Autonomy toward the exit

In the final phase of care, the focus is on promoting autonomy as the adolescent prepares for discharge from the hospital. The patient gradually engages in out-of-hospital activities, initially accompanied by staff and eventually on their own in a safe environment near the hospital. This process allows them to re-establish autonomy and learn to manage stressful situations independently, while still having the option to seek support if needed. Weekend leaves also provide opportunities to practice autonomy and ensure the safety of both internal and external spaces. During this stage, it is important for the patient to distinguish between manageable and more challenging situations and symptoms, and to develop a safety plan that outlines coping strategies and available resources for each level of stress. This plan may include activities such as listening to music, reading, drawing, engaging in breathing exercises, going for walks with a trusted person, reaching out to caregivers or close individuals, practicing body calming exercises, taking prescribed medications, or contacting the referring psychiatrist or the hospital. As discharge approaches, the focus shifts to preparing the adolescent for greater autonomy outside of the hospital setting. This includes gradually reintegrating into daily life, spending time alone at home, engaging in social activities with friends, and taking trips to the city. It is essential to consider the stability of the patient's clinical state at discharge, as borderline symptoms may require multiple hospitalizations to achieve regulation. Therapeutic strategies can be employed to minimize the risk of suicidal acts, such as implementing a watch and call system or planning for sequential re-hospitalization if necessary.

If medication has been part of the treatment, this phase allows the adolescent's input to be considered when adjusting the treatment plan, as they will ultimately decide whether to continue taking medication on a daily basis after discharge. As the patient identifies their sources of difficulty, they take progressive steps toward moving forward and finding meaning and purpose in their internal states. Discharge signifies a planned and announced end to the hospitalization, representing a positive change in the therapeutic relationship, which is often different from the initial patterns experienced by hospitalized patients. It is also a time for parental guidance, managing attenuated symptoms that still carry a risk of recurrence. Parental guidance involves learning to manage risk while providing a containing function for the child.

At the end of hospitalization, in addition to the significant reduction in borderline symptoms, the therapist evaluates various factors, including the evolution of self-esteem, the level of hope for both the adolescent and their parents, the insight of each individual, and the ability to engage the family group in action. These objectives align with the aims of successful psychotherapy, which include building confidence, exploring conflicts, regulating emotions, and managing separations and reunions. However, the temporal dynamics of hospitalization differ significantly, and the emotional intensity experienced over the course of a few weeks plays a crucial role in shaping the psychological impact of these experiences.

In summary, the last stage of care focuses on the patient's own capacities and their ability to translate and regulate their internal states. The goal is to support them in resuming their life journey with increased serenity and equipped with the necessary skills and resources.

Conclusion

The quote by Marcus Aurelius, "May I be given the strength to bear what cannot be changed and the courage to change what can be, but also the wisdom to distinguish one from the other," encapsulates the therapeutic approach for adolescents with borderline symptoms.

In pursuit of the holistic objective, the three levels of containment, crucial in the treatment of adolescents with borderline symptoms, are interconnected and intertwined. They progress sequentially over time, with global containment being planned and organized prior to the patient's admission, followed by local containment during the hospitalization period, and individual containment becoming more prominent toward the end of the hospitalization. At each level, the preventive and therapeutic objectives are inherently linked to the analysis of the contributing factors. The hospital objectives align with those of the outpatient psychotherapeutic model for patients with borderline symptoms. These objectives include establishing a structured framework, involving the parents, actively engaging therapists with the adolescent, addressing resistance factors associated with the disorder, and restoring coherence and psychological continuity (92). For the most severe cases, additional layers of containment are implemented, requiring strong cohesion and collective creativity within the care teams. It would be beneficial in the future to compare this holistic model with singular models to quantitatively assess how different theoretical approaches complement each other within a comprehensive perspective. Overall, this model aligns with the current global health trend, which emphasizes transnational health issues, determinants, and solutions for enhancing overall wellbeing. By providing a detailed description of the individual's environment, it enables better preventive and therapeutic interventions to be implemented.

The therapeutic work aims to assist these patients in achieving a more balanced position within their environment. This involves fostering a sense of differentiation from others (psychic separation) while maintaining emotional connection in a secure manner. The goal is to support them in representing the adversities they have experienced and developing protective and coping strategies to navigate these experiences. It also involves reactivating the process of autonomy-dependence that is inherent to adolescence. Furthermore, the aim is to help the family identify their challenges, difficulties, and available individual and collective resources. Ultimately, the objective is to help the adolescent achieve inner harmony and find peace within their environment.

Author contributions

Conceptualization: MR, RR, and MC. Methodology: MR, LB, KK, and JB. Clinical investigation: LB, JB, and MR. Original draft preparation: RR and MR. Reviewed by: MC, JB, LB, and KK. Supervision: MC. The manuscript was written by MR and approved by all co-authors. All authors contributed to the article and approved the submitted version.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Joint family consultations for psychiatric inpatients with severe eating disorders transitioning to adulthood: psychoanalytic design of a therapeutic setting

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While family work is acknowledged as relevant in the care of eating disorders (EDs), not much literature has explored it in the period of transition from adolescence to young adulthood (16–18 to 30 y.o.). Yet this period is of significant importance in the prognosis and evolution of EDs; but its particular stakes require specific therapeutic settings–especially for inpatient EDs. In this paper, we start from the paradoxical observation that some families refuse this type of work in its usual form, with a family-dedicated therapist, and require to only exchange with the psychiatrist in charge of the treatment plan. We use a psychosomatic-informed psychoanalytic approach to shed light on this refusal as a latent denial of the contribution of family dynamics to the current symptom, and an unconscious tendency to stick to a dependency-laden family scheme. We then explain the conception of a specifically dedicated therapeutic setting, designed to address this specific type of resistance, offered to families as a therapeutic compromise designed to give them a specific position in the care of their child. In our joint therapeutic consultations, family dynamics are addressed on the basis of exchanges regarding treatment and in particular feeding. While such exchanges start from medical considerations, the therapeutic couple (psychiatrist-psychologist) uses them to address the parent and patient expectations underlying the symptom. We propose to call this act "inscription"; it enables a separation from the underlying dependency-oriented family scheme, while stressing the importance to care for associated parental anxieties.

KEYWORDS

inscription, therapeutic setting, eating disorders, family, denial, model, psychosomatic, psychoanalysis

1. Introduction

1.1. Eating disorders in late adolescence and young adulthood: complexity and family care

Eating disorders (EDs) encompass a vast array of complex psychiatric pathologies present in adolescents and young adults, such as binge eating, *anorexia nervosa*, etc. The complexity of these pathologies comes from their notoriously multi-factorial nature – factors encompass culture, family, somatic and biological conditions, and psychiatric comorbidities (1). The absence of a univocal causality requires a multi-faceted approach to care.

Amongst these factors, therapeutic paradigms have stressed the need to address family dynamics, thereby including families to the treatment plan [cf. e.g., Rienecke and Le Grange (2)]. Offering them punctual psychotherapeutic consultations or regular therapy sessions is standard practice for adolescent inpatient EDs in French hospital Units (3, 4).

Less literature has stressed the importance of family work in the transition to young adulthood (16–18 to 30 y.o.). Yet we believe, like (5–7), that this period is of importance in the prognosis and evolution of EDs, and requires specific therapeutic settings. This is especially the case for serious forms, requiring inpatient care.

In this respect, we have been struck by a recurring feature encountered in many inpatient clinical situations in this age range (the vast majority of EDs in the adult Psychiatry Unit at Strasbourg University Hospital). A significant number of families refuse to explore the family dimension of the symptoms in a dedicated family consultation with a psychologist, in spite of the seriousness of their child's condition–which leads others to accept every therapeutic option. Instead, when proposed such a (state-funded) dedicated consultation, they say that it is not that they do not want it, but that since it is now the hospital's job to cure their child, they see no reason to discuss the situation and its family effect.

Drawing on psychoanalysis – an acknowledged therapeutic paradigm in EDs [e.g., Thompson-Brenner et al. (1)], we consider this refusal as a form of specific family resistance against the anxiety induced by the offer to explore family dynamics, and *a fortiori* subsequent family changes. To work with this subset of families, we devised a specific therapeutic setting adapted to this resistance: that of joint family consultations (JFCs), jointly led by a psychiatrist and a psychologist-psychoanalyst. The goal of this paper is to sketch out the design, therapeutic stakes and effects of this specific setting and its use, by explaining how these recurrent clinical situations led us to model it. The data presented in this paper did not need ethical clearance, as it was a secondary account of our experiences in healthcare.

2. Institutional context

In late adolescence, EDs are triggered by the challenges of approaching adulthood: leaving home, facing important choices (e.g., higher education), etc. This differs from earlier adolescence: what is now at stake is the possibility to withstand the perspective of psychologically separating oneself from one's family environment and become autonomous. This has direct implications for ED inpatient care: caretakers no longer envision hospital stays as temporary breaks from family life, as they mostly are during adolescence. Hospital stays in our unit are conceived as an intermediate step before the beginning of (young) adult life, where patients separate themselves from the hospital and establish a new relationship with their families to start an adult life.

But when patients initially come to the hospital after a consultation request, and meet with the psychiatrist overseeing the treatment plan (MR), medical concerns are in the forefront–especially body mass index (BMI) issues. They are understandably the main focus of both patients, families and caretakers. Yet, as mentioned above, a growing consensus is that efficient EDs care requires family work: thus, in the case of potential inpatient admission, the psychiatrist quickly proposes to meet with the parents. Most of the time, she then offers them to plan a consultation (generally with the patient) with the unit's dedicated psychologist/family therapist (OP), to discuss the family effects of the situation; such consultations typically explore the difficulties associated with becoming psychically autonomous, for both families and patients. This offer is presented as an addition to all standard inpatient care: daily psychotherapeutic sessions with a psychiatrist (other than the head of ED Unit), exchanges with another psychiatrist about treatment adjustment (medication, somatic follow-up, stay duration, etc.), and a wide range of institutional care: individual or group dietary support, physical and bodywork and somatic therapies, group psychotherapy, dance lessons, etc. In addition, we have the opportunity to interact with the hospital's Clinical Nutrition Unit. Therefore, all therapeutic effects of family work take place within a dense web of activities; and it is never meant to replace individual psychotherapy, but it facilitates it (more in Conclusion).

3. From manifest refusal to latent common denial

But a significant subset of parents refuse to meet with the unit's family therapist (OP) to discuss the family effects of the situation: it is not that they do not want it, but since it is now the hospital's job to take care of their child, they see no reason to come discuss the situation and its family effect. This is particularly interesting, since EDs – especially *anorexia nervosa*, with its extremely slim bodies – tend to put many families in a constant state of preoccupation (8): quite a few immediately and gratefully accept dedicated consultations with a specialized psychologist. How are we to understand this?

While this clinical observation matches Minuchin et al.'s (9) psychosomatic remarks on change-averse families in *anorexia nervosa*, we understand it exclusively through a psychoanalytic lens: as a resistance of the family group (parents + inpatient + siblings, potentially), expressing a mechanism of denial of the family dimension at play in EDs. This refusal, and the subsequent claim that it is now the hospital's task (and not theirs anymore) to take care of the patient (including feeding them), should be understood as an unconscious projection, onto the Psychiatry Unit, of the family's representation of what it is to take care of someone. Considering how families tell us that they are now solely concerned with how the Psychiatry Unit takes care of their child, we contend that there is something specific in their unconscious representation of such care – and most likely, that it is a relation of exclusive dependency [cf. Corcos (3)], whereby the child would receive from the parents everything they need. While often feeling hurt because they believe they have failed to provide their child with everything they need, parents nonetheless transpose the same exclusive relational scheme onto the caregiving team, without realizing that it is its exclusive character that should be re-examined and questioned: exclusive dependency is both impossible in reality, and an obstacle to separation and individual self-realization (This accounts for the two extremes often to be found in these families: those who try and provide everything, and those who renounce because it looks impossible – both share a representation of exclusive care).

This understanding relies on the psychoanalytic concept of denial, first laid out extensively by Freud in 1938 (10), subsequently elaborated by Klein (11) and Bion (12–14), and recently developed by Ogden (15) under the name of projective disavowal. Gabbard (16) stresses its relevance and sums it up like this: denial, or disavowal, is an active process whereby denied representations or affects are projected onto someone else, in actual interactions, part of which take place at an unconscious level. The task of the therapist is to "process and contain them," i.e., understand them as such, in order to signify to the patient (or the group) what they have thus unconsciously set aside.

We believe that when refusing a dedicated EDs consultation with the psychologist, families project onto the psychiatrist who offers it their unconscious representation of caring for a child *qua* exclusive dependency. Hence the shift of the burden of care, so to speak: it was all on the family, and it is now all on the hospital.

To specify the "interpersonal pressure" exerted on the psychiatrist, and what "process and contain" means, we expand on Braunschweig and Fain's (17) concept of "community of denial" [cf. also (18)], following recent work (19, 20). Braunschweig and Fain (17) stressed that in situations of denial, projection is an attempt to persuade the therapist to adopt one's perspective and jointly disavow the representations one seeks to ignore ("common" denial). In essence, these families tell the psychiatrist (a doctor) that they agree to come and see her to receive medical information about the treatment course, as long as family dynamics are not brought forward as a therapeutic dimension *per se*. This unconscious invitation to denial generally emanates from the whole family (the child partakes in it), but is most often expressed by parents. Therefore, to "process and contain" it means that the psychiatrist, in charge of both mental and physical health, needs to acknowledge the denied representation of parental care that underlies it – one of exclusiveness and dependency, which she is encouraged to take on as a parental substitute. In terms of psychoanalytic technique, she needs to acknowledge a specific form of emerging transference emanating from the whole family group and especially the parents, whereby she is envisioned as a potential accomplice of a specific, unquestioned understanding of care. This emerging transference thus represents the main therapeutic indication for our specific therapeutic setting.

4. Offering a joint therapeutic setting

Following a line of thought first laid out by P. Marty and M. Fain, which OP's work develops, we decided that to preserve potential psychotherapeutic effects, the psychiatrist overseeing the treatment plan should offer a creative compromise adapted to the families' initial resistance. On the one hand, her stance should implicitly acknowledge families' denial-based resistance to address their relational dynamics. On the other, she needs to communicate that it is not possible to fully separate medical/somatic work from family dynamics: there can be no "splitting" between the two (10, 16, 17) since family dynamics are affected by EDs, and affect them in return. On the basis of OP's previous research, we devised a specific therapeutic offer: that of a joint therapeutic setting, with the psychiatrist (MR) and the unit's specialized family therapist (OP). This offer's goal is to communicate that optimal care for the patient will respect their resistance to some extent (the psychiatrist's presence means that it will still be possible to talk about medical care, incl. feeding/eating); but that making care optimal would *also* require to explore family feelings (this is embodied by the presence of the family therapist).

The psychiatrist tries to enact an "inscription" [cf. e.g., Chervet (21, 22)]. In our use of the term, in institutional and family group dynamics, "inscription" refers to an intervention going, in part, against families' resistance, by shedding light on a representation that they expect the psychiatrist to deny along with them – i.e., their exclusive representation of parental care. In therapeutic technique, this inscription amounts to a specific handling of the family's emerging transference, whereby the psychiatrist is cathected as a parental substitute mirroring their representation of care. While they expect her to mirror this representation, she draws on this transferential expectation to surprise them by expressing that she cannot take this role alone, and instead needs someone to help her. She thus communicates two elements that embody a different caregiving stance:

1. there should be no individual/exclusive caretaker(s). This is a dismissal of their projected expectation of an almighty caretaker/caretaking team.
2. care is not only about manifest behavioral parameters (weight, caloric intake, etc.). From the Unit's point of view, care includes more psychological elements: at least, taking into account how everyone feels about the situation. Underlying this is a technical premise: elements regarding medical care can be handled as a therapeutic medium to address family dynamics.

The goal is a re-mobilization of the family: caregivers are not almighty, they need the parents to contribute to the care, as it involves a psychological parameter – their relation to their child. That is, ED treatment is a 3-tier challenge: patient/family/caretakers. This first inscription leaves a trace potentiating further therapist interventions during joint consultations, by delineating an alternative, non-exclusive model of care which does not require the patient to remain dependent upon the caregiver.

5. The technique of joint family consultations: inscription as separation

Offering such a compromise-based therapeutic setting is not psychoanalytically orthodox, since it leaves room to discuss medical and eating-related preoccupations while seeking to address elements pertaining to intra- and interpersonal psychological dynamics. It could thus at first sight be understood as feeding families' resistance. We believe that on the contrary, in cases of strong family denial, it helps induce authentic therapeutic effects by handling their specific transference (projected expectation of an almighty caretaker). Yet, this should not be confused with family therapy *per se*, which is more constrained in terms of rhythm, participants and therapeutic aim [cf. e.g., Robert (23) or Berger (24)] since it is focused on restructuring relationships within the family – while our consultations are more punctual and seek to increase individual autonomy with respect to family dynamics.

In transition to adulthood, the therapeutic aim of these JFCs is to enable parents and patients to withstand the patient's separation and self-realization. Such self-realization requires, as a necessary condition, feeding oneself well enough (not too little, not too much, etc.): that is, (1) not to depend on an external figure to regulate or even control eating behavior, as well as (2) accept to feed on something which does not come from one's parents. Thus, leaving room for families and especially parents to express their feelings about the concrete care of their child is a springboard to explore how families can tolerate transitioning out of a relationship of exclusive dependency. But this requires that psychological separation be explicitly expressed in the course of a therapeutic exchange or sequence, which will echo the first inscription carried out by the psychiatrist.

For the sake of brevity, we give one frequent example: parents often tell the psychiatrist they are preoccupied because in the Unit, their child will not be offered the food they are used to; surely they will not like it as much, so how are they to be cured from their disorder? This manifest question is underlain by latent issues, through what psychoanalysis calls unconscious overdetermination. For example, in the later course of family consultations, some families admit they are concerned their child could be poisoned by what comes from the outside, while others say they are afraid the child could not control him/herself (and would always need someone to regulate their intake, like an infant). Sometimes, they say: the child is not gaining enough weight–you must feed them the wrong way. Both exemplify unconscious motives to maintain an exclusive relationship with the family: they are types of an exclusive, dependency-laden family relationship, split off from the explicit concern about feeding, and unconsciously projected onto the child who, in return, identifies with them. It is these projections that JFCs wish to disentangle, by showing that the child's behavior and discourse to some extent echoes that of parents, and that both play a part in the current symptom.

Drawing on the presence of the psychiatrist, parents thus use manifest issues to express their rivalry with the Unit. These exchanges call for an inscription of the main psychological stake underlying EDs and their medical care: psychological separation between parents and child/patient – that is, a separation of individual perspectives through an explicit re-attribution to parents of what they project onto the child, and vice-versa. From a technical point of view, in the aforementioned type of situations, the therapeutic couple (psychiatrist-psychologist) often proceeds as follows. The psychiatrist shifts her stance out of the medical level, and stresses that from the perspective of the treatment plan, medical parameters (BMI etc.) are but one level of information, and that the stakes are more general and revolve around their child's personal evolution, and how they feel about it. This paves the way for the psychologist to mention that maybe their worry about feeding means that it is they who suffer from being separated from their child – a suffering which they project onto him/her, and to which the child might conform to show them how irreplaceable they are, thereby maintaining dependency.

In these situations, we respectfully tell parents that their anxieties should be discussed without any form of prejudice, as they play a role in the situation, which cannot therefore be fully ascribed to the child. Upon hearing this, the patient is put in a position where s/he is both relieved of parental projections (s/he can refer to this later), while parental separation anxieties can be acknowledged; at the same time, s/he becomes authorized by a third party (the therapeutic couple) to express personal heretofore unexpressed concerns, while evolving away from such projections.

6. Conclusion: deferred and distributed therapeutic effects

The trace of this 2-step inscription often appears in a deferred manner, when the Unit's individual psychotherapists tell us in a subsequent staff meeting that the patient now expresses new desires (with which e.g., their parents might not be happy); or that, more generally, they associate more freely about future perspectives. Of course, in other instances, we see that the patient's relation to their parents evolves, often by witnessing how previous inscriptions affect their exchanges.

Finally, such a therapeutic setting is devised as a potential preparation for, or adjunction to, individual therapy; but by no means as a substitution. Indeed, some parents start a therapy after JFCs, when realizing their child's symptom echoes their concerns.

We even sometimes decide to see parents without the patient when we feel they need a temporary dedicated space to express concerns that should remain out of the child's conscious awareness, to help them transition toward individual therapeutic work.

Data availability statement

The original contributions presented in this study are included in this article/supplementary material, further inquiries can be directed to the corresponding author/s.

Author contributions

OP: Conceptualization, Investigation, Methodology, Supervision, Writing – original draft, Writing – review & editing. MR: Conceptualization, Investigation, Methodology, Writing – original draft, Writing – review & editing. NB: Conceptualization, Writing – review & editing. JP: Conceptualization, Methodology, Writing – review & editing. MD: Conceptualization, Methodology, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Alcohol and psychoactive substance use in a cohort of children followed by child protection in France

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Background and aim: Many studies have investigated the association between maltreatment and substance use in adulthood.

In this study, we sought to explore the association between substance use during adolescence, diverse forms of child maltreatment, and psychological symptoms within a cohort of individuals under the purview of child protection services in France.

Method: The dataset was culled from a retrospective, population-based study encompassing minors and young adults aged 0 to 21, who were under the care of child protection services. Specifically, we conducted a comparative analysis between minors exhibiting substance use (N = 72) and those without such use (*N* = 776).

Result: The odds ratios predominantly illuminated a significant correlation between Substance Use and the manifestation of self-destructive behavior (OR = 4.35; CI 2.02–9.59), as well as aggressive behavior (OR = 5.75; CI 2.87–11.84). Univariate analysis also hinted at an association between SUD and suicidal ideation (OR = 3.52; CI 2.1–5.90).

Conclusion: Children in France who are in the care of child protection services and who use psychoactive substances are at greater risk of dropping out of school and of having other psychological symptoms. It is important that the public authorities take account of these results in order to adjust the care given to these minors, who often do not receive psychological support.

KEYWORDS

substance use, child protection, mental health, youth & adolescence, maltreatment

1. Introduction

In France, alcohol and substance use are major public health problems. In the general population, initial consumption appears around the age of 13.3 years for boys and 13.5 years for girls (1). It is estimated that 7% of deaths the population aged 15 and over are related to alcohol consumption (2). Alcohol consumption starts very early, and is often associated with the use of other substances. Indeed, nearly 10% of 17 year olds are regular multiple users of alcohol, tobacco or cannabis (3). This issue appears to have an international dimension, as a recent

Australian study showed that alcohol was the main cause of death and hospitalization among young Australians (15–24 years) (4). At this age, substance use is often associated with partying (5), youth justify their consumption by the sensations it provides, the desire to get intoxicated, or to facilitate integration into a group (3). However, several studies have highlighted the risk of correlation with other psychopathologies (6) and a risk of future mental health impairment (7).

A key point highlighted in the literature is that alcohol or substance use in adolescence and adulthood is significantly associated with maltreatment and negative childhood experiences (7). More globally, children's experiences and living conditions, from pregnancy onwards (8) have a major influence on the future mental health of victims. Indeed, the World Health Organization (WHO) claims that one in four adults worldwide report having experienced physical violence in childhood (9). A WHO study (10) reports that childhood adversity accounts for 29.8% of all disorders in all countries. In France, in 2015, between 17.6 and 22.0% of women, and between 12.9 and 18.0% of men reported having experienced at least one form of abuse during childhood (11), which may have major consequences in terms of public health. Numerous studies have highlighted the consequences of child maltreatment on the mental health of individuals. Moreover, numerous studies have pointed out the bidirectional relationship between mental health and SUD (12). Alcohol consumption can significantly trigger and exacerbate symptoms of internalization, and conversely, symptoms of internalization increase a person's vulnerability to developing SUD (13). Therefore, these adolescents who exhibit vulnerabilities appear to be at a higher risk of developing psychological symptoms associated with SUD. While these factors have been identified in the general population, we have less evidence for children in child protection care, despite overexposure to maltreatment, but also to mental health disorders (14). Additionally, studies on substance use among minors in child protection services are less existing, especially in France. It therefore seems important to us to focus on this population, which is exposed to multiple forms of abuse (15). Indeed, a cumulative phenomenon of exposure to abuse during childhood seems to amplify the risk of developing a psychological pathology (16–18). Besides, this cumulative aspect has a negative impact on the reduction of psychological symptoms over time (19).

A recent study underlined the complexity of estimating substance use among minors in child welfare services, which depends on the way in which it is measured and the age of the sample (20). Thus, rates ranging from 20% (21) to 50% (22) have been identified. It is essential to take this issue into account, as substance use is strongly associated with delinquency (23) and homelessness (24) in child protection populations, as reported by Bransard et al. (14), who point out that 25% of homeless people and 20% of adults in prison in France were previously child protection beneficiaries.

Considering maltreatment and its specificity as a risk factor for mental health disorders, and in particular substance and alcohol abuse, implies modifying care to adapt it to this population by favoring trauma-focused approaches to combat the long-term impact of the abuse (25). While many studies highlight the impact of maltreatment on the risk of illicit substance use and mental health deterioration, few studies have focused specifically on populations of maltreated children in the care of social services. The primary aim of the study was to ascertain whether Substance Use Disorders (SUD) among these minors varied based on the type of maltreatment experienced. The second objective was to examine the association between SUD and the presence of other psychological symptoms.

2. Methods

The present study relies on an analysis of data collected as part of the population-based study on children and young adults accompanied by the Essonne departmental council in the context of child protection. This is a retrospective cross-sectional study. The aim of this population-based study is to highlight the characteristics of maltreatment of minors in the Aide Sociale à l'Enfance (ASE) and to examine the psychological disorders of minors and young adults. It was developed from the Canadian incidence study (26). To conduct this population-based study, an email was sent to all social workers in charge of measures, in which a link was provided to allow them to complete the tool. We used the LimeSurvey software, so that the survey could be completed online. Professionals had between June 2020 and November 2020 to complete the survey. In total, 1,557 records were partially completed. 242 files were discarded because of too many missing answers. The result is a set of 193 variables containing information on 1,315 completed files, i.e., 19.185% of minors in care in the whole department.

As part of this research, we worked on the following variables Socio-demographic data: age, sex, duration of care, legal status of the child, interaction of the child with the parents, schooling, MDPH recognition (departmental house for the disabled people) and psychological care of the minor.

Data related to maltreatment

The data collected in the files are those that were included in the reason for care by the child protection services. These are physical abuse, sexual abuse, neglect, exposure to domestic violence, and psychological abuse. In terms of the timeline of events, the maltreatment comes first. Indeed, they prompted the need for intervention by child protection services. The symptoms were assessed at a later stage.

Data on psychological disorders

From the administrative files, social workers were asked to record psychological symptoms in children: depression, anxiety, suicidal ideation, self-destructive behavior, Attention Deficit Hyperactivity Disorder (ADHD), aggressive behavior. This research was recorded in the Université de Lorraine's research register.

In this research, we focused on young individuals who engage in psychoactive substance consumption. Substance consumption is defined as a daily consumption of cigarettes and/or cannabis, as well as regular alcohol consumption. A first group of SUD youth was formed (N = 72) was compared to a group without substance use

($N = 776$). To do so, we excluded all children with an age of 11 years or younger because in our cohort, no child under the age of 11 presented cigarette, cannabis, and/or alcohol consumption.

Statistical analyses

Data analyses were performed using R software (version 4.0.2) and the level of significance was set at 0.05. In a first step, we performed descriptive analyses. The distribution of continuous data was estimated using the Anderson-Darling test. As the continuous variables are all non-parametric, they were described using medians and interquartile ranges (IQR). Categorical variables were reported in terms of number (n) and percentage (%).

We opted to perform several multivariable binary logistic regression models to explore the association of SUD. The results are presented in terms of odds ratios (OR) with 95% confidence intervals (95% CI). First, we explored the association of CPSA with sociodemographic variables (Model 1). Second, we investigated the association of CPSA with all childhood maltreatment variables (Model 2). Third, we examined the association between CPSA and the set of psychological symptoms variables (Model 3). Fourth, we included both sociodemographic and maltreatment variables (Model 4). Fifth, we included both sociodemographic and psychological symptoms variables (Model 5). Finally, we integrated sociodemographic, maltreatment, and psychological symptoms variables into the model (Model 6). Variation inflation factors (VIF <10) were calculated to check for non-correlation and multicollinearity (27). All the eigenvalues of the VIF were less than 10. With these results, multicollinearity has been ruled out as a concern. Finally, we calculated the Nagelkerke and McFadden R^2 for each model.

Results

In the entire cohort, a total of 72 adolescents were afflicted by SUD. The median age for this subgroup of minors was 17 years [15–19], with 41 (56.9%) being male. The predominant legal status among these adolescents with SUD was categorized as parental authority, encompassing 38 cases (52.8%). Interactions between parents and offspring were predominantly characterized as "frequent," representing 25 instances (40%). A substantial portion of this cohort of minors, approximately 47.2%, is found to be disengaged from formal education. Among these individuals, 9 cases (12.5%) had received formal recognition for having a recognized disability. Notably, a considerable subset of these adolescents, totaling 30 cases (41.7%), were concurrently receiving psychological support. All socio-demographic variables are described in Table 1.

Univariable analysis

Table 2 presents the odds ratios estimating the association for SUD (Substance Use Disorder) with different variables. SUD was associated with higher age [1.21; CI (1.08–1.36); $p < 0.001$], infrequent parental interactions [2.91; CI (1.14–8.96); $p = 0.037$], suicidal ideation [3.52 (2.10–5.9); $p < 0.001$], Self-destructive behavior [5.86; CI (3.54–9.71); $p < 0.001$], and Aggressive Behavior [6.05; CI (3.6–10.19); $p < 0.001$]. Conversely, being enrolled in school appeared to reduce the risk of SUD [0.16; CI (0.09–0.27); $p < 0.001$]. Our results highlight that a form of maltreatment is significantly associated with SUD.

Analysis of models

Table 3 presents the results of multivariable binary logistic regression models applied to analyze Substance Use Disorder (SUD). Elevated age and disengagement from formal education demonstrated persistent associations with SUD across the models. Additionally, it can be highlighted that self-destructive behavior and aggressive behavior also exhibited consistent associations across all models. However, the association with frequent parental interactions was significant only within Models 5 and 6. Moreover, the status of being classified as an emerging adult exhibited an association with SUD only within Models 1 and 4.

Association with child maltreatment

Our results suggest a lack of association between distinct forms of maltreatment and SUD (Table 2). In Model 2, the associations were as follows: [1.43; CI (0.79–2.6); $p = 0.241$] for instances of physical violence, [1.41; CI (0.73–2.59); $p = 0.29$] for instances of sexual abuse, [0.96 (0.54–1.74); $p = 0.895$] for instances of neglect, [0.81; CI (0.44–1.50); $p = 0.503$] for instances of psychological violence, and [0.66; CI (0.37–1.16); $p = 0.154$] for instances of domestic violence. All models confirm the lack of significance obtained in Model 2.

Discussion

This study examined the association between SUD and forms of child maltreatment, but also with psychological symptoms. To our knowledge, this is the first French study to address this issue in a population of children in child protection care. The main hypothesis of our study was that in the specific population of minors in child protection care, SUD by these minors would be associated with different forms of maltreatment. In our results, on a population of subjects who had suffered abuse and were accompanied by child protection services, the type of abuse did not seem to influence SUD. However, our findings predominantly indicate that SUD is significantly linked to higher age, disengagement from schooling, Self-destructive behavior, and Aggressive Behavior. Moreover, certain models have indicated a significant inverse association between individuals with disabilities and SUD. Children who engage in frequent interactions with their parents, as opposed to those with permanent interactions, exhibit a significant association with SUD.

These results on the absence of association between maltreatment and SUD seem to contradict the existing literature that highlights the association between childhood maltreatment and the risk of substance use in adolescence or adulthood (7, 28).

TABLE 1 Characteristics of the SUD[a] and others.

	SUD (72)	Other (776)	All (848)
	Median [IQR] / *n* (%)	Median [IQR] / *n* (%)	Median [IQR] / *n* (%)
Socio-demographic variables			
Age	17 [15–19]	16 [12–20]	16 [4]
Sex			
Female	31 (43.05)	339 (43.68)	370 (43.63)
Male	41 (56.94)	437 (56.31)	478 (56.37)
Status of the child			
Parental authority	38 (52.78)	451 (58.12)	489 (57.67)
Adult CJM[b]	21 (29.16)	175 (22.55)	196 (23.11)
Delegation of authority	6 (8.33)	37 (4.77)	43 (5.07)
Pupil of the state	1 (1.38)	21 (2.71)	22 (2.59)
Unaccompanied minors awaiting status	5 (6.94)	60 (7.73)	65 (7.67)
Other	1 (1.38)	32 (4.12)	33 (3.89)
Parent–child interaction			
Permanent	5 (6.94)	121 (15.59)	126 (14.86)
Often	25 (34.72)	243 (31.31)	268 (31.6)
Rarely	20 (27.78)	166 (21.39)	186 (21.93)
Never	22 (30.56)	246 (31.70)	268 (31.6)
Education			
Yes	34 (52.78)	676 (87.11)	714 (84.2)
No	38 (47.22)	100 (12.89)	134 (15.8)
Recognition MDPH (Departmental house for disabled people)			
Yes	9 (12.5)	170 (21.91)	179 (21.11)
No	63 (87.5)	606 (78.09)	669 (78.89)
Psychological support			
Yes	30 (41.67)	360 (46.39)	390 (45.99)
No	42 (58.33)	416 (53.61)	458 (54.01)
Child maltreatment			
Physical Abuse			
Yes	38 (52.78)	332 (42.78)	366 (43.16)
No	34 (47.22)	444 (57.22)	482 (56.84)
Sexual abuse			
Yes	16 (22.22)	138 (17.78)	154 (18.16)
No	56 (77.78)	638 (82.22)	694 (81.84)
Neglect			
Yes	47 (65.28)	527 (67.91)	574 (67.69)
No	25 (34.72)	249 (32.09)	274 (32.31)
Psychological violence			
Yes	39 (54.16)	444 (57.22)	483 (56.96)
No	33 (45.83)	332 (42.78)	365 (43.04)
Domestic violence			
Yes	27 (37.5)	349 (44.97)	376 (44.34)
No	45 (62.5)	427 (55.03)	472 (55.66)

(Continued)

TABLE 1 (Continued)

	SUD (72)	Other (776)	All (848)
	Median [IQR] / *n* (%)	Median [IQR] / *n* (%)	Median [IQR] / *n* (%)
Psychological symptoms			
Depression			
Yes	40 (55.56)	354 (45.62)	394 (46.46)
No	32 (44.44)	422 (54.38)	454 (53.54)
Anxiety			
Yes	54 (75)	557 (71.78)	611 (72.05)
No	18 (25)	219 (28.22)	237 (28.95)
Suicidal Ideation			
Yes	27 (37.5)	113 (14.56)	140 (16.51)
No	45 (62.5)	663 (85.45)	708 (83.49)
Self-destructive behavior			
Yes	44 (61.11)	164 (21.13)	208 (24.53)
No	28 (38.89)	612 (78.86)	640 (75.47)
ADHD[4]			
Yes	21 (29.17)	221 (28.48)	242 (28.54)
No	51 (70.83)	555 (71.52)	606 (71.46)
Attachment disorder			
Yes	42 (58.33)	396 (51.03)	438 (51.65)
No	30 (41.67)	380 (48.97)	410 (48.35)
Aggressive Behavior			
Yes	49 (68.05)	202 (26.03)	251 (29.6)
No	23 (31.94)	574 (73.97)	597 (70.4)

[a] Substance use disorders.

[b] Contrat jeune majeur (Young Adult Contract: This refers to a type of legal agreement or program that provides support and assistance to young individuals who are transitioning from the status of minors to adults in the child protection context).

However, most studies compared maltreated children with non-abused children. In our study, all the minors were exposed to childhood maltreatment. We note high odd ratios with physical abuse and sexual abuse. The lack of significance may be an effect of the small size of our consumer group. This could also mean that the type of adversity does not influence SUD, but that it would be child maltreatment in a general sense. The literature points out (29) that child maltreatment can be a stressful life event, often chronic and long-lasting, and where substance use can be used to alleviate the affect (29, 30).

In contrast, our study highlighted strong associations with psychological symptoms, including suicidal ideation. These findings are consistent with previous literature (31, 32). This is important as recent research has noted that children in child protection settings had less suicidal ideation than the general population but were more likely to act on it (33). This aspect raises inquiries into the mentalization capacity of the youth. Previous studies have underscored a correlation between attachment disorders, mentalization, and suicidal ideation (34). Youth in child protection are more exposed to attachment disorders, potentially influencing their mentalization processes and exacerbating the risk of transitioning from ideation to action (35). Further studies should delve into this highly significant aspect to enhance the management of children in child protection.

Regarding psychological symptoms, our results indicate also an association with a risk of aggressive behavior. These findings align with the work of Dória et al. (20) and call for early intervention to prevent progression toward legal action (11). Previous studies have underscored, in the general population, that substance consumption during adolescence is linked to a range of adverse outcomes, including aggressive behavior, antisocial conduct, and school disengagement (36). Substance Use Disorder (SUD) associated with aggressive behaviors acts as predictors of delinquent involvement (23). These youths exhibiting aggressive behaviors tend to identify themselves within peer groups, facilitated by their disengagement from schooling (37). Within these groups, aggressive behaviors and SUD are glorified, thereby escalating the risk of behavioral issues and SUD over the course of years (37). If our study does not measure the bidirectional aspects between SUD and psychological symptoms, it highlights numerous associations that would require further investigation into these dimensions. Indeed, bidirectionality is a complex and significant

TABLE 2 Univariable binary logistic regression models.

	OR 95% CI	*p* value
Age	**1.21 (1.08–1.36)**	**<0.001**
Sex (Male)	1.02 (0.63–1.67)	1
Status of the child		
Parental authority (reference)	–	
Adult CJM[1]	1.42 (0.8–2.47)	0.216
Delegation of authority	1.92 (0.69–4.55)	0.165
Pupil of the state	0.56 (0.03–2.82)	0.582
Unaccompanied minors awaiting status	0.99 (0.33–1.40)	0.982
Other	0.37 (0.02–1.80)	0.335
Parent–child interaction	–	
Permanent (reference)	–	
Often	2.49 (1.01–7.52)	0.069
Rarely	**2.91 (1.14–8.96)**	**0.037**
Never	2.86 (0.53–6.59)	0.128
Education (Yes)	**0.16 (0.09–0.27)**	**<0.001**
Recognition MDPH (Yes)	0.51 (0.25–1.04)	0.07
Psychological support (Yes)	0.82 (0.51–1.34)	0.46
Physical Abuse (Yes)	1.20 (0.74–1.94)	0.53
Sexual abuse (Yes)	1.32 (0.73–2.37)	0.34
Neglect (Yes)	0.89 (0.53–1.47)	0.693
Psychological violence (Yes)	0.88 (0.54–1.43)	0.62
Domestic violence (Yes)	0.73 (0.45–1.21)	0.26
Depression (Yes)	1.49 (0.92–2.42)	0.11
Anxiety (Yes)	1.17 (0.68–2.06)	0.68
Suicidal Ideation (Yes)	**3.52 (2.10–5.90)**	**<0.001**
Self-destructive behavior (Yes)	**5.86 (3.54–9.71)**	**<0.001**
ADHD (Yes)	1.03 (0.61–1.76)	0.89
Attachment disorder (Yes)	1.34 (0.82–2.19)	0.27
Aggressive Behavior (Yes)	**6.05 (3.6–10.19)**	**<0.001**

[1] Contrat jeune majeur (Young Adult Contract: This refers to a type of legal agreement or program that provides support and assistance to young individuals who are transitioning from the status of minors to adults in the child protection context).

issue in public health. However, it is not necessarily straightforward. Previous studies had emphasized the bidirectional aspect between SUD and aggressive behaviors (38). On the other hand, a more recent study pointed out a reciprocal association between alcohol consumption and internalized symptoms in an adolescent population (13). However, when the analysis accounted for gender and externalized symptoms, no bidirectional association was found. Despite these contradictory results, the bidirectional relationship between SUD and psychological symptoms has been documented in the general population (12, 39–41), further studies should further investigate populations exposed to adversity, especially in child protection settings.

Beyond the aspects related to symptomatology, our study highlights a significant association between school dropout and substance use. Studies have noted the effects of substance use, particularly cannabis, on school results (42, 43) and on school absenteeism. However, these studies were in general populations. It has been pointed out that children followed by the child protective services were significantly more likely to drop out of school than other children (44), but beyond the effects of maltreatment on school dropout (45), we have little information on the profile of minors. Our results suggest that children who are abused and use substances are more likely to drop out of school. This element must be taken into account in the holistic support of the child. In particular, by strengthening interaction between schools and child protection services, and by making reports to reduce the risk of children dropping out of school.

We propose not a distance but a multi-professional line of action, as it can be developed in the open dialog (46) protocols for network to address the issue of transference, which is partly undermined by the presence of attachment disorders. In this sense, it seems preferable to us to favor institutional psychotherapeutic approaches that favor the presence of a strong, multi-transferential collective (47). This approach would make it possible to fight against the process of desocialisation encouraged by attachment disorders and which can generate the risk of dropping out of school (48).

Limitations

Our study has several limitations. The main bias of the study is its design. A longitudinal study with repeated measurements to assess placed children would have allowed for greater data reliability. Secondly, the small sample size of the SUD may influence the significance of the results. Thirdly, there may be a recall bias in the case of retrospective data, especially with regard to sexual abuse where it is well known that there may be traumatic dissociation and lower reporting. Finally, the measurement of consumption was not carried out using a scale but from the elements recorded in the files. These are mainly related to a psychological assessment, but may also be the transcript of the observations of the referral educator.

Conclusion

This study highlights the association between psychological symptoms and susceptibility to psychoactive substance use. Importantly, it highlights a pronounced vulnerability to suicidal ideation and underscores a significant inclination toward aggressive behavior. Moreover, our results emphasize that among the cohort of minors under the purview of child protection services, those who partake in substance use are at an elevated risk of discontinuing their education. These comprehensive findings contribute crucial insights to the nuanced decision-making landscape within this institution (49).

There's no doubt that exposure to childhood trauma has a profound impact on mental well-being. As evidenced by prior research, it serves as a pivotal determinant that can shape the

TABLE 3 Comparison of logistic regression models to explain SUD.

	Model 1 N = 848	Model 2 N = 848	Model 3 N = 848	Model 4 N = 848	Model 5 N = 848	Model 6 N = 848
	OR 95% CI	OR 95% CI	OR 95% CI	OR 95% CI	OR 95% CI	OR 95% CI
Age	**1.23 (1.01–1.52)**	–	–	**1.24 (1.01–1.53)**	**1.40 (1.11–1.79)**	**1.39 (1.09–1.79)**
Sex (Male)	1.04 (0.61–1.78)	–	–	1.05 (0.60–1.88)	1.08 (0.59–2.02)	0.97 (0.5–1.89)
Status of the child		–	–			
Parental authority (reference)	-	–	–	–	–	–
Adult CJM[1]	**0.37 (0.14–0.94)**	–	–	**0.36 (0.14–0.92)**	0.51 (0.18–1.41)	0.5 (0.17–1.40)
Delegation of authority	2.37 (0.77–6.50)	–	–	2.21 (0.71–6.15)	2.99 (0.85–9.66)	3.03 (0.84–9.92)
Pupil of the state	0.51 (0.02–3.24)	–	–	0.49 (0.02–3.15)	1.36 (0.06–9.98)	1.17 (0.05–8.92)
Unaccompanied minors awaiting status	0.38 (0.10–1.23)	–	–	0.33 (0.09–2.32)	0.70 (0.17–2.54)	0.6 (0.14–2.28)
Other	0.41 (0.02–2.17)	–	–	0.44 (0.98–8.19)	0.44 (0.02–2.81)	0.45 (0.02–3)
Parent–child interaction	-	–	–	–	–	–
Permanent (reference)	-	–	–	–	–	–
Often	2.51 (0.96–7.91)	–	–	2.58 (0.99–8.19)	**3.73 (1.31–12.64)**	**3.57 (1.23–12.24)**
Rarely	2.21 (0.80–7.21)	–	–	2.34 (0.82–7.75)	2.78 (0.91–9.77)	3.08 (0.98–11.14)
Never	1.57 (0.53–5.32)	–	–	1.60 (0.54–5.50)	2 (0.62–7.41)	2 (0.6–7.55)
Education (Yes)	**0.15 (0.08–0.27)**	–	–	**0.15 (0.08–0.27)**	**0.24 (0.12–0.46)**	**0.23 (0.12–0.45)**
Recognition MDPH (Yes)	0.45 (0.19–0.97)	–	–	0.47 (0.20–1.02)	**0.37 (0.14–0.91)**	**0.39 (0.14–0.96)**
Psychological support (Yes)	1.12 (0.63–1.95)	–	–	1.11 (0.62–1.97)	0.91 (0.47–1.72)	0.95 (0.49–1.82)
Physical Abuse (Yes)	–	1.43 (0.79–2.60)	–	1.28 (0.67–2.47)	–	0.97 (0.46–2.01)
Sexual abuse (Yes)	–	1.41 (0.73–2.59)	–	1.29 (0.62–2.59)	–	1.07 (0.46–2.37)
Neglect (Yes)	–	0.96 (0.54–1.74)	–	1.02 (0.54–1.95)	–	1.11 (0.55–2.26)
Psychological violence (Yes)	–	0.81 (0.44–1.5)	–	0.74 (0.37–1.46)	–	0.68 (0.32–1.44)
Domestic violence (Yes)	–	0.66 (0.37–1.16)	–	0.72 (0.39–1.34)	–	0.60 (0.28–1.11)
Depression (Yes)	–	–	0.95 (0.51–1.77)	–	0.67 (0.34–1.32)	0.67 (0.33–1.33)
Anxiety (Yes)	–	–	0.66 (0.33–1.35)	–	0.83 (0.39–1.80)	0.85 (0.4–1.85)
Suicidal Ideation (Yes)	–	–	1.54 (0.77–3.07)	–	1.28 (0.59–2.77)	1.32 (0.60–2.87)
Self-destructive behavior (Yes)	–	–	**3.17 (1.63–6.26)**	–	**4.28 (2.01–9.33)**	**4.35 (2.02–9.59)**
ADHD (Yes)	–	–	0.57 (0.29–1.04)	–	0.81 (0.36–1.77)	0.83 (0.37–1.85)
Attachment disorder (Yes)	–	–	0.83 (0.45–1.50)	–	0.74 (0.37–1.47)	0.84 (0.41–1.74)
Aggressive Behavior (Yes)	–	–	**4.42 (2.42–8.26)**	–	**5.26 (2.66–10.63)**	**5.75 (2.87–11.84)**
McFadden R^2	0.13	0.01	0.15	0.14	0.28	0.29
Nagelkerke R^2	0.17	0.01	0.19	0.18	0.34	0.35

[1] Contrat jeune majeur (Young Adult Contract: This refers to a type of legal agreement or program that provides support and assistance to young individuals who are transitioning from the status of minors to adults in the child protection context).

Bold font indicates statistical significance ($p < 0.05$).

trajectory of an individual's psychological health. Addressing the effects of childhood maltreatment and ensuring effective management strategies emerge as paramount endeavors for safeguarding mental wellbeing or positive mental health and prevent the risk of SUD.

Data availability statement

The datasets used and/or analyzed in this study are available from the corresponding author upon reasonable request.

Ethics statement

Ethical approval was not required for the study involving humans in accordance with the local legislation and institutional requirements. Written informed consent to participate in this study was not required from the participants or the participants' legal guardians/next of kin in accordance with the national legislation and the institutional requirements.

Author contributions

AE and MM co-wrote the first draft of the article. AE, MM, AA, FW, JM, MR, and GS participated in the data collection and correction of the article. GS supervised the work. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Father's perceptions and care involvement for their very preterm infants at French neonatal intensive care units

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Objectives: We aimed to evaluate (1) fathers' perceptions and care involvement for their very premature infants and their views of the hospitalization period based on parental reports and (2) their evolution over time.

Methods: We used an online parental survey to assess answers from parents of very preterm infants who were successfully discharged from French neonatal units. We analysed answers from February 2014 to January 2019 to an anonymous internet-based survey from the GREEN committee of the French Neonatal Society. Responses were compared for period 1 (P1, 1998 to 2013) and period 2 (P2, 2014 to 2019).

Results: We analyzed 2,483 surveys, 124 (5%) from fathers and 2,359 (95%) from mothers. At birth, 1,845 (80%) fathers were present in the hospital, but only 879 (38%) were near the mother. The presence of fathers in the NICU increased from P1 to P2 (34.5% vs. 43.1%, $p = 0.03$). Nearly two thirds of fathers accompanied their infants during transfer to the NICU (1,204 fathers, 60.6%). Fathers and mothers had similar perceptions regarding relationships with caregivers and skin-to-skin contact with their infants. However, more fathers than mothers felt welcome in the NICU and in care involvement regarding requests for their wishes when they met their infant (79% vs. 60%, $p = 0.02$) and in the presentation of the NICU (91% vs. 76%; $p = 0.03$). Mothers and fathers significantly differed in the caring procedures they performed ($p = 0.01$), procedures they did not perform but wanted to perform ($p < 0.001$), and procedures they did not perform and did not want to perform ($p < 0.01$).

Conclusion: Most fathers were present at the births of their very preterm infants, but fewer fathers were near the mother at this time. Less than two thirds of fathers

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accompanied their infants to the NICU. There should be further changes to better meet the specific needs of the fathers of infants requiring care in the NICU. Continuing assessment with an online questionnaire may be useful to monitor changes over time in father's involvement in NICUs.

KEYWORDS

online survey administration, fathers, preterm infants, NICU, care involvement and presence

1 Introduction

The birth and hospitalization of a very preterm infant (VPI) in a neonatal intensive care unit (NICU) is a major disruption in the family's life. The stressful and intimidating NICU environment and the uncertain health outcomes for newborns is especially traumatic for parents (1). The burden of these multiple stresses may have long-term consequences on the parent-child relationship and parents' mental health (2). The parents of premature neonates have increased risk of depression, post-traumatic stress disorder, and anxiety (3–5). Infant-and family-centered developmental care strategies can prevent these complications and meet the family's needs. Recent studies have recommended providing support for increased involvement of mothers and fathers in the care of their premature infants (6–8).

The presence of fathers in the NICU is now believed to promote the experience of fatherhood with the premature infant, increase the well-being of the mother and infant, and contribute to better infant brain development (8–10). Premature birth may reverse the roles of fathers and mothers in that fathers may be on the front line of care. In particular, fathers of such newborns often receive information on the infant's health and communicate this information to their partners (11). However, often, fathers feel unable to care for their infants, lack self-confidence, and are intimidated by the small size and apparent fragility of their premature newborn (11–13).

Biological and neuroscientific studies have shown that fathers have an innate ability to bond with and care for their premature infants (14, 15). Several studies have shown that fathers of preterm infants wanted to play an active role in their infants' care (9, 12, 16–19). Father-infant bonding appears to be facilitated by the development of fathering skills and increased involvement in infant care (9, 10).

Thus, fathers seeking to care for their premature newborns need support and guidance that meet their specific needs. Previous parental surveys have assessed parental experiences and needs in the NICU (20, 21). However, only a few studies have been performed at the national level, and they rarely specifically examined fathers' views. A recent survey was submitted to parents in France (21, 22). This tool gave the opportunity to collect the presence and experiences of fathers' in French NICUs.

Our main objective was to evaluate the perception of fathers' presence at the time of birth of their VPIs and their experiences during their infants' transfer to the NICU. Secondary objectives were to evaluate fathers' experiences of feeling welcome in the NICU, their perceptions of their relationships with caregivers, and their participation in care and skin-to-skin contact, as compared to mothers' experiences. Finally, we aimed to assess the evolution of these items over time.

2 Patients and methods

2.1 Development and distribution of the questionnaire

In France, an internet-based survey was started in February 2014 as a collaborative project between members of the French Neonatology Society and parental associations within the *Groupe de Réflexion et d'Evaluation du Nouveau-né* (GREEN Committee) (21, 22). The objective was to develop recommendations to improve family integration into NICUs and to modify the hospital environment so that it better meets the needs of parents and their infants (7).

This anonymous online survey for the parents of premature infants who were hospitalized in NICUs consists of 222 questions regarding neonatal care (some multiple-choice and some open-ended) and covers 9 distinct areas. The topics covered include the particular circumstances of the birth, the parent's perceptions of feeling welcome, transfer of the infant to different units, breastfeeding, participation in care, and preparation for hospital discharge. It is intended for all parents who had a newborn infant hospitalized in a NICU.

2.2 Characteristics of parents and infants and data collection

This study was an analysis of quantitative data based on responses collected up to January 2019. We focused on fathers whose infants were born before 32 weeks of gestational age and successfully discharged from the hospital (Figure 1).

A total of 29 items described the population of fathers as a whole: demographic characteristics, presence at the time of birth, and support provided during the infant transfer to the NICU. For these 29 items, we used the responses of all fathers and mothers.

A total of 34 items focused on both parents' experiences. These allowed us to specifically analyse responses about feeling welcome in the NICU, relationships with caregivers, participation in different caring procedures (detailed in Table 1) and skin-to-skin contact. For these 34 questions, we compared the answers of mothers and fathers.

Abbreviations: GA: Gestational age; NICU: Neonatal intensive care unit; VPI: Very preterm infant.

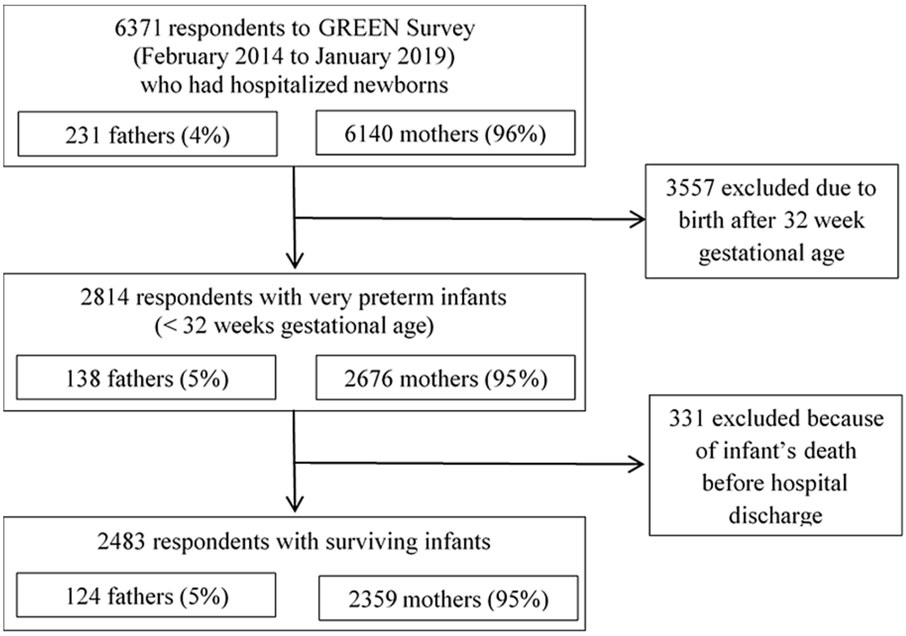

FIGURE 1
Study flow chart of the parents of very preterm infants who responded to the GREEN survey.

Finally, we compared the periods of VPI birth from 1998 to 2013 (period 1; P1) and 2014 to 2019 (period 2; P2) to analyse changes over time as the first recommendations of the GREEN committee, aiming to improve family integration into NICUs, were presented in 2014.

Moreover, open-ended questions were included in the survey to allow the expression of individual feelings of the responders. They were used to describe more precisely the father's perception and to illustrate the presentation of the results of the survey with individual testimonies about fathers' care involvement.

2.3 Statistical analysis

Quantitative variables are presented as mean and standard deviation (SD) or number (%) and were compared by student *t*-test or Fisher's exact test, as indicated. For multiple comparisons, the *p*-value was adjusted using Holm's method (23). To compare the proportions of fathers and mothers among numerous items, we used the signs test (24). All statistical analyses were performed with R v3.5.2.

3 Results

Table 2 shows the characteristics of responding parents and their VPIs. Significantly more fathers than mothers reported living as couples, fathers had significantly more education than mothers, and the infants of fathers were significantly more preterm. As compared with P1 (1998 to 2013), during P2 (2014 to 2019), fathers were older [mean age 32.7 (5.6) vs. 31.3 (5.9) years, $p < 0.0001$], more families were living as couples (94.2% vs. 89.4%; $p < 0.0001$), and the proportion of families with siblings was greater (35.3% vs. 29.8%, $p < 0.01$) (data not shown). Not all fathers and mothers responded to all the questions of the survey. The number of responders is specified for each question when it is necessary.

3.1 Presence of fathers at birth and during transfer to the NICU

Most fathers were present at birth, but less than half were near the mother, mainly due to medical restrictions.

The fathers regretted not being at their wives' side: "*They did not come to get me to attend the cesarean*" or "*The nurses initially took my wife. After 3 min that seemed like an eternity, I had to insist twice and impose myself to attend the delivery. Our children arrived a few minutes later.*"

However, the father's presence near the mother increased over time. Most fathers were separated from their infants less than 24 h. Fathers visited before mothers. An increasing majority of fathers accompanied their infants to the NICU and most fathers accompanied their infants during intra-hospital transfers with caregivers.

The transfer of the newborn to the NICU remained a moment of particular vulnerability: the lack of information, the lack of welcome time and the feeling of loneliness were difficult for fathers: "*My twins were taken to the unit without me. When I arrived, the doctors and nurses were all busy caring for my twins. So no one greeted me*" or "*When he was born, we could not see him; he was immediately taken to the next room to be intubated. We did not hear him scream*" or "*I wish I had accompanied my child to NICU. I found myself without my baby and without my wife, alone and distraught. It was a very difficult time for me.*"

The fathers reported easily finding the location of the NICU, even when going alone. This was similar for inter-hospital transfer, which fathers mostly performed on their own. In comparing P1 to P2 (Table 3), the father's presence near the mother at birth increased over time.

TABLE 1 Caring procedures.

Caring procedure domains	Type of caring procedures
Feeding	Preparing the milk in a syringe or cup Giving the bottle or cup Inserting or removing the nasogastric tube
Monitoring	Weighing/measuring your child Reporting monitoring data on your infant's chart (oxygen saturation/heart rate) Taking your child's temperature Turning off monitor alarms Putting electrodes on the chest
Hygiene care	Fully washing your child in the incubator Performing umbilical cord care Bathing your child wearing high flow or nasal CPAP cannula Bathing your child intubated
Diaper care and stools	Changing a nappy in the incubator Performing an abdominal massage Helping your child to stool by bending the knees
Positioning and clothing	Dressing your child wearing high flow or nasal CPAP cannula Dressing your child intubated Set up the cocoon or swaddling Cleaning the incubator or bed Cleaning feeding equipment (cup/bottle) Bringing clothes and sheets, washing them at home
Skin-to-skin contact or holding	Deciding when your unventilated child can be in skin-to-skin contact Deciding when your child can be in skin-to-skin contact with ventilation or nCPAP Deciding when your unventilated child can be held Deciding when your child can be held with ventilation or nCPAP Deciding when your intubated child can be held
Medical care	Putting on the cannula and repositioning the child in phototherapy Giving glucose before a painful procedure Hold the child by swaddling and comforting during a painful procedure Stimulating the child in case of apnoea Changing O2 high flow cannula Changing the CPAP equipment Suctioning at the nose or mouth
Administering pharmacological treatment	Administering medications orally Administering medications through the gastric tube Putting in eyedrops Administering inhalation

CPAP, continuous positive airway pressure; nCPAP, nasal continuous positive airway pressure.

3.2 Feeling welcome in the NICU

Overall, parents felt very welcome by the medical team, they reported that they received adequate and easily understood information, and the efficiency of the healthcare workers made them trust the team (see Tables 4, 5). More fathers than mothers had positive feelings during the first visit and during explanations of the unit's guidelines. Mothers met physicians later than did fathers but not significantly.

3.3 Relationships with caregivers

Overall, parents were very satisfied or satisfied about their relationships with caregivers. Most fathers and mothers perceived the staff as being available and felt confident in asking questions. They also felt involved in decision-making about the infant's well-being, health, and daily schedule. The parents felt comfortable talking openly to the caregivers, and the information they received seemed consistent among different caregivers. Fathers and mothers did not significantly differ in caregiver relationships (see Tables 6, 7).

3.4 Participation in care

Overall, most parents felt very positive regarding the support they received from caregivers for participating in their infants' care (Table 8). We also analysed participation in care more precisely by identifying different caring procedures (see Table 1).

However, when comparing caring procedures overall between mothers and fathers, we found significant differences in the number of caring procedures that were performed, procedures not performed but that the parent wanted to perform, and procedures not performed and the parent did not want to perform (Figure 2). Overall, fathers performed more caring procedures than mothers (Figure 2A; $p = 0.01$), including bathing and dressing the intubated infant, preparing the infant for phototherapy, and stopping the monitor alarms. More mothers than fathers wanted to perform caring procedures that they did not actually perform (Figure 2B; $p < 0.001$), including cord care, bathing and dressing of the intubated infant, preparing for breast feeding, and administering eye drops. Finally, more fathers than mothers reported not performing caring procedures that they did not want to perform (Figure 2C; $p < 0.01$), including washing the infant completely in an incubator, administering a sweet solution, and performing facilitated tucking during a painful procedure. However, more mothers than fathers did not perform certain other caring procedures that they did not want to perform, including installing and removing a nasogastric tube and performing nasal or oral suction procedures.

Fathers and mothers did not differ in specific caring procedures, except for care of the umbilical cord, with significantly more mothers than fathers not performing cord care although they wanted to (29% vs. 11%, $p = 0.026$).

3.5 Participation in skin-to-skin contact

Most parents were informed of the benefits of skin-to-skin contact, performed this procedure, and felt confident when doing so: "*Skin-to-skin contact was very strongly recommended to us, which we obviously agreed with* (see Table 9). *We had a comfortable chair, available staff, everything was always done with patience, with measured and careful gestures.*" Fathers and mothers did not significantly differ in this care, especially regarding the timing of the first skin-to-skin

TABLE 2 Characteristics of respondents and their infants.

Characteristics	Fathers' responses (N = 124)	Mothers' responses (N = 2,359)	p
Father's age, mean (SD), N = 2,414	32.6 (5.6)	31.9 (5.8)	0.19
Mother's age, mean (SD), N = 2,478	31.6 (5.1)	30.8 (5.4)	0.1
Mother's family situation at the time of birth: in couple, n (%), N = 2,483	124 (100)	2,256 (95.6)	**<0.01**
Mother's current family situation: in couple, n (%), N = 2,483	118 (95.2)	2,155 (91.4)	0.18
Father's education level, n (%), N = 2,402			**<0.001**
College level	37 (29.8)	1,084 (47.6)	
Baccalaureate level	1 (0.8)	103 (4.5)	
Higher education	86 (69.4)	1,091 (47.9)	
Mother's education level, n (%), N = 2,482			**<0.05**
College level	30 (24.2)	810 (34.4)	
Baccalaureate level	1 (0.8)	59 (2.5)	
Higher education	93 (75)	1,489 (63.1)	
Father's occupation, n (%), N = 2,409			0.63
Employed	121 (97.6)	2,180 (95.4)	
Unemployed	3 (2.4)	98 (4.3)	
Student	0 (0)	4 (0.2)	
Other (pensioner etc.)	0 (0)	3 (0.1)	
Mother's occupation, n (%), N = 2,482			0.17
Employed	109 (87.9)	1,961 (83.2)	
Unemployed	13 (10.5)	380 (16.1)	
Student	2 (1.6)	15 (0.6)	
Other (pensioner etc.)	0 (0)	2 (0.1)	
Father's native language, n (%), N = 2,418			0.1
French	118 (95.9)	2,077 (90.5)	
Other language	1 (0.8)	83 (3.6)	
Bilingual, including French	4 (3.3)	135 (5.9)	
Mother's native language, n (%), N = 2,483			0.08
French	114 (92)	2,220 (94.1)	
Other language	5 (4)	32 (1.4)	
Bilingual, including French	5 (4)	107 (4.5)	
Single or multiple births, n (%), N = 2,483			0.27
Singleton	103 (83.1)	1,988 (84.3)	
Twins	20 (16.1)	351 (14.9)	
Triplets	0 (0)	17 (0.7)	
Quadruplets	1 (0.8)	3 (0.1)	<0.05
Infant's gestational age at birth, median (min–max), N=2,483	28 (24-31)	29 (24-31)	**<0.05**
Birth weight, g, n (%), N = 2,483			0.37
<500	1 (0.8)	33 (1.4)	
500 to 1,500	113 (91.1)	2,002 (84.9)	
>1,500	10 (8.1)	324 (13.7)	
Family with siblings living at time of birth, n (%), N = 2,480	43 (34.7)	757 (32.1)	0.56
Number of hospitals where the infant received care, median (min–max), N = 2,282	1 (1–5)	1 (1–5)	0.36

Bold values indicate statistically significant differences.

contact. However, a higher proportion of mothers than fathers would have liked to perform skin-to-skin contact more often.

Nonetheless, some fathers' open-ended comments about skin-to-skin contact described a stressful experience: "*For the first skin-to-skin contact, the nurse insisted when I wasn't ready; it was too early for me, I was tired and was afraid he would breathe wrong on me*" or "*During skin-to-skin contact, the temperature was dropping too fast, and I was afraid I was making him worse.*" or "*I felt more uncomfortable than insecure, a little ridiculous unclothed in the nursing unit*" or "*I was afraid of respiratory arrest when the assistants were removed and afraid of falling asleep with the baby and not doing it right.*"

4 Discussion

The results of this national web-based survey indicated that most fathers were present at birth, but less than half were near the mother at this time, although this number increased slightly over time. Very few fathers reported no separation from their infants, although most met their VPIs within 1 day of birth. Less than two thirds of fathers accompanied their infants during transfer to the NICU. We found many similarities between the responses of fathers and mothers; however, there were significant differences in the perceptions of fathers and mothers regarding feeling welcome in the neonatal unit and involvement in care of their infants.

The main limitation of this study was that it used an internet-based open-access questionnaire that required knowledge of the French language, which probably explains why the study population had a high percentage of parents with high socio-economic class and traditional family structure. Thus, our results may not be applicable to fathers from economically vulnerable families. In addition, our study population probably had more fathers that were involved in their infant's care. In agreement, a European study also reported that fathers with higher education were more likely to be present in the unit during medical rounds than other fathers (25). This situation could limit the generalizability of our results regarding fathers' perceptions. In addition, because our data were collected retrospectively, there was a risk of incorrect reporting of information by parents, whose perceptions could have changed over time and after discharge. Data

TABLE 3 Comparison of fathers' presence at birth and transfer of newborns to other hospital units during period 1 (P1; 1998–2013) and period 2 (P2; 2014–2019).

	Total (*N* = 2,483)	Overall population from P1 (1998–2013) (*N* = 1,371)	Overall population from P2 (2014–2019) (*N* = 1,112)	Adjusted *p*
Father at birth was, *n* (%), *N* = 2,291				0.03
Present in birth room with mother	879 (38.4)	439 (34.5)	440 (43.1)	
Present but in a room next door because not allowed in birth room by medical staff	932 (40.7)	543 (42.7)	389 (38.1)	
Present but in a room next door, because did not want to attend	34 (1.5)	21 (1.7)	13 (1.3)	
Not present because of no time or unable to come	318 (13.9)	196 (15.4)	122 (12)	
Not present for other reasons	128 (5.5)	72 (5.7)	56 (5.5)	
Length of time between birth and mother's first visit with baby, *n* (%), *N* = 1,960				0.001
No separation	130 (6.6)	58 (5.3)	72 (8.4)	
Separation less than 24 h	1,330 (67.9)	701 (63.7)	629 (73.1)	
Separation more than 24 h	500 (25.5)	341 (31)	159 (18.5)	
Length of time between birth and father's first visit with baby, *n* (%), *N* = 2,009				0.15
No separation	795 (39.6)	420 (37)	375 (42.8)	
Separation less than 24 h	1,148 (57.1)	667 (58.8)	481 (55)	
Separation more than 24 h	66 (3.3)	47 (4.2)	19 (2.2)	
Number of fathers who accompanied baby to the first neonatal unit, *n* (%), *N* = 1,986	1,204 (60.6)	648 (57.9)	556 (64.2)	0.19
Means by which father followed his baby to another hospital, *n* (%), *N* = 73				1
By himself	71 (97.3)	46 (97.9)	25 (96.2)	
In an ambulance	2 (2.7)	1 (2.1)	1 (3.8)	
If father went by himself, those who easily found the service where the infant was transferred, *n* (%) (*N* = 58)	53 (91.4)	32 (91.4)	21 (91.3)	1
For infant's transfer in the same hospital, fathers who were accompanied by a member of the healthcare team, *n* (%), *N* = 1,042	901 (86.5)	455 (83.5)	446 (89.7)	0.17
If fathers went there alone, those who easily found the service where the infant was transferred, *n* (%), *N* = 132	115 (87.1)	74 (89.2)	41 (83.7)	1

were also missing for some items in the questionnaire for some respondents. However, the very high number of total answers from mothers regarding the fathers' behaviors suggests the validity of our data.

To our knowledge, this was the first nation-wide study that used quantitative data to assess fathers' presence at the birth of their preterm infants and during their transfer to the NICU. Previous studies have reported fathers' perceptions during pregnancy and birth (26) but not the proportions of fathers present at birth and the presence of the father with the mother at that time. Similar to our results, a German survey from 2011 interviewed 111 fathers of very low birth weight infants in 2 NICUs and found that nearly all the fathers met their infants on the first day of life. However, first contact was earlier for these German fathers than the fathers in our study in that 33.3% of them saw their infants at birth and 61.3% saw them within 1 h of birth (27).

There is a general lack of data regarding the presence and role of fathers at birth of a VPI and during transfer to the NICU, even though fathers reported being very satisfied when accompanying their infants immediately after birth and when encouraged to touch and hold them (18). Current recommendations for infant-and family-centered developmental care strongly support parents being as close as possible to their infants (6) and that separation should be avoided. European studies showed that policies regarding parental presence and involvement in the NICU varied widely among countries and among NICUs within individual countries (28, 29). There is greater support for parental presence in the NICU in northern than other European countries. However, increasing emphases are being placed on applying fewer restrictions for parents regarding access to the NICU, more encouragement for parents to provide skin-to-skin contact, and improved accessibility of parents to bedrooms, family kitchens, and private bathrooms in NICUs (28, 30). These changes of NICU policies

TABLE 4 Parents' perceptions of feeling welcome in neonatal units.

	Fathers	Mothers	Adjusted *p*
Upon arrival in the neonatal care intensive unit, *n* (%)			
The parent felt expected by the medical team (*N* fathers = 95 and *N* mothers = 1,944)	83 (87)	1,606 (83)	1
The team devoted the time required (*N* fathers = 95 and *N* mothers = 1,944)	93 (98)	1,731 (89)	0.49
The team asked about the parents' wishes/needs when meeting with the infant (*N* fathers = 86 and *N* mothers = 1,880)	68 (79)	1,128 (60)	0.02
During this first visit, the parent was introduced to, *n* (%) (*N* fathers = 95 and *N* mothers = 1,944)			
The service and its operating rules	87 (92)	1,474 (76)	0.03
The caregivers of the infant	85 (89)	1,597 (82)	1
The infant's room and with explanations about the function of each device	81 (85)	1,497 (77)	1
The infant	85 (89)	1,720 (88)	1
Waiting time before talking with a member of the healthcare team (nurse, caregiver, intern, physician) about the condition of infant, *n* (%) (*N* fathers = 100 and *N* mothers = 1,960)			
After 24 h	6 (6)	294 (15)	0.56
Waiting time before meeting a physician, *n* (%) (*N* fathers = 91 and *N* mothers = 1,731)			
After 24 h	20 (22)	658 (38)	0.08
During the first interview with the physician, the parent felt, *n* (%) (*N* fathers = 95 and *N* mothers = 1,890)			
Supported	83 (87)	1,561 (83)	1
Listened to	86 (91)	1,647 (87)	1
Included in the practice	82 (86)	1,536 (81)	1
The explanations given at that time were, *n* (%) (*N* fathers = 95 and *N* mothers = 1,930)			
Understandable	83 (87)	1,702 (88)	1
Adequate	77 (81)	1,424 (74)	1
These explanations were, *n* (%)			
Too technical (*N* fathers = 92 and *N* mothers = 1,870)	12 (13)	227 (12)	1
Of an appropriate technical level (*N* fathers = 92 and *N* mothers = 1,870)	76 (83)	1,533 (82)	
Insufficiently technical (*N* fathers = 100 and *N* mothers = 1,975)	5 (5)	79 (4)	
This interview generally gave confidence to the team, *n* (%) (*N* fathers = 95 and *N* mothers = 1,830)	87 (92)	1,683 (92)	1

are essential for successfully promoting a father's physical proximity with their infant. Father-infant closeness appears to be important in fostering the father-infant bond (8), mainly through care involvement and skin-to-skin contact (9, 11, 12, 31–33).

We found slight differences between fathers and mothers regarding their perceptions of feeling welcome in the NICUs and involvement in their infants' care. No previous studies have provided accurate comparisons of self-reported fathers' and mothers' involvement in the care of preterm infants. A few studies compared maternal and paternal perceptions on this topic, but these were not nation-wide studies and very few parents were interviewed (18, 31, 32, 34, 35). Some other studies on the topic considered only the father's views (9, 13, 16, 17, 19, 27, 33, 36). Altogether, these studies indicated that fathers needed explanations about the function of the NICU and its equipment and about the baby's sensory and relational abilities (31, 34, 36). Fathers reported feeling the need to be included early in care and to feel valued as a parent (35), in accordance with our results.

The responding fathers in our study were also very motivated to participate in care, notably expressing a desire to perform caring procedures that typically only a minority of parents perform. Moreover, fathers performed more technical procedures than mothers in our study. Interpreting this difference was difficult because (1) our study design did not allow for comparing mothers and fathers of the same infant; (2) we recruited parents from a large number of NICUs, which may differ in their parental integration policies; and (3) we could not exclude selection bias that favored the participation of fathers who wanted to participate in care (32). However the architectural design of a NICU and the level of implementation of infant-and family-centered care strategies could increase the involvement of parents and the frequency of skin-to-skin contact, particularly among fathers (25). In this case, the healthcare team plays an essential and supportive role (11, 16, 31, 35) in building trust and putting fathers at ease when providing care, such as skin-to-skin contact, and helping them be more involved with infant care (27, 33).

The need for fathers to be present near their partners and to be involved in infant care requires changes such as provision of supportive policies in the NICU and encouragement from healthcare teams of both parents to participate in their infants' care (31). Caregivers can have a decisive influence on fathers (31) and should be aware of the barriers that fathers have described such as the newborn's physical appearance, the technical NICU environment, and changes in parental roles, which may lead them

TABLE 5 Fathers' words about their perception and their welcome in the NICU.

Negative feelings	Surprised and lost	*“In the hours following the birth, one is ‘stunned’.” “The transfer from the NICU was abrupt and this memory will remain one of the most striking”*
	Not needed “technical” explanations	*“No impatience about the technical questions, which I knew I would have time to ask later, and if possible with the mother”*
	Ambivalence between health to the newborn and mother	*“Big ambivalence, between the fact of needing to accompany and be reassured on the state of the baby, and the anxiety of leaving the mother alone in the birth room”*
	Feeling of loneliness and anxiety in the absence of welcome	*“And when we arrived in the NICU, not a glance, not a word, we were spectators of the placement of our baby in incubator, that seemed so abrupt that we cried. For many minutes, everyone was around the incubator, without anyone noticing our presence”*
Positive supports	A human and benevolent welcome	*“The team was waiting for us” “The reception in the unit was of an exceptional quality. At the same time, it was dedramatizing, human, empowering and involving”*
	Parental involvement with feeling as “care partner”	*“I was taken into account” “They asked me what I wanted with my wife” “They said that we had a great role to play” “They involved us in every decision, and as a father, it feels good to be able to take on that role, they gave me room.” “The doctor immediately named me as Robin's father, thus helping me to take this place for my son. This was beneficial and involving.”*
	A team that takes the time and answers questions, in a comprehensible medical language	*“They never tried to hide information from us, they took our concerns into account, our questions were always answered.” “The nurse and the pediatricia, took the time to talk to us, to answer our questions and to talk about what was next without lying to us and without falling into incomprehensible medical language.”*

TABLE 6 Relationships of parents and caregivers.

	Fathers	Mothers	Adjusted *p*
Relationships with caregivers were satisfactory or very satisfactory, *n* (%)			
In neonatal intensive care unit (*N* fathers = 90 and *N* mothers = 1,795)	88 (98)	1,768 (98)	1
In neonatal ward (*N* fathers = 85 and *N* mothers = 1,795)	83 (98)	1,585 (88)	1
In kangaroo unit (*N* fathers = 23 and *N* mothers = 500)	23 (100)	440 (88)	1
Relationships with the health care team, *n* (%) (*N* fathers = 82 and *N* mothers = 1,736)			
Staff was available when parents requested them	74 (90)	1,495 (86)	1
Parents felt confident with the staff who cared for the infant	73 (89)	1,509 (87)	1
Parents felt comfortable asking questions or for clarification when explanations were unclear	67 (82)	1,425 (82)	1
Overall, explanations and parents' involvement were possible in decision-making regarding, *n* (%) (*N* fathers = 82 and *N* mothers = 1,728)			
The infant's well-being	68 (83)	1,452 (84)	1
The infant's health (treatment, ventilation, nutrition)	69 (84)	1,387 (80)	1
The organization of the infant's day (care schedules, skin-to-skin contact, etc.)	71 (87)	1,407 (81)	1
The team considered the parent's comments about the infant's health and well-being, *n* (%) (*N* fathers = 78 and *N* mothers = 1,690)	63 (81)	1,301 (77)	1
Overall, the information was consistent from one person to another, *n* (%) (*N* fathers = 82 and *N* mothers = 1,715)	67 (82)	1,372 (80)	1
The parent felt comfortable speaking openly to the healthcare team, *n* (%) (*N* fathers = 82 and *N* mothers = 1,700)	64 (78)	1,224 (72)	1
The reasons why they did not feel comfortable talking with the health care team, *n* (%)			
Difficulty in formulating their requests (*N* fathers = 5 and *N* mothers = 131)	4 (80)	105 (80)	1
Shyness (*N* fathers = 6 and *N* mothers = 159)	5 (83)	135 (85)	
Fear of being judged (*N* fathers = 6 and *N* mothers = 166)	5 (83)	141 (85)	
Lack of availability of the healthcare team (*N* fathers = 6 and *N* mothers = 173)	5 (83)	152 (88)	
Fear that openness will influence caregivers' relationships with the infant (*N* fathers = 7 and *N* mothers = 219)	7 (100)	197 (90)	
Possibility of attending physicians' visits to the infant, *n* (%) (*N* fathers = 83 and *N* mothers = 1,725)	62 (75)	1,190 (69)	1
It helped the parent (*N* fathers = 59 and *N* mothers = 1,142)	50 (85)	959 (84)	1
The parent missed it (*N* fathers = 21 and *N* mother s = 496)	17 (81)	372 (75)	1

TABLE 7 Fathers' feelings and words expressed about caregivers.

Positive feelings "What helped you the most during the newborn's hospitalization?"	*"Humanity"* *"Kindness"* *"Smile"* *"Professionalism"* *"Benevolence"* *"Moral support and comfort"* *"Availability"* *"Know-how"* *"Listening"* *"Involvement"* *"Reassurance,"* *"Presence and support"* *"Gentleness"* *"Empathy"*
Negative feelings	*"We were not really parents"* *"Under the control and judgment of the caregivers"* *"Caregivers contradicted each other in their methods"* *"We are nothing, not even the parents, nothing is chosen or asked of us"* *"Some caregivers purposely keep parents out of their child's care by telling us that we do not know how to do it"* *"No communication about the risks, the tests performed on the baby. We feel like spectators or even on the sidelines, powerless and passive"* *"Preference given to the mother systematically. Father's opinion not often asked"*

TABLE 8 Participation of parents in care of their infants.

	Fathers	Mothers	Adjusted *p*
Concerning the infant's care, *n* (%) (*N* fathers = 78 and *N* mothers = 1,580)			
Staff offered to participate in care as soon as possible	73 (94)	1,505 (95)	1
The first time parents were involved in care, the staff gave them confidence and accompanied them	76 (97)	1,493 (95)	1
Opportunity to take on the role of parent	67 (86)	1,285 (81)	1
Feeling of being judged by, *n* (%)			
Nurses (*N* fathers = 80 and *N* mothers = 1,606)	8 (10)	273 (17)	1
Physicians (*N* fathers = 83 and *N* mothers = 1,555)	5 (6)	171 (11)	1
Caregivers were able to respect preferences for the care that parents wanted to provide, *n* (%) (*N* fathers = 75 and *N* mothers = 1,530)	66 (88)	1,316 (86)	1
Caregivers were able to respect the time parents needed before they began to participate in their infant's care, *n* (%) (*N* fathers = 78 and *N* mothers = 1,572)	70 (90)	1,368 (87)	1

to feel they cannot adequately care for their infant (11–13). Caregivers must get to know each parent, learn about the type of involvement they want (13), and estimate the time they need before providing care (9, 32). The Family Initiative's International Neonatal Fathers Working Group drafted 12 practical recommendations to be used by neonatal teams to support the development of father-infant bonds and enable fathers to experience more equal co-parenting. These recommendations included assessing the needs and wishes of the father; ensuring flexibility and ease of access to the neonatal unit for fathers; providing information about infants directly to fathers, not exclusively through mothers (37); and providing information in real time (16, 35). Research has recommended encouraging the presence and care involvement of fathers and supporting them during their transition to fatherhood (31, 37). Other research reported that fathers felt the need to meet with other fathers who would better understand their own fears and difficulties (37). This could be facilitated by the establishment of discussion groups for fathers. Some authors have proposed the creation of interactive social media support that provides fathers with electronic updates on their infants' health condition and allows communication with other fathers (35).

One highly effective procedure is to encourage the presence of fathers in family rooms during the entire hospitalization period, a strategy with proven short-term benefits for the infant (38). This approach is also supported by the implementation of infant-and family-centered care, which has widespread social support at the national level (39). European countries have disparities in the social support and parental leave policies provided to new parents (40), with Nordic countries providing the most generous benefits.

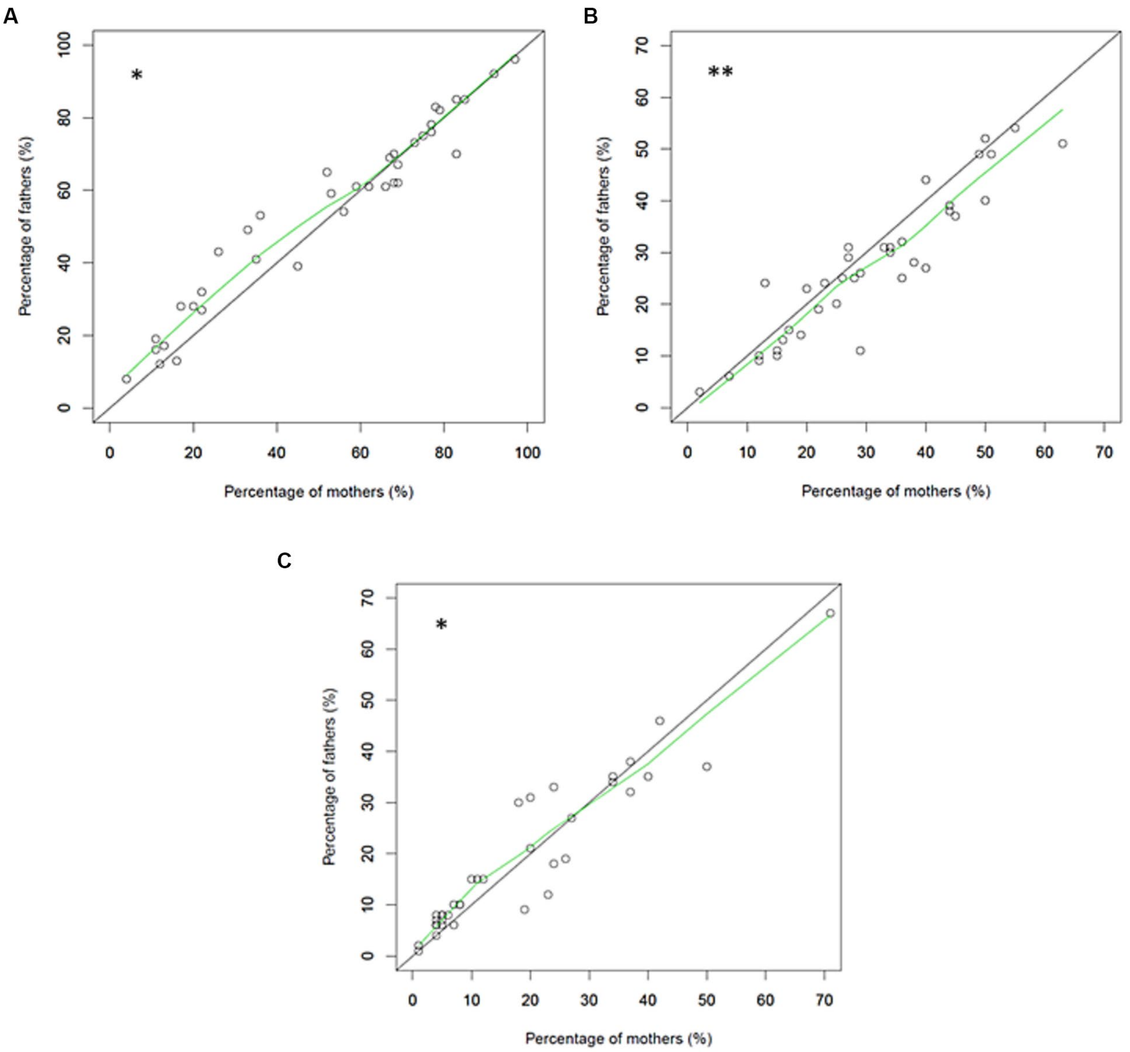

FIGURE 2
Relationships of the percentages of mothers and fathers who performed different caring procedures **(A)**, who did not perform different caring procedures that they wanted to perform **(B)**, and who did not perform different caring procedures that they did not want to perform **(C)**.

However, most member states of the European Union now provide statutory parental leave (41). They aim at providing greater support for fatherhood an promoting more gender equality. The European Standards of Care for Newborn Health called for continuous parental support and access as well as high parental involvement in the care of newborns (42). There are similar recommendations in France (7), and the laws in France regarding social support for fathers of very preterm infants have evolved. Since July 1, 2019, all fathers whose newborn infants require immediate hospitalization at birth receive 30 days of paternity leave (43) in addition to the 25 days allocated to all fathers since July 2021 (44). Before that last date, and at the time of the survey, only 11 days were allocated. Fathers in France are now able to spend more time with their preterm infants. In view of the demonstrated benefits of father's involvement in the care of their very preterm infants through early interventions, there is a need to further support this evolution in every country (45).

Large prospective studies are necessary to evaluate fathers' presence and perceptions regarding their very preterm newborns. Quantitative studies should measure actual involvement during the whole duration of hospitalization, and qualitative studies should assess the feelings and needs of fathers. Future research should also examine the mothers and fathers of VPIs who have lower socioeconomic status because these parents may have different needs.

5 Conclusion

Most fathers were present at the births of their VPIs, but less than half were near the mother at this time. Only a small number of fathers reported no separation from their infants, although most met their infants during the first day of life. Less than two thirds of the fathers accompanied their infants on transfer to the NICU. Altogether, these data indicate room for improvement to meet the specific needs of fathers in the broad context of infant-and family-centered developmental care for premature infants. The continuation of this online questionnaire will allow for assessing future progress.

TABLE 9 Participation of parents in skin-to-skin contact with their infants.

	Fathers	Mothers	Adjusted *p*
Number of parents informed about the benefits of skin-to-skin contact, *n* (%) (*N* fathers = 74 and *N* mothers = 1,530)	70 (95)	1,443 (94)	1
Either parent was able to practice skin-to-skin contact with the infant at least once, *n* (%) (*N* fathers = 74 and *N* mothers = 1,530)			
No, neither parent did	1 (1.3)	26 (1.7)	0.002
Yes, the other parent did	5 (6.8)	12 (0.8)	
Yes, both parents did	63 (85)	1,242 (81)	
Yes, I did	5 (6.8)	267 (17)	
Infant's age when they first had skin-to-skin contact, *n* (%) (*N* fathers = 68 and *N* mothers = 1,510)			
1 to 3 days	28 (41)	652 (43)	1
3 to 7 days	23 (34)	441 (29)	
1 to 2 weeks	4 (6)	195 (13)	
2 to 3 weeks	7 (10)	100 (7)	
> 3 weeks	6 (9)	121 (8)	
Concerning this first skin-to-skin contact, the parent thought that, *n* (%) (*N* fathers = 68 and *N* mothers = 1,468)			
It was the right time	59 (87)	1,128 (77)	0.63
It was offered too soon	1 (1.5)	51 (3.5)	
It was offered too late	7 (10)	296 (20)	
Number of parents who would have liked to have skin-to-skin more often, *n* (%) (*N* fathers = 65 and *N* mothers = 1,468)	37 (57)	1,087 (74)	0.026
The duration of skin-to-skin contact times was generally determined by, *n* (%) (*N* fathers = 68 and *N* mothers = 1,510)			
Parents	17 (25)	358 (24)	1
Nurse	19 (28)	329 (22)	
Physician	3 (4.4)	165 (11)	
Mutual agreement between the health care team and parents	5 (7.4)	83 (5.5)	
The infant's state of health	24 (35)	572 (38)	
Number of parents who felt confident during skin-to-skin contact, *n* (%) (*N* fathers = 68 and *N* mothers = 1,505)	65 (95.6)	1,404 (93)	1
Parents' preference to have their infant, *n* (%) (*N* fathers = 68 and *N* mothers = 1,508)			
In skin-to-skin contact	6 (9)	80 (5.3)	1
In their arms	48 (71)	1,160 (77)	
No preference	14 (21)	267 (18)	

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving humans were approved by Comité d'Ethique de la Faculté de Médecine et d'Odontologie de l'Université de Strasbourg. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation was not required from the participants or the participants' legal guardians/next of kin in accordance with the national legislation and institutional requirements.

Author contributions

AS-D analyzed the data, reviewed the literature, and wrote the first draft of the manuscript. IL conducted the extraction and the analyses of the data and generated the figures and tables of the manuscript. LC, OD, and JS discussed the interpretation of the results and reviewed the manuscript. MA, AR, CB, and AE contributed actively to the conceptualisation, the accessibility and dissemination of the GREEN parental questionnaire. CT supervised the conceptualisation and dissemination of the GREEN parental questionnaire and discussed the results, and reviewed the manuscript. PK contributed to the conceptualisation of the GREEN questionnaire, designed the study, supervised the analyses, and thoroughly reviewed the final manuscript. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Investigating the borders of autism spectrum disorder: lessons from the former diagnosis of pervasive developmental disorder not otherwise specified

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Introduction: Autism Spectrum Disorder (ASD) diagnosis is relatively consensual in typical forms. The margins of the spectrum and their degree of extension, however, are controversial. This has far-reaching implications, which extend beyond theoretical considerations: first, peripheral forms of autism are more prevalent than central forms; second, we do not know how relevant typical-targeted recommendations are for atypical forms. In DSM-IV-TR, these margins of autism were studied within the category of Pervasive Developmental Disorder – Not Otherwise Specified (PDD-NOS). In spite of its low reliability, this former diagnosis was of particular interest to shed light on the gray area of margins. The aim of this systematic is therefore to investigate the clinical characteristics of PDD-NOS in comparison with Autistic Disorder.

Method: A stepwise systematic PRISMA literature review was conducted by searching PubMed and Web Of Science databases to select corresponding studies.

Results: The systematic review included 81 studies comprising 6,644 children with PDD-NOS. Cross-sectional and longitudinal studies comparing PDD-NOS and AD showed that PDD-NOS corresponds to milder form of autism with less impact and less associated disorder, with the exception of schizophrenia and mood disorder.

Discussion: Our review challenges initial views of PDD-NOS, and shows the clinical relevance of this diagnosis when dealing with the margins of autism, and the *de facto* diversity included in the spectrum. However, in view of the many limitations of PDD-NOS (low reliability, instability through time, low acceptability), we suggest taxonomic changes in DSM-5: we introduce a new category based on three main dimensions related to socialization impairment, emotional lability and psychotic symptoms.

Conclusion: Our review argues for a distinction between AD and PDD-NOS on clinical characteristics and thus highlights the need to study the margins of autism. While the limitations of the PDD-NOS category made it irrelevant to investigate these margins from a research perspective, we believe that a multidimensional approach for mental health professionals taping socialization, emotion lability and psychotic symptoms would be interesting. Our review therefore encourage future studies to test relevant criteria for a new category and possibly identify developmental trajectories, specific interventions and treatments.

KEYWORDS

autism spectrum disorder, pervasive developmental disorder, DSM-IV-TR, DSM-5, pathological demand avoidance, diversity, heterogeneity

Introduction

Since 2013 and the DSM-5, the diagnostic category of Autism Spectrum Disorder (ASD) has included 2 behavioral dimensions which represent the core defining features of ASD (1): (a) communication and social interaction deficits on the one hand, and (b) repetitive behaviors and restricted interests on the other hand (1). The term "ASD" suggests that the core features of the disorder can be measured dimensionally and, that they fall along a continuum of severity. No diagnostic subtypes are listed (1); instead, specifiers are provided to indicate associated dimensions, such as intellectual and/or language impairment, as well as the severity level of core ASD symptoms (2). Further, any known genetic or medical disorders are recorded and other co-occurring neurodevelopmental, mental, or behavioral disorders are indicated (2) to characterize subgroups.

In the DSM-5 (2), whatever the level of severity, even in the mildest forms, ASD symptoms are viewed as intrinsically autistic. However, to take one example, this conceptualization does not take into account the overlap between, on the one hand, some ASD symptoms, and on the other, Intellectual disability (3, 4), language impairment (5, 6), Attention Deficit/Hyperactivity Disorder (7, 8) or schizophrenia (9, 10). Moreover, mild ASD symptoms might be more akin to a natural variant of typical development than a formal disorder (11). Boundaries issues with typical development are also longitudinal, as has been shown in studies on "optimal outcome" or "Loss of Autism Diagnosis (LAD)" (12). These studies (13) focused on a group of individuals meeting criteria for ASD in childhood who no longer met them later in development. This definition entailed clear documentation of early ASD diagnosis, not meeting current diagnostic criteria (with ADOS scores similar to neurotypical peers with no history of ASD), and having overall cognitive, language, and social functioning standardized test scores within the average range (12). Confirmation that meeting criteria for ASD is not necessarily a lifelong state appears evident in the strikingly similar proportions (about 9%) of individuals experiencing LAD in prospective and retrospective studies (12). In line with the scope of our review, it is worth noticing that one study (14) showed that among ASD children included at age 2, those with PDD-NOS were significantly more likely than those with AD to move off the spectrum by age 4. ASD symptoms might be thus less specific than suspected in the DSM-5, especially in milder forms.

Envisioned in line with Kannner's (15) first description and seminal work, autism is relatively consensual in its typical forms (full syndrome) (16). However the margins of the spectrum and their degree of extension are controversial (17) because of overlaps with other dimensions and blurred boundaries with typical development. This question is much more than a theoretical issue, since peripheral or "marginal" forms of autism might be more prevalent than central forms (see below). Furthermore, one could question the usefulness and relevance of recommendations made for typical forms when facing atypical ones. Past conceptualizations (18) contrasted the more prototypical form of autism, called "Autistic Disorder (AD)," with the margins of autism studied within the Pervasive Developmental Disorder - Not Otherwise Specified (PDD-NOS) category. Is therefore of special interest to explore these margins through the lens of this former diagnosis.

The PDD-NOS diagnosis category was first introduced in 1987 (19) to contrast with typical forms for autism labeled "Autistic Disorder" (AD). The DSM-III-R (19) states that *"this category should be used when there is a qualitative impairment in the development of reciprocal social interaction and of verbal and nonverbal communication skills, but the criteria are not met for autistic disorder, schizophrenia or schizotypal or schizoid personality disorder. Some people with this diagnosis will exhibit a markedly restricted repertoire of activities and interests, but others will not."* In DSM-IV (20) and DSM-IV-TR (18), AD and PDD-NOS were included within the Pervasive Developmental Disorders (PDDs) that prelude ASD. PDDs also included Asperger Syndrome (AS), Childhood Disintegrative Disorder and Rett Syndrome (18, 20).

In DSM-IV-TR (18), the PDD-NOS category included atypical autism, i.e., presentations that did not meet the criteria for AD because of late age of onset, atypical symptomatology, or subthreshold symptomatology, or all of these. These recommendations echoed Towbin's conceptualization (21) described in a reference textbook section published in 1997. He recommended the use of PDD-NOS in 4 indications: (a) as a temporary diagnosis when data are lacking or when the child is too young; (b) to designate a mild form of ASD; (c) in the case of late age of onset of autistic symptoms; (d) to depict a clinical picture with early symptom onset and impairment in social reciprocity (21). In terms of diagnostic use, the PDD-NOS, often envisioned as a diagnosis of subthreshold autism, seems to cover a wide range of clinical variability, sometimes referred to as heterogeneity; it would benefit from being studied in a more positive manner, by drawing on its clinical diversity (22).

Epidemiological studies show that at least half of PDD were PDD-NOS. In 2005, Fombonne (23) found a PDD-NOS prevalence of 37.1 per 10,000 for a PDD prevalence of 63 per 10,000 and an AD prevalence of 13 per 10,000. In 2010 in the United States, the results of the Centers for Disease Control and Prevention (CDC) showed a

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Abbreviations: ASD, Autism Spectrum Disorder; ADHD, Attention Deficit Hyperactivity Disorder; DSM, Diagnostic and Statistical Manual; ID, Intellectual Disability; MCDD, Multiple Complex Developmental Disorder; MDI, Multi-Dimensional Impairments; PDA, Pathological Demand Avoidance; PDDs, Pervasive Developmental Disorders; PDD-NOS, Pervasive Developmental Disorder - Not Otherwise Specified; PRISMA, Preferred Reporting Items for Systematic Reviews and Meta-Analyses; SCD, Social Communication Disorder.

PDD prevalence of 14.6 per 1,000 in a population of 363,749 8-year old children (24). PDD-NOS were 46% of the PDD, AD 43% and AS 11%. Noticeably, between 2006 and 2010 the distribution of AD and PDD-NOS has not undergone substantial changes (24).

The diagnosis of PDD-NOS had a low inter-rater reliability. The inter-rater reliability was good to excellent (*k* from 0.95 to 0.67) for the distinction between PDD and non-PDD. However, the disentanglement of different subtypes of PDD was much less reliable (*k* from 0.18 to 0.65) (25–27). The DSM-IV-TR criteria were therefore more successful in differentiating PDD (25) from other psychiatric disorders than in distinguishing between the three main subtypes of PDD, namely AD, PDD-NOS and AS (28). In line with these results, a study (29) tried to assess the variation between behavioral phenotypes and clinical diagnoses of different autism spectrum disorders across 12 university-based sites. They found that clinical distinctions among categorical diagnostic subtypes of PDDs (AD, PDD-NOS and AS) were not reliable even across sites with well-documented fidelity, using standardized diagnostic instruments (29).

In spite of its low reliability, the former diagnosis of PDD-NOS is very interesting to study the margins of autism because of both its conceptualization at the end of the spectrum, and its high prevalence. The aim of our systematic review is thus to investigate the clinical characteristics of PDD-NOS in comparison with AD. The objectives were therefore to study comparatively (in both diagnoses) synchronic and diachronic characteristics such as autism symptoms, cognitive function, daily impact, associated mental health or physical health condition and developmental trajectory from infancy to adolescence. The comparison is intended to address the distinction between the typical form and the margins of autism as well as the relevance of the PDD-NOS/AD distinction. Our study therefore aims to determine whether this distinction helps mental health professionals to account for diversity within the spectrum, and to identify divergent trajectories, better than the current ASD category.

Materials and methods

This study follows the Preferred Reporting Items for Systematic Reviews and Meta-analyses reporting guidelines (PRISMA) (30).

Search strategy and selection criteria

A stepwise systematic literature review (30) was conducted by searching PubMed, MEDLINE, and Web Of Science databases for published peer-reviewed papers using the following keywords: "PDD-NOS" OR "Pervasive Developmental Disorder Not Otherwise Specified" from January first 1987 (the year of the DSM-III-R publication, introducing this diagnostic category for the first time) to June 16th 2022. The keywords were screened in the titles and abstracts.

Eligibility criteria

Inclusion criteria

To be included in this review, the articles had to meet all following criteria: (a) include children and/or adolescents (31), (b) compare toddlers, children or adolescents with PDD-NOS with children with AD (c) be published in English, (d) use a quantitative design. RC and OP applied the eligibility criteria and screened the records to select included studies.

Exclusion criteria

The following exclusion criteria were used: (a) no reviews, comments or clinical cases, (b) methodological features: number of participants included in the AD or the PDD-NOS groups ≤10, adults included only, lack of comparison group, comparison with a group different from AD (e.g., AS), participants whose diagnosis has been confirmed by a classification other than the DSM-III or the DSM-IV, test validation studies, (c) epidemiological data in order to focus on mostly clinical well-characterized populations, (d) therapeutic trials, (e) anatomical, biological or fMRI studies (isolated, contradictory and/or non-replicated results) are (difficult to interpret), (f) language other than English, and (g) qualitative studies.

Outcome measures and data extraction

The following variables were extracted: First author (second, third author and journal if needed to distinguish from other studies); Year of publication; Sample Size; Age (mean ± SD); Sex (% Males); Functioning level when available and when age at inclusion (at baseline in longitudinal studies) > 6 y.o. (to assess comorbid ID); Objective; Design of the Study; Methods (only with longitudinal studies); Main assessment tools; Results.

Quality assessment

All included studies were observational, with a similar comparative framework. Quality assessments in such observational studies is controversial, with no clear agreement on rating methods (32). We therefore did not assess quality.

Results

Database

Our search from 1987 to 2022, undertaken on June 16th 2022, found 510 articles on PubMed and 431 on Web of Science. After manually removing all duplicates, 681 references remained. Based on their titles and abstracts, 256 papers were excluded for lack of relevance as they displayed no separate groups for AD and PDD-NOS. Most of these articles focused on PDDs (not PDD-NOS), ASD or grouped PDD-NOS, AD and/or AS into common groups. Other articles studied PDD-NOS in specific groups such as X-Fra, cerebral palsy, Down Syndrome... Our search strategy yielded 425 full-text articles assessed for eligibility. After conducting a full-text analysis of all these papers and excluding those not meeting our inclusion criteria, we ended up with 81 relevant studies, 67 of which were cross-sectional and 14 longitudinal.

Studies

A total of 6,644 children with PDD-NOS and 11,156 children with AD were included. 47 studies were from the United States (58%), 12 from Netherlands (15%), 5 from Turkey (6%), 3 from Italy (4%), and 3 from Sweden (4%) The mean age in each group (when available) was 5.85 and 5.84 years, respectively. The mean age was low because many studies included toddlers or/and pre-schoolers with PDD-NOS (52%). Sex was not systematically specified in each group. Based on available data, PDD-NOS groups comprised 75.1% of males. IQ was not always specified, in particular in studies in toddlers or preschoolers. Based on available IQ measures, average IQ was 81.93.

Within the 67 selected cross-sectional studies, we distinguished those relating to (a) autism symptoms (22 studies), (b) perinatal, developmental and functional aspects (31 studies), (c) associated mental health conditions (14 studies) and (d) associated physical health conditions (6 studies). We chose these categories in part because they best served our goal, of investigating the clinical characteristics (autism symptoms, associated conditions/dimensions, developmental trajectories and adaptive behavior) of PDD-NOS in comparison with AD, and in part because they were those that best matched selected studies. All cross-sectional studies could be included in these three categories. As explained above, 14 longitudinal studies were also selected (Figure 1).

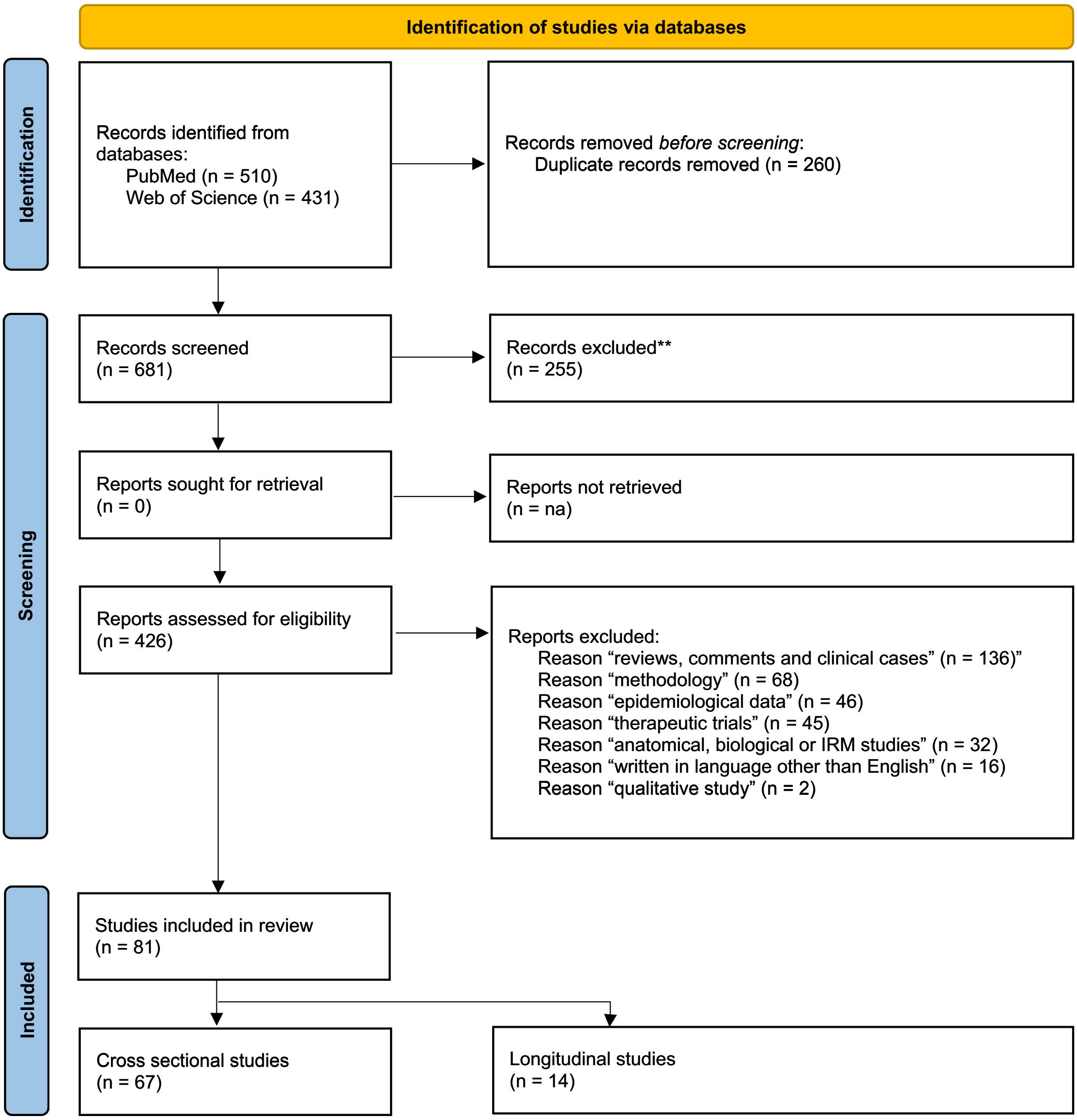

FIGURE 1
PRISMA flowchart.

The total number of the studies presented in the corresponding tables (Tables 1–5) is 87 and not 81 because Carigi et al.'s study informed the 4 domains, and was thus selected in the corresponding categories (33). Walker et al. study informed 2 domains (34). Finally, Mandy et al.'s study informed 3 domains (35).

Cross-sectional studies

Autism symptoms

Studies on autism symptoms in PDD-NOS in comparison with AD deeply investigated the social-communication domain. 13 studies (33, 34, 36–46) (Table 1) showed a lesser socialization impairment whereas 3 studies (including one on gaze performance) (47–49) did not show any differences (Table 1).

7 studies found lower restrictive and repetitive behaviors (34, 35, 42, 43, 46, 50, 51) (Table 1). However, 3 studies did not show such a difference (33, 41, 52) (Table 1).

The mean age of onset of parental concerns in the group with AD was comparable to that reported in the PDD-NOS group (53). These parental concerns in PDD-NOS referred to non-specific problems linked to sleeping, eating, and level of activity (53).

Perinatal, developmental and functional aspects

Three studies (54–56) showed distinct perinatal risk factors in PDD-NOS in comparison with AD (Table 2). However, involved factors differed: hyperbilurinemia (55), higher exposure to smoking during pregnancy (56), threatened abortion, a labor duration of less than 1 h, cephalopelvic disproportion, and deliverance by an elective or emergency cesarean section (54). Even if perinatal risk factors might differ, odds ratios for birth defects were similar for AD and PDD-NOS (57).

Early development was less impacted in PDD-NOS with respect to AD (Table 2). Pointing was less impaired in PDD-NOS than in AD (33, 58). Skill loss was less frequent in PDD-NOS (58, 59). Children with AD began crawling significantly later than those with PDD-NOS (60). Using the Revised Educational Profile, Portoghese et al. showed that all behavioral areas and almost all assessed developmental domains (with the exception of the cognitive verbal) were more severely impaired in the AD group than in the PDD-NOS group (61). Children with AD and ID displayed a Visual Rooting Reflex more often than children with PDD-NOS and ID (62), suggesting less severe neurological impairments.

The investigation of language development in PDD-NOS in comparison with AD did not show any difference or particularity (63, 64) (Table 2). Also, fine and gross motor development did not differ between the 2 groups (35, 65). However, Ghaziuddin et al. found lower clumsiness in PDD-NOS (66).

Regarding socio-emotional development, social cognitive performance did not differ between both groups (67). One study shows a distinct profile of emotional information treatment in PDD-NOS in comparison with AD (68) (Table 2).

Children with PDD-NOS and AD did not differ with respect to global intellectual functioning (35, 69, 70). However, one study (35) showed higher visio-spatial performance in PDD-NOS when others did not (71, 72) (Table 2).

Executive functions lead to contradictory results, showing lesser impairment in PDD-NOS (73, 74) or no difference (75) (Table 2).

Sensory profile did not differ between the two groups (76).

Adaptive function was higher in PDD-NOS in 2 studies (34, 77). However, other studies did not show any difference (78, 79) (Table 2).

Quality of life of children and parents was less impaired in the PDD-NOS groups (33, 80) (Table 2).

Only one study showed an impact of sex on AD vs. PDD-NOS (81).

Associated mental health conditions

Children with PDD-NOS showed less challenging behavior than those with AD (82–86). Symptoms related to ADHD were overrepresented in the AD group (38, 85) (Table 3). Studies showed more Conduct Disorder (87) and Oppositional Defiant Disorder (88) in the PDD-NOS group.

Anxiety led to contradictory results (Table 3). Anxiety has been shown to be higher in the AD group (83, 85, 89) or in the PDD-NOS group (90). A study investigated the interaction between diagnosis (AD or PDD-NOS) and communication deficits (91). Children with AD experienced less anxiety as communication deficits increased while children with PDD-NOS experience more anxiety as communication deficits increased highlighting distinct type of interaction (91).

Chien et al. showed that children with PDD-NOS had a higher risk for developing Schizophrenia Spectrum Disorder, Bipolar Disorder, Major Depressive Disorder than those with AD (92). These results concur with another study comparing youth with ASD attending a specialty clinic to those attending a general psychiatry clinic (88). The latter group suffered from a greater burden of psychopathologies (Major Depressive Disorder and psychosis), higher levels of dysfunction and more pharmacotherapy [*cf* Carigi et al. (33); Table 3].

Associated physical health conditions

Studies showed less sleep disorders in the PDD-NOS-group (33, 93, 94) but another did not (35) (Table 4). Feeding disorders were less common in the PDD-NOS group than in the AD group (35, 93, 94). However, one study did not confirm this result (85) (Table 4).

Parmeggiani et al. showed that pathological neurological examination, cerebral lesions, abnormal EEG background activity and associated genetic pathologies were more common in PDD-NOS. Moreover, Epilepsy seizure in PDD-NOS had an earlier onset (2 years 8 months) and a better outcome (95).

Longitudinal studies

Longitudinal studies showed lower diagnosis stability in PDD-NOS than in AD (27, 96–99) with the exception of Chawaska et al. study (100) (Table 5).

Cognitive development was less impaired in PDD-NOS (Osada et al., Takeda et al., Thurm et al.) (101–103). However, another study did not show such a better outcome in this group (96).

As a whole, autistic symptoms were lower in the PDD-NOS group than in the AD group (96, 101) (Table 5). More specifically, studies showed better communication and socialization skill outcomes in PDD-NOS than in AD (103–105). Kim et al.'s study showed that the likelihood of having restrictive and repetitive behavior was the same for both AD and PDD-NOS groups; though the rated severity of these

TABLE 1 Autism symptoms in PDD-NOS in comparison of AD.

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Functioning level	Objective	Study design	Main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Carigi et al.	2014	46	71	AD: 67.78 months (39.59)[1] PDD-NOS: 45.20 months (17.28)	56	79	Italy	DQ[2]: 59.86 (15.42)	To identify clinical features that might prove able to differentiate AD from PDD-NOS	Comparative study	The ADI-R[3] The ADOS[4] The CARS[5] The Griffith's Mental Development Scale	Higher scores were rated on the ADI-R (with the exception of restrictive and repetitive behavior), on the ADOS and on the CARS in AD in comparison with PDD-NOS.
Mahoney et al.	2014	72	105	AD: 53.12 months (18.31) PDD-NOS: 52.86 months (18.47)	86	82	USA		To characterize and compare social interaction profiles of young children with AD and PDD-NOS	Comparative study	The Screen for Social Interaction	Children with AD and those with PDD-NOS had similar social interaction profiles
Joseph et al.	2013	128	46	AD: 4.14 years (1.57) PDD-NOS: = 4.14 (1.57)	41	89	USA	Non-Verbal Developmental Quotient: 80.37 (17.60)	To investigate restricted, repetitive and stereotyped patterns of behavior, interests and activities in children with AD and those with PDD-NOS	Comparative study	The Repetitive Behavior Scale-Revised	The AD group did not differ from PDD-NOS on any subscale (Self-Injurious Behavior, Stereotyped Behavior, Compulsive Behavior, Ritualistic/Sameness Behavior and Restricted Behavior).
Matson et al.	2013	115	113	26.63 months (5.22)			USA		To compare ASD symptoms in children with AD and those with PDD-NOS	Comparative study	the BISCUIT[6]-Part 1	The AD group had higher rates of ASD symptoms than the PDD-NOS group in all three domains (repetitive behaviors, social interaction and communication)

(Continued)

TABLE 1 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Functioning level	Objective	Study design	Main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Kozlowski et al.	2012	20	20	8.37 years (3.22)	13	65	USA	No ID	To determine differences between AD and PDD-NOS on appropriate and inappropriate social skills	Comparative study	The Matson Evaluation of Social Skills in Youngsters-II	Social skills did not differ between children with AD and PDD-NOS
Falck-Ytter et al.	2012	40	25	AD: 2107 days (339) PDD-NOS: 2224 days (252)			Sweden	Total IQ: 84.15 (10.53)	To compare gaze performance in children with AD and those with PDD-NOS	Comparative study	Eye tracking The Wechsler Preschool and Primary Scale of Intelligence – Third Edition.	Gaze performance did not significantly differ between the AD and the PDD-NOS group
Hattier et al.	2011	197	197	AD: 26.59 months (4.75) PDD-NOS: 25.54 (4.48)	143	73	USA		To examine whether the AD group would have significantly greater levels of impairment in communication and socialization than the PDD-NOS	Comparative study	The BISCUIT-Part1 The Battelle Developmental Inventory, 2nd Edition	In regards to communication, no significant differences were found between children with AD and PDD-NOS. For socialization, the comparison AD vs. PDD-NOS were found to differ significantly
Karabekiroglu	2012	47	94	AD: 4.1 (2.0) PDD-NOS: 5.4 (2.4)	71	76	Turkey		To investigate differential features of PDD-NOS such as presenting symptoms, developmental history, and comorbidity with respect to autism	Comparative study	The K-SADS[7] The First Clinical Admission Questionnaire and Clinical Assessment Form Preliminary PDD-NOS symptom screening scale	The most common presenting symptoms in the PDD-NOS and AD groups are reported significantly less in prevalence in the PDD-NOS group

(Continued)

TABLE 1 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Functioning level	Objective	Study design	Main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Mandy et al.	2011	97	66	AD: 9.43 years (3.50)[2] PDD-NOS: 9.05 years (3.51)	52	79	UK	Verbal IQ: 87.60 (15.98)	To characterize a group of individuals diagnosed according to a clearly operationalized DSM-IV-TR definition of PDD-NOS	Comparative study	The Developmental, Dimensional and Diagnostic Interview The ADOS	The AD and PDD-NOS groups did not differ significantly according to reciprocal social interaction or social communication measures. The PDD-NOS group had less repetitive, stereotype behaviors than the AD group.
Sipes et al.	2011	270	270	17 to 34 months			USA		To examine the effects of a diagnosis of ASD (AD or PDD-NOS) and motor skills impairments and their interaction on social skills impairments	Comparative study	The Biscuit-part 1 The Battelle Developmental Inventory–Second Edition	Those with AD had significantly higher (i.e., more impaired) social skill impairments than those with PDD-NOS. Level of fine motor skills affected social skills more so in those with AD than in those with PDD-NOS
Fein et al.	2010	15	13	10.42 years (2.2)			USA		To compare the rates of contagious yawning in a group of children with AD and PDD-NOS	Comparative study	A contagious yawning experiment	Children diagnosed with AD were less likely to exhibit contagious yawning than children diagnosed with PDD-NOS

(Continued)

TABLE 1 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Functioning level	Objective	Study design	Main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Horovitz et al.	2010	220	220	AD: 27.22 months (4.74) PDD-NOS: 26.44 (4.77)	156	71	USA		To compare communication impairments of toddlers diagnosed with AD and PDD-NOS	Comparative study	The BISCUIT-Part 1	Toddlers with AD had more total communication impairments than did toddlers with PDD-NOS
Fodstad et al.	2009	161	140	26.53 months (5.02)			USA		To determine the nature and rate of verbal and non-verbal communication as well as social skills	Comparative study	The BISCUIT part 1	Deficits in communication and social skills were more obvious and pronounced in those with AD, then PDD-NOS
Matson, Dempsey & Fodstad (*Developmental Neurorehabilitation*)	2009	140	121	26.63 months (5.12)			USA		To determine the nature and rate of stereotypies and ritualistic behaviors in a population of infants with AD or PDD-NOS	Comparative study	The BISCUIT	Stereotypies and repetitive/ritualistic behaviors were most common in AD, then PDD-NOS
Matson, Dempsey & Fodstad (*Journal of Developmental and Physical Disabilities*)	2009	69	34	7.99 years (3.25)			USA		To distinguish between AD and PDD-NOS To evaluate specific diagnostic profiles within and across ASD is also critical	Comparative study	The Autism Spectrum Disorders-Diagnostic for Children	Severity of impairment was greatest for AD, followed by PDD-NOS. AD and PDD-NOS differed on Social Relationships and Verbal Communication but not on Insistence on Nonverbal Comm./ Socialization, Sameness/Restricted Interests

(Continued)

TABLE 1 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Functioning level	Objective	Study design	Main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Matson, Fodstad & Dempsey	2009	171	144	26.50 (4.97)			USA		To distinguish between a diagnosis of AD from PDD-NOS in a sample of infants and toddlers	Comparative study	The BISCUIT-Part 1	The study identified 11 items that distinguished young children with autism from those with PDDNOS. This items were engages in repetitive motor movements for no reason, use of language in conversations, shares enjoyment with others, interest in participating in social activities, restricted interest and activities, sticking to purposeless odd routines or rituals, abnormal preoccupation with object parts, reads non-verbal cues of others, use of non-verbal communication, abnormal of repetitive hand or arm movements and development of social relationships

(Continued)

TABLE 1 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Functioning level	Objective	Study design	Main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Chawarska et al.	2007	51	24	AD: 28.5 months (4.7) PDD-NOS: 27.8 months (5.7)			The ADI-R	Mullen visual reception DQ: 84 (16)	To examine factors that influence the timing of parental recognition of developmental problems including severity of autistic symptoms	Comparative Study	The ADI-R The Mullen Scales of Early Learning	The mean age of onset of parental concerns in the group with AD was comparable to that reported in the PDD-NOS group. Parents of children with AD were significantly more likely to report their children as having medical problems and motor delays, as well as unusual autistic-like stereotyped behaviors than parents of children with PDD-NOS. Children with PDD-NOS had more non-specific problems related to sleeping, eating, and activity level

(Continued)

TABLE 1 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Functioning level	Objective	Study design	Main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Verté et al.	2006	57	31	AD (without ID): 8.8 years (1.8) PDD-NOS: 8.8 years (1.6)	26	84	The Netherlands	Full Sclale IQ: 98.0 (14.7)	To explore whether children with AD (without ID) and PDD-NOS can be differentiated in terms of their Children's Communication Checklist profile	Comparative Study	The Children's Communication Checklist	The AD (without ID) group had more problems of coherence than the PDD-NOS groups. The AD (without ID) group had lower conversational rapport scores than the PDD-NOS Group. The PDD-NOS group had fewer restricted interests than the HFA and AS groups. The HFA group had a lower average pragmatic composite score than the PDD-NOS group
Walker et al.	2004	216	21	AD: 86.3 months (38.0) PDD-NOS: 99.0 months (71.8)	18	86	USA	Leiter IQ: 82.38 (33.40)	To describe the characteristics of children with PDD-NOS, diagnosed using a consensus best estimate	Comparative study	The ADI-R The ADOS The ABC[8] The Leiter International Performance Scale	Children with PDD-NOS have fewer autism symptoms than children with AD.

(Continued)

TABLE 1 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Functioning level	Objective	Study design	Main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Buitelaar et al.	1999	205	80	Pre-school children to young adults	69	86	The Netherlands	ID in 25% of cases	To explore the boundaries of PDD-NOS vis à vis AD	Comparative study	ICD 10/DSMIV criteria of AD and PDD-NOS	The three items with the strongest differentiating power were (a) lack of varied spontaneous make-believe play, (b) preoccupation with restricted patterns of interest, and (c) impairment of play before age 3 years
Klin et al.	1999	34	34	AD: 7.37 years (2.93) PDD-NOS: 6.59 years (2.01)			USA	Non-Verbal Mental Age: 5.19 years (1.00)	To explore whether or not any deficits found in face recognition applied to AD and PDD-NOS	Comparative study	The K-ABC	Children with AD performed significantly worse on face recognition than the PDD-NOS group
Mayes et al.	1993	40	40	AD: 6.9 years (5.4) PDD-NOS: 6.6 years (4.8)	33	75	USA	Mental age: 3.74 (2.60)	The delineation, of specific clinical features that differentiate PDD-NOS from AD	Comparative study	24 items selected from the DSM III-R criteria for AD, the ICD-10 Draft Research Criteria for childhood autism, the ABC and the VABS	Items that distinguished AD with PDDNOS from those with autism related to the degree of socialization and relatedness with children with PDD-NOS showing less severe disturbances in relatedness

[1](SD). [2]Developmental Quotient. [3]Autism Diagnostic Interview -Revised. [4]Autism Diagnostic Observation Schedule. [5]Child Autism Rating Scale. [6]Baby and Infant Screen for Children with aUtIsm Traits. [7]Schedule for Affective Disorders and Schizophrenia for School-Age Children. [8]Vineland Adaptive Behavior Scale. [9]Autistic Behavior Checklist.

TABLE 2 **Perinatal, developmental and functional aspects in PDD-NOS in comparison of AD.**

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Functioning level	Objective	Study design	Main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Carigi et al.	2014	46	71	AD: 67.78 months (39.59)[1] PDD-NOS: 45.20 months (17.28)	56	79	Italy	DQ[1]: 59.86 (15.42)	To identify clinical features that might prove able to differentiate AD from PDD-NOS	Comparative study	The CGI[2] The Griffith's Mental Development Scale The Health Related Quality of Life Questionnaire	Absence of pointing, were more frequent in the AD group. The impact on patients and families were also greater in the AD group.
Hill et al.	2014	40	44	6 to 13 years			USA	Verbal Comprehension/ Verbal IQ and Processing Speed composite scores: 158.18 (22.408)	To examine the potential moderating effects of anxiety and cognitive functioning on the relation between local processing and social skills in children with ASD	Comparative study	The Wechsler Preschool and Primary Scale of Intelligence – Third Edition. The Wechsler Intelligence Scale for Children – Fourth Edition The Anxiety and Social Skills subscale scores from the Parent Report Form of the Behavioral Assessment System for Children – Second Edition	The Block Design subtest scores did not differ between children with AD and those with PDD-NOS. Anxiety and cognitive functioning moderated the association between local processing and social skills whatever the diagnosis considered

(Continued)

TABLE 2 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Functioning level	Objective	Study design	Main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Thurm et al.	2014	125	42	AD: 48.6 months (16.9) PDD-NOS: 48 months (15.7)	35	83	USA	DQ: 72.5 (19.9)	To provide novel data on timing information about both attainment and any loss of skills to capture onset patterns in young children	Comparative study	A detailed caregiver interview	In children with AD, pointing to express interest and show object were lesser frequently attained skills than in those with PDD-NOS. The percentage of children reported to have lost at least one skill revealed that the AD group had the largest percentage of children with caregiver report of any loss followed by PDD-NOS
Kose et a.	2013	46	38	AD: 8.6 years (3.8) PDD-NOS: 6.3 years (2.5)	32	84	Turkey		To investigate the Health Related Quality of Life of children within ASD groups.	Comparative study	The Pediatric Quality of Life Inventory 4.0 scored by mothers	Psychosocial, social, school functioning and total summary score of the AD group were lower than PDD-NOS
Peters-Scheffer et al.	2013	87	24	AD: 5.58 years (17.19) PDD-NOS: 5,92 years (19.88)			The Netherlands	IQ: 32.85 (14.42)	To compare behavioral flexibility in those with AD and those with PDD-NOS	Comparative study	The Behavior Flexibility Rating Scale – Revised The Mullen Scales of Early Learning	No differences in behavioral flexibility toward persons were found between children with PDD-NOS plus ID and children with AD plus ID

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TABLE 2 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Functioning level	Objective	Study design	Main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Schlooz et al.	2013	15	21	AD: 131.27 months (13.59) PDD-NOS: 130.29 (16.73)	21	100	The Netherlands	Full Scale IQ: 109.43 (12.16)	To test whether the AD group reach higher scores on the Embedded Figures Test than the PDD group	Comparative study	The Children's Embedded Figures Test The Adult Embedded Figures Test	Children and adolescents with AD and PDD-NOS did not differ on visual-perceptual tests
Visser et al.	2013	121	75	AD: 32.6 months (6.6) PDD-NOS: 34.6 months (5.8)			The Netherlands		This study compared the occurrence of pre- and perinatal risk factors between the narrowly (AD) versus broadly defined autistic phenotypes (PDD-NOS)	Comparative study	A parental questionnaires derived from the Prechtl optimality scales with addition of relevant items like paternal age, intoxications and maternal stress	Cases with PDD-NOS differed from those with AD by higher exposure to smoking during pregnancy and by a negative association of smoking with IQ. SDP appears to contribute more to broadly defined than to narrowly defined autistic phenotypes
Blijd-Hoogewys et al.	2012	35	65	11.23 years (3.35)			The Netherlands	Performance IQ: 91.98 (19.26)	To examine whether different ASD subtypes (Autistic disorder and PDDNOS) show different profiles	Comparative study	The Behavior Rating Inventory of Executive Functions	The BRIEF scale scores for AD and PDDNOS do not differ significantly on executive funtions
de Bildt et al.	2012	60	95	11.90 years (3.92)			The Netherlands	ID	To examine the rates of Visual Rooting Reflex in the AD and PDD-NOS subgroups	Comparative study	The ADOS[3] The VABS[4]	Children with AD + ID displayed a Visual Rooting Reflex more often than children with PDD-NOS + ID

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TABLE 2 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Functioning level	Objective	Study design	Main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Kjellmer et al.	2012	22	44	69 months (8.5)			Sweden	Full Scale IQ: 86.3 (11.3)	To compare language comprehension between children with AD and those with PDD-NOS	Comparative study	The Comprehension Scale of the Reynell Developmental Language Scales III	Children with AD and PDD-NOS did not differ in language comprehension
Mandy et al.	2011	97	66	AD: 9.43 years (3.50)[2] PDD-NOS: 9.05 years (3.51)	52	79	UK	Verbal IQ: 87.60 (15.98)	To characterize a group of individuals diagnosed according to a clearly operationalized DSM-IV-TR definition of PDD-NOS	Comparative study	The Developmental, Dimensional and Diagnostic Interview The ADOS	For gross motor and fine motor impairment there were no group differences. By contrast, on measures of visuo-spatial impairment and auditory sensitivity the PDD-NOS group were less impaired that the AD group
O'Donnell et al.	2012	28	14	36 to 59 months			USA		To explore the sensory processing differences between two subgroups of these children (AD and PDD-NOS)	Comparative study	The Short Sensory Profile	Short Sensory Profile scores between the two subgroups of children (AD and PDD–NOS) did not significantly differed

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TABLE 2 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Functioning level	Objective	Study design	Main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Beeger et al.	2010	11	20	AD: 9,33 years (1.67) PDD-NOS: years 9,17 (1.75)			The Netherlands	Full Scale IQ: 96.1 (11.1)	To test whether children with AD and PDD-NOS differ in their tendency to account for the impact of an emotionally charged initial situation in their emotional reaction to a successive situation (emotional transfer).	Comparative study	Two parallel sets of four stories	Children with AD reported equal effects of preceding positive and negative emotions, and children with PDD-NOS were relatively unaffected by the preceding emotions
Huemer et al.	2010	171	119	AD: 10.41 years PDD-NOS: 10.08 years	91	76	USA		To compare decoding and comprehension in children with AD and those with PDD-NOS	Comparative study	The Woodcock reading mastery test—revised Slosson oral reading test-revised Gray oral reading test-revised, 4th edition Lindamood auditory conceptualization test The Peabody picture vocabulary test third edition Detroit tests of learning aptitude-4th edition, word opposites Detroit tests of learning aptitude-2nd edition, Oral directions	Group differences on decoding and comprehension between the AD and the PDD-NOS group were not significant

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TABLE 2 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Functioning level	Objective	Study design	Main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Matson, Mahan, Fodstad et al.	2010	117	112	AD: 26.83 months (4.78) PDD-NOS 25.76 months (4.48)	80	71	USA		To analyze the differences between motor skills in infants and toddlers with various AD and PDD-NOS	Comparative study	The Battelle Developmental Inventory, 2nd Edition	Toddlers with PDDNOS did not have significantly different fine or gross motor skills than toddlers with AD
Matson, Mahan, Kozlowski et al.	2010	165	166	AD: 27.52 months (4.61) PDD-NOS: 26.64 months (4.69)	123	74	USA		To examine whether there are differences in age of onset for developmental milestones, such as saying one's first word, first phrase, onset of crawling and onset of walking, between the AD and the PDD-NOS group	Comparative study	The BISCUIT[5] demographic form	The children with AD began crawling significantly later than those with PDD-NOS. No other comparison was significant
Zander et al.	2010	85	94	AD: 10.22 years (2.16), PDD-NOS: 10.73 years (2.15)	51	54%	Sweden		To examine index score profiles and their characteristics in a large Swedish sample consisting of children with AD and PDD-NOS	Comparative study	The Swedish version of the Weschler Intelligence Scale for Children – third edition	There were no significant differences between children with AD and those with PDD-NOS on any index score of the Weschler Intelligence Scale for Children
Dawson et al.	2009	314	84	4 to 19 years			Australia		To investigate the association between birth defects and ASD for AD and PDD-NOS	Comparative study		Odds ratios for birth defects were similar for AD and PDD-NOS

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TABLE 2 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Functioning level	Objective	Study design	Main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Meilleur et al.	2009	80	44	6.3 years (4.1)			Canada		To compare regressive and non-regressive ASD children for their profile of diagnosis	Comparative study	The ADI-R	Children with AD were significantly more likely to regress in skills other than language when compared with children with PDD-NOS
Perry et al.	2009	192	66	51.70 months (12.51)			Canada		To examine whether the adaptive behavior of children with AD differ from individually matched children with PDD-NOS	Comparative study	The VABS	No differences were found between matched pairs of children with AD and PDD-NOS on adaptive behavior
Portoghese et al.	2009	21	25	AD:3.2 years (1.04) PDD-NOS: 3.3 years (0.92)	25	92	Italy		To discern differences between preschool children with AD and children with PDD-NOS as regards their developmental and behavioral levels	Comparative study	The Revised Psychoeducational Profile	All behavioral areas and almost all developmental domains (with the exception of the cognitive verbal) assessed were more severely impaired in the AD group.
de Bruin et al.	2006	13	76	AD: 8.62 years (1.81) PDD-NOS: 8.39 years (1.86)	65	86	The Netherlands	Full scale IQ: 89.58 (19.38)	To compare VIQ–PIQ and subtest patterns in children with AD from children with PDD-NOS	Comparative study	The WICS-R	No overall differences between VIQ and PIQ were found in PDD-NOS and autism.

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TABLE 2 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Functioning level	Objective	Study design	Main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Verté et al.	2006	50	25	AD: 8.7 years (1.9) PDD-NOS: 8.5 years (1.4)	20	80	The Netherlands	Full scale IQ: 98.3 (14.4)	To investigate whether children with AD (without) and PDDNOS can be differentiated from each other on their neurocognitive executive functioning (EF) profile	Comparative study	The change task The circle drawing task Test of Everyday Attention for Children, Subtest Opposite Worlds The Self-Ordered Pointing Task, Abstract Designs The tower of London The Wisconsin Card Sorting Test An adaptation of the Controlled Word Association Task	The executive function profile of the PDDNOS group was less disturbed than the profile of the AD group (higher Visual Working Memory and Planning scores).
Glason et al.	2004	314	84	4 to 19 years			Australia		To examine the association of obstetric factors with autism spectrum disorders for a cohort of children, using obstetric data contained in a statutory database collected at the time of birth	Comparative study		The PDD-NOS group had similar types of complications to the autism group, but some variables reached statistical significance. These variables were greater frequencies of threatened abortion, a labor duration of less than 1 h. Cases were more likely to have experienced cephalopelvic disproportion, been delivered by an elective or emergency cesarean section.

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TABLE 2 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Functioning level	Objective	Study design	Main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Rhea et al.	2004	20	20	AD: 6.5 years (1.8) PDD-NOS 6.2 years (1.6)	18	90	USA	IQ: 76.3 (16)	To provide a microanalysis of differences in adaptive functioning seen between well-matched groups of school-aged children with autism and those diagnosed as having PDD-NOS	Comparative study	The VABS	Findings indicate that children with PDD-NOS scored higher in expressive communication; specifically, the use of elaborations in syntax and morphology and in pragmatic use of language to convey and to seek information in discourse
Walker et al.	2004	216	21	AD: 86.3 months (38.0) PDD-NOS: 99.0 months (71.8)	18	86	USA	Leiter IQ: 82.38 (33.40)	To describe the characteristics of children with PDD-NOS, diagnosed using a consensus best estimate	Comparative study	The VABS The Leiter International Performance Scale	Children with PDD-NOS have higher functioning levels than children with AD.
Allen et al.	2001	176	18	AD without ID: 57.8 months (15.2) AD with ID: 59.6 months (16.4) PDD-NOS without ID: 56.0 months (13.3) PDD-NOS with ID: 66.0 months (15.15)	14	78	USA	ID in 90% of cases	To determine whether preschool children having AD differed in kind from those of with PDD-NOS	Comparative study	The VABS The Wing Autistic Disorder Interview Checklist The Stanford-Binet 4th edition	PDD-NOS children did not differ significantly from the AD children in verbal and adaptive skills. The PDD-NOS did also not differ from the AD children in maladaptive behaviors.

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TABLE 2 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Functioning level	Objective	Study design	Main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Juul-Dam et al.	2001	61	16	5 years			USA		To examine various pre-, peri-, and neonatal factors	Comparative study	Risk factors assessment	The PDD-NOS group showed results similar to those of the AD group, with significantly lower increased incidence of hyperbilirubinemia
Buitelaar et al.	1999	20	20	AD: 12.5 years (3.2) PDD-NOS: 12.4 years (3.1)	17	85	The Netherlands	Full Scale IQ: 98.7 (18.3)	To explore wether autistic children without ID and autistic-like (PDD-NOS) children could be distinguished in social cognitive performance	Comparative study	First order ToM tasks Second order ToM tasks Emotion recognition	AD and PDD-NOS children could not be significantly differentiated from each other on social cognitive performance
Ghaziuddin et al.	1998	12	12	AD: 10.3 years (2.9) PDD-NOS: 10.1 years (2.7)	10	83	USA	Full IQ: 78.2 (14.5)	To assess the presence of clumsiness in patients with AD or PDDNOS	Comparative study	The Bruininks Oseretsky test	Patients with PDD-NOS were found to be less impaired on battery test scores than those with AD
Volkmar et al.	1993	199	74	AD: 13.29 years (7.64) PDD-NOS: 7.47 (6.05)	58	78	USA	IQ:70.23 (22.46)	To explore the nature of sex differences in AD and PDD-NOS	Comparative study	The ABC The VAB	The AD males were 8.8 times more likely than females to have full-scale IQ scores >70; whereas in the PDD-NOS group males were only 1.5 times more likely to have full-scale IQs > 70

[1](SD). [2]Clinical Global Impression Severity Scale. [3]Autism Diagnostic Observation Schedule. [4]Vineland Adaptive Behavior Scale. [5]Baby and Infant Screen for Children with aUtIsm Traits.

TABLE 3 Associated mental health conditions in PDD-NOS in comparison with AD.

Authors	Year	Population		Age	Males in PDD-NOS group		Country	PDD-NOS Functioning level	Objective	Study Design	Methods and main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Chien et al.	2020	2,929	461	AD: 5.6 years (10.6)[1] PDD-NOS: 12.7 years (16.5)	311	68	Taiwan	Absence of ID	To investigate the incidence of comorbid Schizoprenia Spectrum Disorder, Bipolar Disorder, Major Depressive Disorder and ASD associated neurodevelopmental conditions in AD and PDD-NOS using a national insurance database	Comparative study	Claim records completed by physicians	Patients with PDD-NOS had a higher risk for developing Schizoprenia Spectrum Disorder, Bipolar Disorder, Major Depressive Disorder than AD.
Carigi et al.	2014	46	71	AD: 67.78 months (39.59) PDD-NOS: 45.20 months (17.28)	56	79	Italy	DQ[2]: 59.86 (15.42)	To identify clinical features that might prove able to differentiate AD from PDD-NOS	Comparative study	Semi-structured interview to collect information about the developmental history	Pharmacotherapy were more frequent in the AD group.

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TABLE 3 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	PDD-NOS Functioning level	Objective	Study Design	Methods and main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Joshi et al.	2014	ASD clinic: 89 Psychiatry clinic: 25	ASD clinic: 30 Psychiatry clinic: 192	3 to 17 years			USA		To investigate whether phenotypes of ASD and associated psychopathology and dysfunction varied by referral source. To this end, youth with ASD attending a specialty clinic for ASD were compared to those attending a general psychiatry clinic	Comparative study	The K-SASD[3] The DSM Global Assessment of Functioning Scale	More ASD clinic youth met criteria for AD; more youth referred to the psychiatry clinic met criteria for PDD-NOS. General psychiatry clinic youth with ASD suffered from a greater burden of psychopathologies (Oppositional defiant disorder, Major Depressive Disorder and psychosis) higher levels of dysfunction and more pharmacotherapy.
Kozlowski et al.	2012	92	114	AD: 26.57 months (4.84) PDD-NOS: 26.14 months (5.036)	57	50%	USA		To identify differences in challenging behavior endorsement rates between AD and PDD-NOS groups	Comparative study	The BISCUIT[4]-Part 3	Individuals with AD endorse significantly greater amounts of challenging behaviors (aggressive/disruptive behaviors, stereotypic behaviors, and self-injurious behaviors) than individuals with PDD-NOS

(Continued)

TABLE 3 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	PDD-NOS Functioning level	Objective	Study Design	Methods and main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Gjevik et al.	2011	47	12	11.8 years (3.3)			Norway		To assess the prevalence of current comorbid DSM-IV disorders in a special school population of children and adolescents with AD and PDD-NOS	Comparative study	The K-SADS	Obsessive-Compulsive Disorder and Conduct Disorder was more prevalent in children with PDD NOS than in those with AD.
Horovitz et al.	2011	291	241	26.33 months (4.76)	168	70	USA		to examine the differential endorsement of challenging behaviors with respect to diagnosis subtype (AD vs. PDD-NOS)	Comparative study	The BISCUIT part 3	Young children with AD are endorsed for engaging in significantly more challenging behaviors than those with PD-NOS (Destructive/ aggressive behavior, stereotypic behaviors but not self-injurious behavior)
Sipes et al.	2011	247	211	AD: 27.21 months (4.84) PDD-NOS; 26.47 months (4.84)	152	72	USA		To determine if tantrum/conduct problems were rated more frequently in toddlers with AD than in those with PDD-NOS	Comparative study	The BISCUIT-Part 2	Children with AD exhibited greater symptoms of tantrum/ conduct problems
Snow et al.	2011	54	54	AD: 65.6 months (28.7) PDD-NOS: 65.4 months (29.7)			USA	Non Verbal IQ: 78.1 (19.7)	To determine if children in these different diagnostic groups differed on rates and profiles of psychopathology	Comparative study	The ADOS[5] The Child Behavior Checklist	Higher scores in the PDD-NOS group (school-age sample) on two items measuring Anxiety/ Depression on the Child Behavior Checklist

(Continued)

TABLE 3 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	PDD-NOS Functioning level	Objective	Study Design	Methods and main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Davis et al.	2011	33	33	7.46 years (2.79)			USA	91.1% without ID	To examine whether or not there are fundamental differences in the effects of communication deficits on anxiety levels in children with AD and PDD-NOS	Comparative study	The Autism Spectrum Disorders – Comorbidity for Children The Autism Spectrum Disorders – Diagnostic for Children	The AD and PDD-NOS groups interacted with communication deficits such that children with AD experienced less anxiety as communication deficits increased while children with PDD-NOS experience more anxiety as communication deficits increased
Davis et al.	2010	159	154	27.09 months (5.02)			USA		To establish if there are differences in the prevalence of anxiety and avoidance between infants and toddlers diagnosed with AD versus those with PDD-NOS	Comparative study	The BISCUIT-Part 2	Results indicated an overall pattern whereby toddlers with AD had more severe anxious and avoidant symptoms than toddlers with PDD-NOS
Horovitz et al.	2010	141	135	26.68 months (4.97)			USA		To examine gender differences in psychiatric symptoms in infants and toddlers with AD and PDD-NOS	Comparative study	The BISCUIT part 2	No gender differences. The AD group evinced significantly more psychiatric symptoms (tantrum/conduct behavior, inattention/ impulsivity, avoidance behavior, anxiety/ repetitive behavior, eating/sleep problems) than the PDD-NOS

(Continued)

TABLE 3 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	PDD-NOS Functioning level	Objective	Study Design	Methods and main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Matson et al.	2009	169	140	27.30 months (4.81).			USA		To examine the frequency of various comorbid problems in infants and toddlers with an AD or PDD-NOS	Comparative study	The BISCUIT-Part 2	All five disorders (tantrum/conduct behavior, inattentive/impulsive, avoidant behavior, anxiety/repetitive and eating problems/sleep) were more common in the AD group. Differences in the two groups were particularly striking for the anxiety/repetitive behavior and inattention/impulsivity factors
Gurkan et al.	2008	18	11	AD: 10.61 years (3.99) PDD-NOS: 10.27 (3.50)	11	100	Turkey	Full Scale IQ: 91.00 (23.46)	To determine the pattern and frequency of psychiatric comorbidity in children and adolescents with AD and PDD-NOS	Comparative study	The K-SADS The Wechsler Intelligence Scale for children-Revised tests	Differences were significant only for tic disorders which were highest in PDD-NOS group
Pearson et al.	2006	25	25	AD: 9.5 years (3.1) PDD-NOS: 10.5 months (3.3)	19	76	USA	Full-Scale IQ: 94.0 (22.9)	To compare differences in behavioral and emotional comorbidity in children with AD and those with PDD-NOS	Comparative study	The Personality Inventory for Children – Revised	Children with PDD-NOS were at lower risk for depression, social withdrawal, atypical behaviors/psychosis, and social skills problems and higher risk for family problems

[1](SD). [2]Development Quotient. [3]Schedule for Affective Disorders and Schizophrenia for School-Age Children. [4]Baby and Infant Screen for Children with aUtIsm Traits. [5]Autism Diagnostic Observation Schedule.

TABLE 4 Associated physical health conditions in PDD-NOS in comparison with AD.

Authors	Year	Population		Age	Males in PDD-NOS group		Country	PDD-NOS Functioning level	Objective	Study design	Methods and main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Gok et al.	2019	61	41	9.56 years (3.9)[1]			Turkey		To evaluate the relationship between sleep disturbances, gastrointestinal problems and eating behaviors in children who are diagnosed with AD and PDD-NOS	Comparative study	Parental reports on sleeping and gastrointestinal problems The Feeding Assessment Survey The Brief Autism Mealtime Behavior Inventory	Sleeping, gastrointestinal and eating problems are seen in those with AD more commonly than in those diagnosed with PDD-NOS
Carigi et al.	2014	46	71	AD: 67.78 months (39.59) PDD-NOS: 45.20 months (17.28)	56	79	Italy	DQ[2]: 59.86 (15.42)	To identify clinical features that might prove able to differentiate AD from PDD-NOS	Comparative study	Semi-structured interview to collect information about the developmental history	Sleep disorders were more frequent in the AD group.
Kozlowski et al.	2012	506	502	AD: 26.63 months (4.69) PDD-NOS: 26.35 (4.82)	376	72	USA		To compare groups of children with AD and PDD-NOS on their Eating/Sleep Problems	Comparative study	the BISCUIT[3]-Part 2	Children with AD were found to present with significantly more feeding and sleep problems than children with PDD-NOS
Mandy et al.	2011	97	66	AD: 9.43 years (3.50)[2] PDD-NOS: 9.05 years (3.51)	52	79	UK	Verbal IQ: 87.60 (15.98)	To characterize a group of individuals diagnosed according to a clearly operationalized DSM-IV-TR definition of PDD-NOS	Comparative study	The Developmental, Dimensional and Diagnostic Interview The ADOS[4]	For sleep problems there were no group differences. By contrast, on measures of feeding difficulties the PDD-NOS group were less impaired that the AD group
Matson et al.	2009	72	40	8.21 years (3.76)			USA		To better understand the nature and specific interrelationship between feeding problems and autism and PDD-NOS should be	Comparative study	The Autism Spectrum Disorders-Diagnostic for Children The Autism Spectrum Disorders-Comorbidity for Children	Feeding problems did not differed between the AD and PDD-NOS groups

(Continued)

TABLE 4 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	PDD-NOS Functioning level	Objective	Study design	Methods and main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Parmeggiani et al.	2007	77	77	10 years 1 month (range: 3 years to 29 years 2 months)	48	62	Italy	ID in 92.3% of cases	To evaluate the occurrence, features, and causes of epilepsy in pervasive developmental disorder not otherwise specified in comparison with autistic disorder	Comparative study	The CARS[5]	Mild mental retardation, pathological neurological examination, cerebral lesions, abnormal EEG background activity and associated genetic pathologies were more common in PDD-NOS. Epilepsy occurred in 35.1% of patients with PDD-NOS, with no statistically significant difference compared with AD. The mean age of seizure onset was earlier (2 years 8 months) in PDD-NOS. Seizure outcome was better in AD

[1](SD) [2]Developmental Quotient. [3]Baby and Infant Screen for Children with aUtIsm Traits. [4]Autism Diagnostic Observation Schedule. [5]Childhood Autism Rating Scale.

TABLE 5 Longitudinal studies comparing developmental trajectories in AD and PDD-NOS.

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Objective	Study design	Methods	Main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Usta et al.	2019	182	210	72.3 months (45.9)[1]			Turkey	To examine predictors of outcome after 3-year clinical observation, special education and psychiatric treatment in the clinical-based group	Prospective study	The ASD symptoms were assessed at baseline (T0) and 12th (T1), 24th (T2) and 36th (T3) months. The performance of machine learning algorithms was tested on the data.	The ABC[2] The Abberant Behavior Checklist The CGI[3]	In the autism group, older father and mother age In the PDD-NOS group, mental retardation comorbidity, less birth weight and older age at diagnosis have a worse outcome
Hinnebusch et al.	2017	111	82	AD: 27.09 months (4.57) PDD-NOS: 25.88 months (4.04)	65	78	USA	To determine whether children retained an ASD diagnosis (AD or PDD-NOS) and to assess autism severity and degree of cognitive and adaptive progress	Prospective study	Children were initially evaluated at approximately 24 months of age and were then reevaluated about two years later.	The ADOS-G[4] The CARS[5] The VABS[6] The Mullen Scales of Early Learning	The AD group showed a higher rate of diagnostic stability than the PDD-NOS group. The PDD-NOS group showed significanty less improvement than the AD group; however, at both time points, they were still on average in the PDD-NOS range, with lower severity scores than the AD group. No significant differences in learning rate on cognitive or adaptive measures between the AD and the PDD-NOS groups

(Continued)

TABLE 5 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Objective	Study design	Methods	Main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Mordre et al.	2012	74	39	AD: 6.7 years (SD 2.6) PDD-NOS: 7.7 years (SD 2.3)	31	80	Norway	To compare adult outcome of individuals with PDD NOS and individuals with AD	Comparative study	Children with AD and PDD-NOS were followed up from childhood to adulthood. Outcome measures were criminality, mortality and marital status rates		The disability pension award was the only outcome measure that differed significantly between the AD and PDD-NOS group. The lower rate of disability pension award in the PDD-NOS group was predicted by better psychosocial functioning
Osada et al.	2012	67	31	AD: 8.99 years (0.39) PDD-NOS: 8.98 years (0.61)	24	77	Japan	To clarify how IQ, autistic symptoms, educational placements and job status vary between AD and PDD-NOS	Longitudinal comparative study	The participants' data were collected between their first visit to the clinic and the visit at which they applied for basic disability benefits at 20 years of age.	The Japanese version of the Stanford-Binet The CARS	Participants with AD consistently showed lower IQ and more severe autistic symptoms than those diagnosed with PDD-NOS
Malhi et al.	2011	64	13	AD: 2.49 years (SD 0.41) PDD-NOS; 2.43 years (SD 0.37)			India	To describe stability and change of early diagnosis of AD and PDD-NOS	Prospective follow up study	Diagnosis of AD and PDD-NOS was made at age 3 years or less and follow-up was done around 4 years of age.	The CARS	The diagnosis of AD was more likely to persist than the diagnosis of PDD-NOS

(Continued)

TABLE 5 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Objective	Study design	Methods	Main assessment tools	Results
		AD	PDD-NOS		N	%						
Kim et al.	2010	121	71	8 to 30 months	63	89	USA	To study longitudinal data from toddlers and pre-schoolers (from 8 to 56 months old) with AD and PDD-NOS	Longitudinal study	A hierarchical regression analysis was carried out using children's earlier RRB totals (from 8 to 30 months) as one of the predictors for the same children's later RRB totals (from 31 to 56 months)	The ADOS	The likelihood of having an restrictive and repetitive behavior was the same for both AD and PDD-NOS groups; though the rated severity of these restrictive and repetitive behaviors was higher in children over age two with AD than PDD-NOS diagnoses
Anderson et al.	2009	93	51	AD: 29.6 months (4.68) PDD-NOS: 29.45 months (5.67)			USA	To examine the development of social skills between ages 2 to 13 in a sample of children initially diagnosed with AD or PDDNOS	Prospective study	Growth curve analysis was used to examine growth in socialization age equivalents from age 3 to 13 (Assessment at 2, 3, 5, 9 and 13 years)	The ADOS The ADI-R[7] The VABS	The gap between children with AD and the PDD-NOS group widened with time as the social skills of the latter group improved at a higher rate
Chawarska et al.	2009	43	18	21.5 months (SD 4.9)			USA	To examine short-term stability of clinical diagnosis in the second year of life	Prospective study	Cognitive, social, and communication skills of toddlers were assessed at the average age of 21.5 (SD 4.9) months, and reassessed at 46.9 (SD 7.7) months	The ADOS The Mullen Scales of Early Learning	Diagnosis of AD was stable in 74% of cases as compared to 83% in PDD-NOS

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TABLE 5 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Objective	Study design	Methods	Main assessment tools	Results
		AD	PDD-NOS		*N*	%						
van Daalen et al.	2009	40	13	AD: 29.4 (5.6) PDD-NOS: 28.2 (5.2)	9	69	The Netherlands	To evaluate the stability of ASD diagnoses in children	Prospective study	The ASD diagnoses (AD and PDD-NOS) at 23 months and 42 months of 131 patients were compared to evaluate stability of the diagnosis of ASD		The stability was 63% for AD and 54% for PDD-NOS
Anderson et al.	2007	98	58	29 months (SD 5.17)			USA	To contrast the differences between diagnostic groups in the development of verbal skills from age 2 to 9	Prospective study	Growth curve analyses were used to analyze verbal skills trajectories over time	The ADOS The ADI-R The Infant Mullen Scales of Early Learning	The gap between children with AD and the PDD-NOS group widened with time as the verbal abilities of the latter group improved at a higher rate
Takeda et al.	2007	49	77	AD: 31.7 ± 3.3 months PDD-NOS: 34.8 ± 3.5 months	39	51	Japan	To compare the change in DQ or IQ between AD and PDDNOS in preschool years	Comparative study	AD and PDD-NOS children were evaluated at age 2 and at age 5	The Kyoto Scale of Psychological Development and the Japanese version	The PDD-NOS children were significantly higher in DQ/IQ at age 2 and at age 5 than the AD children

(Continued)

TABLE 5 (Continued)

Authors	Year	Population		Age	Males in PDD-NOS group		Country	Objective	Study design	Methods	Main assessment tools	Results
		AD	PDD-NOS		*N*	%						
Thurm et al.	2007	59	24	AD: 29.98 months (SD 4.28) PDD-NOS: 30.38 months (SD 4.69)	19	79	USA	To explore which early variables (e.g., non-verbal cognitive ability, Vineland social and communication skills) at ages 2 and 3, best predicted receptive and expressive language at age 5	Prospective study	The study analyses data obtained at age 2 and age 3, as well as between the ages of 4 and 5	The ADI-R The ADOS The DAS The Mullen Scales of Early Learning The VABS	VABS age equivalent ratio scores (ratio scores = age equivalent scores/ chronological age) and other key variables indicated that Children with AD had significantly lower ratio scores than children with PDD-NOS on non-verbal cognitive ability and on both the socialization and communication domains of the VABS at ages 2 and 3, as well as on the composite expressive and receptive language age equivalent ratio scores at age 5
Lord et al.	2006	84	46	AD:29.1 months (SD 4.7) PDD-NOS 29.1 months (SD 5.6)	35	76	USA	To address the question of diagnostic stability of AD and PDD-NOS	Prospective study	Prospective study of diagnostic classifications made at ages 2 and 9 years	The ADI-R The ADOS	Diagnostic stability at age 9 years was very high for AD at age 2 years and less strong for PDD-NOS
Stone et al.	1999	25	12	31;4 months (SD¯3;4)			USA	To examine the stability of the diagnosis over a 1-year period	Prospective study	This sample included children who received an ASD diagnosis before 3 years and returned for a 1-year follow-up	The ADI-R The ADOS	The diagnosis of AD was more stable over time than was the diagnosis of PDD-NOS

[1](SD). [2]Autistic Behavior Checklist. [3]Clinical Global Impression Severity Scale. [4]Autism Diagnostic Observation Schedule. [5]Child Autism Rating Scale. [6]Vineland Adaptive Behavior Scale. [7]Autism Diagnostic Interview – Revised.

restrictive and repetitive behaviors was higher in children over age 2 with AD than with children with a PDD-NOS diagnosis (106).

Regarding functional outcome, a study form Norway (107) showed that the disability pension award at adulthood was the only outcome measure differing significantly between the AD and PDD-NOS groups previously included during childhood.

Within the PDD-NOS group, worse outcome was associated with ID, lesser birth weight and older age at diagnosis (108). Moreover, a better psychosocial functioning during childhood predicted a lower rate of disability pension award in the PDD-NOS group (107).

Discussion

We conducted a systematic review of PDD-NOS in comparison with AD. Cross-sectional and longitudinal studies comparing PDD-NOS and AD showed a clear trend for lower communication and socialization impairment in children with PDD-NOS. Results on restrictive and repetitive behaviors were more contradictory. Early development was less impacted in PDD-NOS as well as the quality of life of children and families. Children with PDD-NOS showed less challenging behavior than those with AD. Schizophrenia and mood disorders appeared to be higher in PDD-NOS whereas feeding and sleep disorders tend to be less prevalent.

With regard to our results, PDD-NOS appeared as category both quantitatively and qualitatively distinct from AD. In a quantitative perspective, PDD-NOS was a lesser variant of autism with respect to the trajectory of autism symptoms, some associated mental and physical health conditions, or quality of life. This perspective is consistent with the spectrum approach of the DSM-5 (2) and a possible continuum between AD and PDD-NOS within ASD. However, in a qualitative perspective, PDD-NOS differed from AD because of higher associated schizophrenia and mood disorders. These results were unexpected with respect to a spectrum approach and suggest specific links between the margins of autism and other psychiatric dimensions. Our review coheres with Tsai and Ghaziuddin's review (109) which concluded that the literature appeared to suggest that PDDNOS can be separated from AD, and, therefore, does not seem to lie on a "continuum" with that disorder. These authors recommended systematic research to settle the issue and that PDDNOS should be regarded as a separate category to facilitate ongoing research (109). In contrast, a recent systematic review on PDD-NOS (110) concluded that in regard of the limited consistency, sensibility, and stability of this disorder, it would be more consistent to include this diagnosis within ASD. Noticeably, the authors still recommended to collect information from professionals on adults who received a PDD-NOS diagnosis in childhood.

Our search on PubMed found 510 references. On the same period, from 1987 to 2022, the term "Autistic disorder" gave 24,639 references. PDD-NOS was thus referenced 48 times less than AD, and was therefore much less studied. However, the high prevalence of the PDD-NOS category demonstrates its high clinical relevance. This clinical importance of PDD-NOS in psychiatry was also emphasized by a study comparing youth with PDDs attending a specialty clinic for autism with those attending a general psychiatry clinic (88). More autism clinic youth met criteria for AD; more of those referred to the psychiatry clinic met the criteria for PDD-NOS (88). The discrepancy between the past clinical use of the PDD-NOS category and the scarcity of research in the field (in comparison with AD) might be due to the difficulty of studying the margins of autism because of their heterogeneity and complexity - which our review aimed to underline. PDD-NOS therefore illustrated a gap between clinical and research issues and the need to address heterogeneity from a research perspective. Based on research rather than clinical consideration of the PDD-NOS, the DSM-5 missed the complexity of the margins and opted for a continuum approach and ASD.

With regard to the 4 recommendations proposed by Towbin on the use of the PDD-NOS diagnosis category (21), it appears that, firstly, although PDD-NOS is related to earlier developmental disorders, the age of parental concern does not differ between AD and PDD-NOS. The latter result suggests that the PDD-NOS is not commonly used in the case of a later age of onset of autistic symptoms. Towbin (21) and Mandy et al. (35) suggested that PDD-NOS depicted a clinical picture with early onset of symptoms, an impairment in social reciprocity and less to no restrictive and repetitive behaviors. We showed, however, the lack of clear support to the claim of lesser frequency of restrictive and repetitive behaviors in PDD-NOS. Towbin also recommended using PDD-NOS as a temporary diagnosis when data are lacking or when the child is too young (21). This recommendation concurs with less diagnostic stability in PDD-NOS in infants and pre-schoolers, as is shown by our results. Finally, our review mostly supports the recommendation referring to PDDS-NOS as a lesser variant of autism with respect to the trajectory of autism symptoms, associated mental and physical health conditions, or quality of life.

Chien et al. showed that PDD-NOS was more associated than AD with schizophrenia and mood disorder (92). This result concurs with Joshi et al.'s study comparing youth with PDDs attending a specialty clinic for autism to those attending a general psychiatry clinic (88). General psychiatry clinic youth with PDDs (with more PDD-NOS than AD) suffered from more Major Depressive Disorder and psychosis. This result echoes the results of the Sporn et al.'s retrospective study, showing that 25% of the 76 patients who reported early onset schizophrenia had a diagnosis of PDDs during their childhood, with a clear predominance of PDD-NOS (111). In a reference sample of childhood onset schizophrenia, Rapaport et al. also found a high frequency of PDD-NOS (112).

The importance of associated psychosis and mood disorder in PDDs also echoes past syndromic conceptualization such as Multiple Complex Developmental Disorder (MCDD) (113), Pathological Demand Avoidance (PDA) (114) or Multi-Dimensional Impairments (MDI) (115) which linked social impairment, emotional lability and signs of psychosis (without a diagnosis of early onset schizophrenia). These signs of psychosis referred to thought disorder (MCDD), delusions (MCDD), poor ability to distinguish fantasy from reality (MDI, PDA). It is important to stress that MCDD and PDA were initially coined to deal with heterogeneity within PDD-NOS. However, both syndromes were not limited to the PDD-NOS category (116, 117).

The results of our review highlight that PDD-NOS was of great interest because of its high prevalence and clinical relevance. This diagnosis was useful to take into account milder forms of autism. However, it was less reliable and less stable through time than AD. Moreover, a qualitative study emphasized (118) that the autistic community might not acknowledge this diagnosis. This study was a phenomenological analysis of discussion forum dialog among 76 adult participants with self-reported diagnosis of AD and AS but not PDD-NOS (118). Participants constantly agreed that PDD-NOS should be removed in the DSM-5. Several participants expressed concern that the PDD-NOS diagnosis was not specific and reliable enough. They called the diagnosis "a junk category," "a receptacle" (118).

Our review emphasized lower socio-communication impairments in PDD-NOS than in AD. In DSM-5, ASD diagnosis required three criteria out of three in this domain, namely deficits in social–emotional reciprocity, deficits in nonverbal communicative behaviors used for social interaction and deficits in developing, maintaining, and understanding relationship (2). To diagnose PDD-NOS, only two of them was required (18). It might therefore be suspected that the social and communication deficit characteristic of PDD-NOS wasn't important enough for it to remain in the autism spectrum. A review and meta-analysis (119) showed that 70% of PDD-NOS did not meet the criteria for ASD. Another hypothesis postulated that PDD-NOS might belong to the Social Communication Disorder (SCD) diagnosis category. However, this category must meet all three criteria for the social-communication domain, as in ASD (2). Thus, it appears that PDD-NOS has been excluded from ASD and SCD.

Our review challenged initial views of PDD-NOS and showed that this diagnosis mostly included milder forms of autism. PDD-NOS was very clinically relevant to deal with the margins of autism and the diversity within the spectrum. However, PDD-NOS encountered too many limitations (low reliability, instability through time, low acceptability) to be retained in the DSM-5. The fact is that many PDD-NOS did not join either ASD or SCD. Thus, in order to take into account past research on PDD-NOS and the margin of autism, we argue for the creation of a new category alongside ASD and SCD, based on three main dimensions related to socialization impairment, emotional lability and psychotic symptoms and coined on PDA, MCDD and MDI. Criteria for this new category could be selected from the most valid criteria for these three syndromes and tested against the ASD criteria, to ensure that they are truly distinct. In our view, this new category would provide an opportunity for mental health professionals, along with SCD and ASD, to better take into account the neurodevelopmental disorders that have an impact on the development of social communication and its heterogeneity. From a clinical point of view, this new category would trigger the search for associated psychotic symptoms or emotional lability that might currently be understudied by professionals. In addition, new developmental trajectories, specific interventions and treatments could be identified, which would in turn make it possible to better inform and help the people concerned and their families.

Limitations

The first limitation of our study comes from the terms selected for the database search. Few studies directly addressed differences between PDD-NOS and AD. However, when the DSM-IV-TR was in use, some studies included participants with PDDs including those with AD, PDD-NOS or another PDD subtype often compared to controls. In the latter case, PDD-NOS was only mentioned in the abstract or in the title when there was a difference within PDDs subgroups. That is why we may have over-selected references showing a difference between PDD-NOS and AD, and under-selected negative studies that did not. The second limitation is that we studied all clinical features of PDD-NOS. For example, some characteristics such as quality of life were rarely studied and were only related to two references. The third limitation is sample overlap between studies, as 25% of the studies were from Matson et al. Finally, our results cannot be extended to females with PDD-NOS, individuals with PDD-NOS and ID, or adults with PDD-NOS.

Conclusion

Our systematic review shows that PDD-NOS corresponds to milder form of autism with less impact and less associated disorder, with the exception of schizophrenia and mood disorder. Our review challenges initial views of PDD-NOS, echoes past syndromic conceptualizations such as MCDD, PDA or MDI, and argues for a quantitative and qualitative distinction between AD and PDD-NOS. The PDD-NOS diagnosis was very clinically relevant to deal with the margins of autism and the diversity within the spectrum, and therefore massively used by professionals in the field. However, PDD-NOS had many limitations (low reliability, instability through time, low acceptability) and was therefore understudied, thereby generating a discrepancy between clinical and research issues. Thus, in order to overcome this gap and to take into account past research on PDD-NOS, we suggest taxonomic changes in DSM-5, through the introduction of a new category based on three main dimensions: socialization impairment, emotional lability and psychotic symptoms. Future studies are therefore needed to test relevant criteria for this new category and possibly identify developmental trajectories, specific interventions and treatments.

Author contributions

RC and OP designed the systematic review and selected the studies following PRISMA guidelines. OP was contributed mainly in epistemological and methodological. RC wrote the first complete draft of the manuscript. OP, NC, and CS provided substantial modification to the manuscript. All authors contributed to the article and approved the submitted version.

Conflict of interest

Publisher's note

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Current psychopathology models emphasize very early intersubjectivity-based interventions in children to prevent later mental disorders

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Current psychopathology models have evolved toward dimensional models, in which symptoms and diseases are at the extremes of dimensions. Despite these new dimensional proposals, classifications and third-person approach have shown limitations. Their extraordinary evolution nevertheless underlines the contributions of developmental and psychodynamic frameworks. Developmental contributions have made it possible to evolve from disorders centered on a first-person perspective. Complementarily to the first-person/third-person perspectives, we advocate a second-person perspective, based on intersubjectivity. This perspective reverses the intuitive trend to focus our interventions on the most specific symptoms and syndromes, and advocates instead interventions on a "p" general factor that are both generalized and highly targeted. The implications are (1) to intervene as early as possible, (2) to base the definition of our therapeutic targets on an intersubjective perspective, (3) to identify and enhance children's and parents' strengths. These empirically informed directions are not in the current mainstream of psychopathology frameworks, and need to be developed.

KEYWORDS

psychopathology, development, interventions, classifications, P factor, phenomenology, psychodynamic, intersubjectivity

Psychopathology models have evolved toward dimensional models, in which symptoms and diseases are at the extremes of dimensions

Developmental disorders force us to revise our models of psychopathology, which have been essentially built on those of adult psychiatry. The etymological conception of psychopathology, which refers to "psyche/pathos/logy," aims at the study of pathological manifestations of the human psyche. Psychopathology remains first and foremost the study of pathological human behavior. Its object is the human psyche, i.e., a complex epistemic object whose ontological referent belongs to several fields. We have to combine two approaches: the "nomothetic" one, which aims to determine universal laws, and the "idiographic" one, which grasps the particular (Lyon et al., 2017). Different functions have been proposed for psychopathology: the descriptive function deals with the phenomenological experience of psychopathological suffering, the clinical one organizes the classification of this experience, while the theoretical one explores the etiology of this experience. In the APA dictionary of psychology,[1] however, the term "Psychopathology" is defined more narrowly as (1) the scientific study of mental disorders (2) the behavioral or cognitive manifestations of such disorders. The term in this sense is sometimes considered synonymous with mental disorder itself. Yet, there are three ways to assess mental illness (Fuchs, 2010): the positivistic, or 3rd-person approach, using classifications; the phenomenological, subject-oriented or 1st-person approach; and the hermeneutic, intersubjective or 2nd-person approach. We will first develop the interest and limits of the first approach. Although alternatives to categorical diagnoses have been proposed, they do not provide access to a person's experience. We will see that the question of development has paved the way for the second approach. We will finally show how, complementarily, the first two approaches support the absolute necessity of the third one. The implications for early intersubjective interventions will be developed.

Despite new dimensional proposals, classifications, and third-person approaches have limitations

ICD and DSM classifications are unavoidable tools for scientific communication, but they have limitations. They do not reflect the continuous process of psychopathology, but rather its final categorical manifestation (Markon and Krueger, 2005). The diagnostic categories have sometimes been built top-down, and have changed according to theoretical *a priori*, or to the influence of lobbies in order to allow insurance companies to refund the cost of treatments. They do not take into account the developmental continuity between disorders nor the frequent comorbidities (Caspi et al., 2020). Subthreshold symptoms in individuals with other disorders are not taken into account. Lastly, clinicians are encouraged to diagnose several comorbidities, using the "not otherwise specified" label, in order to facilitate access to treatment (Pincus et al., 2004).

New proposals have revisited the classification into discrete disorders. The first dimensional categorization was proposed in children by Achenbach (1966) and Achenbach and Edelbrock (1978), focusing on patterns of co-occurrence among common psychopathological syndromes in childhood, distinguishing internalizing and externalizing dimensions.

The Research Domain Criteria (RDoC) framework (Insel et al., 2010) was created as an alternative to diagnosis, and is devoted to informing future classifications, and to understanding the transdiagnostic biobehavioral systems underlying psychopathology. The RDoC framework is operationalized in the RDoC matrix, built with eight columns representing units of analysis (genes, molecules, cells, circuits, physiology, behavior, paradigms, and self-reports), and six rows representing functional higher-level domains: Negative Valence Systems, Positive Valence Systems, Cognitive Systems, Systems for Social Processes, Arousal/Regulatory Systems, and Sensorimotor Systems, each domain containing 3 to 6 constructs. All these dimensions were defined "top-down," based on expert consensus from relevant fields.

The Hierarchical Taxonomy of Psychopathology (HiTOP) alternative (Kotov et al., 2017) is a dimensional classification system, based on observational clinical assessments. It offers a hierarchical organization, from general functioning, to spectra, subfactors and symptoms, which distinguishes traits and syndromes. A general factor is at the top of the system, then increasingly detailed dimensions (internalized, externalized…), conceptualized as extremes of normal psychological functions rather than as categories, represent a continuous spectrum of risk and severity. Caspi and colleagues (Caspi et al., 2014) proposed that common categorical diagnoses of adult psychopathology could be best explained and structured by a general psychopathology latent factor alongside unique internalizing and externalizing latent factors. Covariation among symptoms of psychopathology and maladaptive traits define clusters. The treatment decision-making of clinicians is more aligned with the HiTOP description than with traditional diagnoses (Hopwood et al., 2020). In view of the respective limitations of these two models, it has been proposed that RDoC and HiTOP should be complementary models: RDoC may help elucidate the underpinnings of the clinical dimensions included in HiTOP, while HiTOP may provide psychometrically robust clinical targets for RDoC-informed research (Michelini et al., 2021).

Another alternative, bottom-up built, considers that psychiatric disorders can best be viewed as sets of symptoms that are connected through a system of causal relation. "Symptoms of psychopathology are causally connected through myriads of biological, psychological and societal mechanisms [...] The network theory holds that this is a general feature of mental disorders, which can therefore be understood as alternative stable states of strongly connected symptom networks" (Borsboom, 2017). This new alternative (Borsboom and Cramer, 2013) has led to a new way of conceiving of psychopathology and of answering the question: "What kind of things are psychiatric disorders?" (Kendler et al., 2011). These models do not consider that kinds exist whether or not we recognize them; they "are defined not in terms of essences but in terms of complex, mutually reinforcing networks of causal mechanisms" (Kendler et al., 2011). These Network-based methods are becoming increasingly widespread (Barabási et al., 2011). In the end, however, while we need models to describe the biobehavioral systems underpinning psychiatric disorders, none can avoid the question of the descriptive theoretical context of the basic symptom, which is theoretically and conceptually influenced.

The evolution of adult models of psychopathology thanks to developmental contributions

Most of the models of psychopathology described above make little reference to development and developmental trajectories. Yet, the main preoccupation of clinicians is to understand when (and why) a psychopathological trajectory begins, and how we can prevent it. Development is a trajectory, and atypical development is a trajectory that results from atypical constraints over time which may have cascading effects on how other skills are acquired (Thomas and Baughman, 2014). To capture atypical profiles, we need developmental and dynamic assessments that draw "trajectories." Yet, the above-mentioned models seldom refer to time-scales. Measuring trajectories

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[1] https://dictionary.apa.org/psychopathology

requires the repetition of assessment points in each individual, which is time-consuming and costly. By contrast, methods such as cross-sectional studies that study the differences by comparing assessments at certain points in development detect large-scale associations but do not inform on the idiographic dimension of individual trajectories.

This question concerns not only the appearance of disorders but also the organization of personality traits. Are they precursors of disorders, and if so, how does the interaction between them evolve during development? An interesting review (Durbin and Hicks, 2014) questions the theoretical background of relationships between traits or personality, and disorders. "Trait-disorder associations are dynamic in that their mechanisms differ across persons depending upon their developmental contexts, and within person, based on the idiographic histories of their traits and experience with disorder" (Durbin and Hicks, 2014). Periods of developmental tasks or transitions, such as the transition from adolescence to young adulthood, are especially at risk for these changes. These questions thus require a developmental framework such as that proposed by developmental psychopathology, which assumes that deviations from normal development are likely to signal psychopathological conditions (Cicchetti, 1993). These deviations in trajectories are particularly relevant as first manifestations of a disorder, and as targets for interventions.

Developmental considerations make it possible to evolve from a disorder-centered to a first-person perspective

How do we determine the aim of our therapeutic interventions? If we follow our two assumptions, that models are now more dimensional, and that they are developmentally informed, we need to determine the following: what is the focus of our interventions? Does the intervention concern a disorder? Or a trait, before the appearance of a disorder? An individual suffering? How can the limit between normal and pathological functioning be identified if all the dimensions assessed are continuous? A disorder arises when it causes the subject problems. Sometimes, however, the subject does not complain, or is not able to complain, for instance due to the person's developmental stage, as in infants.

All the psychopathological models we have presented are disorder-centered, ranging from normality to pathology. None of them is person-centered, even if network models make it possible to describe individual clusters. None of them refer to phenomenology, only at best to the subject's inner experience. The terms of phenomenology and experience tend to surreptitiously disappear from the field of psychopathology. The interest in phenomenology, which has progressively lost ground, was recently underlined in psychotherapy practice (Stanghellini and Cravaro, 2014). Our aim is not to develop the concept of phenomenology, but to see how developmental and diachronic frameworks have re-actualized this first-person approach.

De Ajuriaguerra (1989), introducing the developmental approach, argued that the constraint/freedom relationship changes diachronically from birth to adulthood and synchronically during the developmental stages. The constraint/freedom dualism means that the biological equipment constrains the function, but that the functioning of the function, which broadens the degree of freedom, depends more on the way the subject self-organizes the functioning of his/her biological skills. A different way to consider psychopathology would be to take into account both functions (determined by genetic, neurophysiological, and cognitive equipment, occurring in a particular environment at a particular moment), and the functioning of these functions (determined by psychological dimensions, in a social and cultural environment). This "complementarist" approach (Devereux, 1972) would make it possible to avoid opposing the various interpretations readings and interventions. While the HiTOP and RDoC models are interesting in that they distinguish these different levels, neither of them takes into account the way the subject copes with a dysfunctional function. The subject determines how he/she makes the function operate, while the clinician's role is to determine to what extent and how the function is impaired, but also how the dysfunctioning impairs, or not, the interactive and inner world of the subject. We refer to Ey's "organodynamism" (Ey, 2006), which "substitutes for monism or dualism the idea of a living dialectic between the vital infrastructure and the psychic superstructure of the person." It dialectically combines the negativity of the psychopathological process (the pathogenic organic action) with the positivity of the symptoms (the psychic reaction to this action). Therefore, the psychopathological process can hinder being-in-the-world by affecting the synchronic field (the lived experience), as well as the diachronic field of the person (by affecting the progressive integration of these experiences). Mental illness could thus be conceptualized as the knitting together of a vulnerable self, even due to genetical or developmental reasons, and the way a person tries to cope with. Narratization, rather than semi-structured interviews, allows patients to communicate and explain their own experiences, in their own terms, in the context of their personal world and history, and to try to make sense of them, through the method of 'phenomenological dissection' (Stanghellini and Cravaro, 2014). This involves a shift away from disease-and-variable-oriented strategies, toward person oriented research and treatment strategies (Bergman and Magnusson, 1997). The person-centered perspective, along with network models, emphasizes the role of multifinality (a given factor may result in a variety of outcomes) and equifinality (there are many pathways toward one specific outcome) (Cicchetti and Rogosch, 1996). The psychodynamically based classification PDM (Lingiardi and McWilliams, 2017) aims to bridge the gap between the need for experimental and methodological validity, and clinical complexity. It attempts to "characterize an individual's full range of functioning – the depth as well as the surface of emotional, cognitive and social patterns" (Lingiardi and McWilliams, 2017). Nevertheless, this manual, despite its assessment of mental functions in the M axis, remains a categorical classification.

Towards a second person perspective in interventions, implying targeting a non-specific p factor

The extraordinary shift from a categorial conception of psychopathology toward dimensional and developmental ones completely transforms our field. We propose a hypothesis: the second-person perspective, established through the interaction between a clinician and the patient, offers the most comprehensive understanding of psychopathological processes, the consequent effects of the primary dysfunction, and how patients cope with it. Primarily, this perspective facilitates the development of targeted interventions. But these three perspectives are complementary: the second-person perspective is rooted in the first-person perspective (requiring access to the patient's subjective experience) and should be interconnected with the third-person perspective (to better delineate the patient's symptoms and the specific psychiatric treatments needed). The framework of psychopathology should be not monadic unity, but rather dyadic unity: child–parent, child-therapist, parent-therapist, patient-therapist interaction, environment-patient interaction.

But we must go further. We have to reappraise psychotherapy as a highly and precisely targeted intersubjective action. There is a discrepancy between the quantity of knowledge we possess about psychiatric disorders, and the interventions that are effective. A task force (Wampold and Imel, 2015) concluded that "adapting psychological treatment (or responsiveness) to transdiagnostic client characteristics contributes to successful outcomes at least as much as, and probably more than, adapting treatment to the client's diagnosis." What works are "transtheoretical common factors" of psychotherapy, that should reduce specific symptoms through their impact on the general factor of psychopathology, and not theoretical ones (Norcross and Wampold, 2018). This conclusion could seem disappointing, but is in fact very important. A very interesting proposal was made by Forbes and colleagues (Forbes et al., 2019). "If all forms of common psychopathology are connected with a general underlying factor that can be observed from the very earliest years of development, then understanding the psychological nature of that general factor […] may provide new directions in contemplating how to reduce levels of the general factor and subsequently prevent a wide range of mental disorders from emerging later in development." In infants and children, specific psychopathological symptoms are less common: infants often express undifferentiated behaviors not yet organized as patterns. Over developmental time, attractors represent recurrent patterns that have stabilized and are increasingly predictable (Granic and Hollenstein, 2003). We have to intervene before these patterns are installed, and thus target non-specific behaviors. Intervening at the very early roots of developmental tasks in a dimensional perspective, and focusing on this general factor could prevent many later pathological traits and specific symptoms in children and adolescents. Forbes et al. (2019) go further and propose "[using] early intervention for general psychopathology as a foundational scaffold on which to introduce gradually more focused interventions later in development." This view breaks the dichotomy between prevention, which usually focuses on reducing the first signs or stages of psychopathology, and interventions, which target psychopathological patterns or symptoms. In an attempt to reach this early and non-specific p factor, Fonagy and Allison (2014) proposed that the "p factor may be a proxy for impairments in epistemic trust," that is, trust in the authenticity and personal relevance of interpersonally transmitted knowledge. For Fonagy and Campbell (2017), psychopathology might be characterized by a temporary or permanent disruption of epistemic trust and the social learning process that it enables. This may have major consequences for future interventions: the p factor could be a "reachability" factor of the patient, and the interventions should be interpersonal or intersubjective ones. The core principles of intersubjectivity, emphasizing the importance of understanding subjective experiences and interactions, are not limited by age. Thus, intersubjective-based interventions can be applicable across various age groups, including youths and adults.

When to intervene? Critical periods support early interventions

When to intervene? The early beginning of trajectories implies targeting our interventions as early as possible to prevent later psychopathology. The role of the clinician could be to identify targets and intervene at different levels simultaneously: at the higher level of general functioning, targeting the roots of multiple dysfunctions, but also at lower levels, aiming to reach a specific symptom or behavior, with a specialized intervention. These two types of intervention become complementary rather than competing.

But when does the "early" time for intervention begin? While there is a legitimate focus on determining the emerging signs of developmental disorders as soon as possible, how can a disorder be determined before the scheduled appearance of the function that is assumed to be impaired? Is it possible to determine an optimal moment to undertake interventions? The notion of sensitive/critical periods gives some insights into the early processes that underline pattern construction.

A critical period (CP) refers to a finite period in which experience provides information that is essential for normal development and alters performance permanently (Knudsen, 2004). A sensitive period refers to a period in which the effect of experience on the brain is particularly strong during a limited period in development. If experiences essential to cortical specialization do not occur during the CP, the functioning of the cortical areas allocated to the particular skill will be altered, without residual plasticity; if they do not occur during the sensitive periods, it may be difficult to redirect development along a typical trajectory; plasticity exists but to a limited degree. These two concepts refer to two mechanisms. Experience-expectant mechanisms (Greenough et al., 1987) facilitate the biological encoding of expectable environmental stimuli during constrained developmental windows, whereas experience-dependent stimuli are idiosyncratic processes that facilitate learning across the lifespan without ontogenetic constraints.

What we know better now (Reh et al., 2020) is that: (1) these windows occur for distinct domains and at different times over development; (2) the expected experience must coincide with the critical period for each circuit to occur; (3) plasticity is regulated at multiple timescales during development. The latter include different time-scaled processes: "(1) rapid, moment-by-moment shifts in circuit physiology; (2) gradual molecular events controlling the maturation of cortical circuits dictating critical period onset and closure in early life; and (3) epigenetic modifications over the life span (or across generations) that set the baseline level of plasticity" (Reh et al., 2020). Early environmental influences thus involve a complex development chronodependency of the child that opens up possibilities, if these CPs are given sufficient stimulation. Moreover, CPs are regulatory processes: they reduce future vulnerability to adversity, as experiences occurring after critical periods impact less brain circuitry (Takesian and Hensch, 2013). On the other hand, early adversity may modify CP processes, including their time of onset and closure.

This knowledge about CPs may help us to orient our intervention schedule (Nelson and Gabard-Durnam, 2020), especially in children experiencing social adversity (Nelson et al., 2019), or high-risk parenting (Feldman, 2015).

Identifying children's and parents' strengths, and not only problems

Another assumption is that when we consider the target of intervention on a continuum from normal to pathological, we identify not only problems, but also individual strengths. This view is totally underestimated and underused in psychopathology. We are trained to detect what does NOT function, and not what the strengths are that we can lean on. A recent shift has therefore appeared in interventions targeting neurodevelopmental disorders. We have shifted from purely behavioral, early, intensive interventions, in a simplified environment, which aimed to decrease deviant behavior, to naturalistic developmental behavioral interventions, community based, peer-or parent-mediated (Schreibman et al., 2015), leaning on children's and parents' skills. These new interventions aim to address all the fields of normal development (cognition, motor development, language...), in order to improve the child's engagement and reduce the vicious circle of unattuned interaction. In these interventions, the targets are normal developmental skills, and not pathological ones. Video feedback interventions with autistic children such as Preschool Autism Communication Therapy (PACT, Green et al., 2010), Video Feedback Intervention to promote Positive Parenting (VIPP, Poslawsky et al., 2014), and Interactive Guidance Therapy (IGT, Rusconi-Serpa et al., 2009) do not aim to teach new skills to the parents or to the child. They are based on the parent's identification, in a co-construction with the therapist, of the respective interactive preserved skills along with the child during successful moments of interaction in short videotaped interactive play between parent and child. The conditions and occurrence of these moments are then generalized, decreasing the vicious negative interactive circle between the two partners. We do not work FOR the patient or the parent, but WITH them. We chose IGT (Ouss et al., 2023) instead of PACT, because it does not intervene at a specific symptom level, but at the intersubjective level. We determine in the here and now the tailored focus we will work on, depending on the actual play between the infant and the parent, and the parents' responses to our questions. In PACT, the topic of each session is predetermined in advance by the method. This tenuous difference in the setting contains the core of what we consider the most important factor: the intersubjective link between the therapist and the patient or the parents. "Compared to the phenomenological approach, hermeneutic understanding is less unidirectional: it implies the co-construction of meaning and narratives in the course of the interactive process" (Fuchs, 2010). The early support of parents has recently developed as a fundamental trend in developmental psychopathology, becoming one of the four key points of interventions, together with the individualization of the intervention according to the developmental profile of the child, the expansion of learning targets, and the consideration of temporal characteristics (Wallace and Rogers, 2010).

Conclusion

The extraordinary evolution of our models of psychopathology underlines the contributions of developmental and psychodynamic frameworks. These models make it possible to relativize the intuitive trend to focus our interventions on the most specific symptoms and syndromes, to the detriment of the uniqueness of each patient. These new models offer opportunities to reduce the "endogenous/exogenous dichotomy" in the mental health field. We have reached an era of "generalist" interventions in child psychopathology, which are paradoxically based on the ultrasingular of the idiographic dimension. We must assume that the clinician's intuition is the guide of what we "feel," and that this feeling is not noise to be eliminated, but rather the core guide of our interventions. The phenomenology of the clinician, mobilized through the interaction with the patient, no doubt needs to be conceptualized: does it refer to counter transference? Access to the unspeakable? The psychodynamic framework and psychoanalytical listening are probably the best tools to identify these dimensions, as has been shown in a very interesting research (Cohen et al., 2011). Instead of demonizing psychoanalysis and second-person perspectives, the richness and the limitations of current models of psychopathology, and the contribution of developmental approaches, should force us to reconsider our theoretical frameworks (Ouss-Ryngaert and Golse, 2010). These empirically informed directions are not yet in the mainstream of psychopathology frameworks, which prioritize Evidenced Based educational or behavioral based and guidelined interventions, as they are more straightforward to train. No guideline will teach how to listen to the singularity of each patient. The question: which skills a clinician must possess and how to equip them adequately with such skills remains a crucial concern. The paradox is that therapist's non-specific skills, required for all types of psychotherapies (openness, attention to the patient's world, non-judgment, empathy…) are not consistently incorporated into training, and are often regarded as inherent to a psychotherapist's personality. We advocate that these skills should be developed during supervisions with experienced psychotherapists. Our proposals have to be developed to reach he deepest and the darkest corners of our patients, and of our own defenses.

Data availability statement

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author.

Author contributions

The author confirms being the sole contributor of this work and has approved it for publication.

Conflict of interest

The author declares that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Psychological and social consequences of deafblindness for siblings: a systematic literature review

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The onset of deafblindness profoundly impacts both the individual with this condition and the individual's family, including siblings. While current studies have primarily focused on the impact felt by parents or spouses, the distinct experiences of siblings have received comparatively less attention. This systematic review addresses the existing research gap regarding the psychological and social consequences experienced by siblings of individuals with deafblindness. A comprehensive search was conducted across multiple electronic databases, including PsycINFO, PsycARTICLES, Dissertations & Theses (on ProQuest), ERIC (Education Resources Information Center), International Bibliography of the Social Sciences (IBSS), Sociological Abstracts, Google Scholar, PubMed, and Cairn Info. Seven studies were identified as meeting the eligibility criteria for inclusion. The review revealed that siblings of individuals with deafblindness face psychological and social challenges, including emotions such as feelings of neglect, resentment, embarrassment, jealousy, and anxiety. Siblings also grapple with communication difficulties, contributing to feelings of exclusion and insecurity. In addition, these siblings take on significant responsibilities within the family and encounter obstacles in forming relationships outside the family. These findings underscore the need of interventions to improve the well-being of siblings of individuals with deafblindness by addressing their psycho-emotional needs and promoting positive social interactions. These findings align with studies conducted on siblings of children with other disabilities. However, additional research is crucial to investigate overlooked dimensions, particularly positive factors like coping mechanisms and resilience, that may influence the mental health and social experiences of these siblings.

KEYWORDS
deafblindness, siblings, family, psychological consequences, social experiences, siblings' psychotherapeutic interventions

Introduction

According to Watters et al. (2005), deafblindness is "a condition that combines any degree of hearing loss with any degree of vision loss that interferes with communicating and acquiring information, even though individuals who are deafblind may still have varying levels of useful vision and hearing" (p. 16). Indeed, deafblindness encompasses a spectrum of manifestations (Rodgers, 2021), and most subjects exhibit residual vision and hearing. The first global report of the World Federation of the DeafBlind indicates that approximately 0.2% of the world's population lives with severe deafblindness, whereas "milder forms" of deafblindness impact around 2% of the global population (International Disability Alliance, 2018). Moreover, individuals with deafblindness frequently exhibit additional physical and cognitive impairments (Heller et al., 1999).

Deafblindness manifests in various forms, including congenital, acquired, and aged-related deafblindness, each with distinct etiological factors and characteristics (Souriau, 2000; Dammeyer, 2010). Congenital deafblindness occurs at birth or shortly thereafter (Dammeyer, 2010) and is caused by genetic factors (e.g.: CHARGE syndrome), prenatal infections (e.g: cytomegalovirus, rubella,), or birth complications (e.g.: as prematurity, low birth weight,). Metabolic disorders, congenital malformations, certain medications, or maternal drug use are other potential causes of congenital deafblindness (Chen, 2004; Holte et al., 2006; National Center on Deaf-Blindness, n.d.).

Acquired deafblindness occurs later in life (following a period of normal sensory functioning) (Dammeyer, 2010) and can be attributed to genetic disorders (e.g.: Usher syndrome), traumatic events (e.g.: severe head injuries), infections (e.g.: meningitis), or specific neuronal conditions (e.g.: multiple sclerosis). Additionally, prolonged use of certain medications or exposure to toxic substances like chemotherapy drugs, antibiotics, or environmental toxins can potentially lead to acquired deafblindness (Chen, 2004; Holte et al., 2006; National Center on Deaf-Blindness, n.d.).

Age-related deafblindness is characterized by a gradual decline of both hearing and vision that arises due to the natural process of aging. This condition predominantly affects older adults, typically occurring after the age of 65. The primary causes of age-related deafblindness include presbycusis (age-related hearing loss), age-related macular degeneration, cataracts, and glaucoma, in conjunction with age-related hearing loss. Furthermore, other age-related health conditions like diabetes, cardiovascular diseases, and neurological disorders can also contribute to hearing loss and vision loss (Simcock, 2017).

Among syndromes associated with deafblindness, CHARGE syndrome is the most frequently encountered. However, in the context of acquired deafblindness, Usher syndrome predominates as the leading cause (Chen, 2004; Holte et al., 2006; National Center on Deaf-Blindness, n.d.). The impact of deafblindness, a distinct condition, is multiplicative (i.e., not simply the sum of vision and hearing impairments) (Fletcher and Guthrie, 2013; Ferrell et al., 2014). For example, individuals with deafblindness cannot compensate for hearing loss through lip reading. Thus, deafblindness is best understood as a precise and individual condition with disabilities distinct from those associated with only vision or hearing impairment (Arcous et al., 2019).

Deafblindness correlates with lower education, increased poverty, and higher unemployment rates (International Disability Alliance, 2018). The age at which the condition starts has a big effect on how difficult it is to cope with, especially when the condition is present from birth, leading to significant limitations (Bruce, 2005; Ronnberg and Borg, 2011). Progressive conditions, like Usher syndrome, bring multifaceted challenges, including access to information, mobility problems, workplace and educational difficulties, social isolation, feelings of insecurity, difficulties in projecting into the future and mental health issues (Arcous et al., 2019). Despite these challenges, appropriate accommodations can mitigate difficulties, which thereby enhances the quality of life for individuals with Usher syndrome (Ellis and Hodges, 2013; Arcous et al., 2019). These accommodations include alternative communication methods, assistive technologies, leisure activities, and social support (given by friends or family) (Arcous et al., 2019).

Indeed, the family plays a fundamental role in the well-being of the person with deafblindness (Spring et al., 2012). The family is the primary source of support (Ellis and Hodges, 2013; Arndt and Parker, 2016). According to Bernard (2013), "The quality of life of deafblind children and adults is greatly influenced by the connection, appropriate support, and interactions with the family" (p. 135). Family members can help with travel, administrative procedures, and daily tasks (Kyle and Barnett, 2012). Family members also play an important role in interpreting and accessing information from the outside world (Kyle and Barnett, 2012; Simcock, 2017). Moreover, the family offers the possibility of social openness (Gullacksen et al., 2011; Arndt and Parker, 2016). Sometimes family members are the only people individuals with deafblindness see during the week. The family can also support self-determination of the deafblind individual (Morgan et al., 2002).

According to Siemon (1984), "Because families are a system, distress in one member affects both the system and each member in it." (p. 294). It is necessary to acknowledge that the presence of a child with a disability in a family can lead to emotional consequences (felt among all family members), along with additional challenges like increased financial responsibilities and the need for extended caregiving. Families may also experience feelings of grief and loss, which can be amplified based on the severity of the disability (Correa-Torres, 2008). Family members may experience both social and psychological consequences due to the presence of a child with a disability.

Social consequences pertain to the effects and repercussions on individuals' social interactions, relationships, and integration within society, stemming from particular circumstances, events, or conditions. These consequences encompass strained social dynamics, social exclusion or isolation, challenges in forming and maintaining relationships, stigma or discrimination, and difficulties in engaging in social activities or fulfilling social roles (Brooks, 1980; Mormiche and Boissonnat, 2003). Psychological consequences encompass the effects and influences on individuals' emotional, cognitive, and behavioral well-being from experienced circumstances or events. These consequences encompass emotional responses such as stress, anxiety, depression, anger, sadness and cognitive changes (Jover, 2014; Ha Namkung and Carr, 2020).

Regarding the social consequences, the presence of an individual living with deafblindness poses communication challenges within the family (Souriau, 2000; Rodgers, 2021). Indeed, a study conducted by Kyle and Barnett (2012), among 39 individuals with acquired deafblindness, demonstrated that family members may encounter difficulties acquiring tactile sign language or other specific communication skills tailored to the disability. It is thus evident that effective communication within the family plays a pivotal role in managing the disability, fostering the self-esteem of the individual with a disability, and promoting family cohesion (Miner, 1995; Wahlqvist et al., 2020).

When deafblindness is acquired progressively, this process can provoke significant familial disruptions, which in many cases leads to entire role realignments assumed by family members (Gullacksen et al., 2011)— a process that is neither straightforward nor undemanding. For instance, according to Watters-Miles (2014), who conducted a study among six individuals with Usher syndrome, parents may become overprotective of their children. Hersh (2013) study on individuals with deafblindness (unspecified type) similarly observes protective behaviors demonstrated by parents. Miner (1995) study, focusing on individuals with Usher syndrome type I, emphasizes the need for adjustments in familial relationships as the syndrome progresses. Similarly, Figueiredo et al. (2013) study, involving eleven individuals with Usher syndrome (type I or II), supports Miner's study results.

The presence of a child with deafblindness can moreover present challenges in socialization beyond the immediate household for family members. A study conducted by Hersh (2013), which examined twenty-seven individuals with deafblindness (without specification of the type), revealed that families with children with deafblindness might experience feelings of stigmatization and embarrassment, ultimately leading to adverse consequences for their social participation.

Regarding the psychological consequences, Watters-Miles (2014) emphasizes that the onset of a disability can be traumatic for the individual and the entire family. Miner (1995), focusing on individuals with Usher syndrome type I, highlights that communication difficulties, hindered by the nature of the disability, can result in additional stress and can increased risk of depression among family members. Indeed, according to Correa-Torres (2008), sighted-hearing parents of children with deafblindness are at a higher risk of developing depression. Hartshorne and Schmittel (2016) who conducted a study on the social–emotional development of children with deafblindness (all types) also found that the risk of sighted-hearing parents developing depression is greater. Emotional problems were also reported by partners of individuals with Usher type 1. These partners appear to have more daily responsibilities and feel tired, depressed and resentful (Miner, 1995).

In summary, the familial implications of deafblindness are both pervasive and multidimensional, extending to siblings who have been relatively overlooked in current literature. Addressing these gaps is vital for the inclusive support and well-being of all family members. Indeed, the siblings of children with deafblindness confront unique challenges due to the very nature of their brother or sister's disability. Because of visual and auditory impairment, siblings are greatly hindered in their ability to communicate and play with their disabled sibling.

Siblings also assume increasingly vital roles within the family, often enduring lifelong relationships with their disabled siblings (Cicirelli, 1995). Indeed, siblings share a deep and enduring history together. As parents age, siblings may take on greater responsibility and play a more meaningful role in their relationship with a sibling with a disability (Vert et al., 2016). Also, siblings serve as primary witnesses to the challenges faced by the disabled child and their parents (Dayan and Scelles, 2017).

Despite this, research has primarily concentrated on parents and spouses, leaving a gap in understanding the psychological and social experiences of siblings of children with deafblindness.[1] To bridge this gap in knowledge, this systematic review aims to explore the existing literature on the psychological and social consequences of deafblindness on siblings. Gaining a comprehensive understanding of their experiences is crucial for providing inclusive support and enhancing the overall well-being of the entire family.

Methodology

This systematic literature review was carried out to explore the existing literature on the psychological and social consequences of deafblindness on siblings.

Search strategy

We conducted a comprehensive literature search using various electronic databases, including PsycINFO, PsycARTICLES, Dissertations & Theses (on ProQuest), ERIC (Education Resources Information Center), International Bibliography of the Social Sciences (IBSS), Sociological Abstracts, Google Scholar, PubMed, and Cairn Info. The following combination of words was used to conduct the systematic review search: "Siblings" OR "Brother," OR "Brothers" OR "Sister" OR "Sisters" OR "Fratrie" OR "Frère" OR "frères" OR "sœurs" OR "Soeur" AND ("Sourdaveugle" OR "Sourd-aveugle" OR "Surdicécité" OR "Deaf-blind" OR "Deafblind" OR "Deafblindness" OR "Dual Sensory loss" OR "dual sensory impair" OR "Dual sensory impairment" or "Dual sensory disability" OR "Usher Syndrome" OR "Wolfram Syndrome" OR "Stickler Syndrome" OR "Charge Syndrome" OR "Alport Syndrome" OR "Bardet-Biedl Syndrome" OR "Rubella" OR "Cockayne Syndrome" OR "Cornelia de Lange Syndrome" OR "Flynn-Aird Syndrome" OR "Goldenhar Syndrome" OR "Deaf-blind Hypopigmentation Syndrome").

Inclusion and exclusion criteria

Studies were included in the review if they were (1) written in English or in French languages (two languages spoken by the authors), (2) published in a peer-reviewed journal/ published dissertation/thesis studies or published reports with no date limits for the publication year (3) explored life experience and/or psychological health and/or social experience of siblings of individuals with deafblindness, and (4) articles mentioning the terms "fratrie" (siblings), "siblings," "frères" (brothers), "frère" (brother), "sœurs" (sisters), "sœur" (sister).

Studies were excluded from the review if they primarily focused on age-related deafblindness. This exclusion criterion was implemented to ensure that the included studies specifically addressed the experiences of siblings who grew up in the same household as individuals with deafblindness during childhood or adolescence.

Date selection and collection process

The articles selection process was conducted by two authors (first and second author). Here are the 6 steps that we followed for the article selection: (1) Database Retrieval; (2) Duplicate Removal; (3) Title and Abstract Screening; (4) Accessibility Check; (5) Criteria-Based Selection; and (6) Final Selection.

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[1] In this context, siblings refer to each of the two or more children or offspring who share one or both parents, specifically brothers or sisters.

1. Number of articles obtained in each database: the number of articles collected from each database was recorded to determine the initial pool of resources available for analysis. One thousand six hundred fourteen (1614) articles were found in total in the databases.
2. Number of articles set discarded as duplicates: to ensure data integrity and avoid redundancy, duplicate articles across databases were identified and removed from the pool. This step helped streamline the analysis process and eliminate any repetition. Two hundred and seventeen duplicates were identified, and 1,397 articles remained after removing these duplicates.
3. Number of articles set aside based on titles and abstract: after obtaining the articles (1,397 after removing the duplicates), a screening process was conducted based on their titles and abstracts. Articles that did not appear relevant to the research topic or did not meet the inclusion criteria were discarded. Many articles discussed the chosen syndromes or the chosen syndromes in relation to siblings. However, most of these studies had a medical perspective, aiming to uncover the genetic factors behind disease transmission rather than exploring the personal and social experiences of the siblings. One thousand three hundred fifty-one (1351) were thus rejected at this step in our methodology. Eighteen articles remained.
4. Among the eighteen remaining articles, we excluded some due to accessibility issues. In cases where we could only access the title or the abstract without the full text, we left these articles out of our analysis. This was done to ensure a thorough analysis, focusing on complete articles rather than just titles or abstracts. Specifically, we excluded two articles because we could only retrieve their titles, and four more because we had access only to their abstracts.
5. Number of articles selected for analysis: from the remaining articles, a selection was made based on the predetermined criteria for inclusion. These criteria could include relevance to the research question or methodology problems. Twelve articles were screened for eligibility using the inclusion and exclusion criteria. Five articles were excluded.
6. Final number of articles selected: after screening and applying the inclusion criteria, the remaining articles constituted the final set of resources selected for analysis. The number of articles (7) at this stage represents the data used in the research study.

Data extraction

The two authors extracted the selected articles based on their relevance to the inclusion criteria. One author was responsible for categorizing the results into psychological and social consequences, utilizing the definitions provided in the introduction. The second author validated this categorization. Information about the country the research was carried out, the age of the siblings, the type of deafblindness of the siblings, the methodology used in the articles and the bias and limitations presented by the authors of each article were also gathered (Figure 1).

Results

General characteristics of the studies

In total, there were five studies conducted in the United States, 1 study in Australia, 1 study in Canada, and 1 study in the United Kingdom. Only four studies directly assessed siblings' experiences with their siblings with deafblindness. The other three studies examined siblings' experiences through the perspectives of parents or individuals living with deafblindness.

Rowan (1990) conducted a study in the United States involving 12 siblings of deafblind children from five families. The age range of the siblings in this study was between 7 and 16 years old. Another study in the United States by Laman (2006) focused on four siblings ranging in age from 19 to 22 years old, with a sibling with congenital deafblindness. Harland and Cuskelly (2000) conducted a study in Australia with four siblings from different families. The age range of the siblings in this study was between 21 and 30 years old.

The causes of deafblindness in siblings with this disability were different. We believe it is necessary to clarify this because the experiences of non-disabled siblings may vary depending on the severity of the visual and hearing impairment. Rowan (1990) discussed different conditions, such as cortical visual impairment, profound hearing loss, and associated complications. Laman (2006) focused on congenital deafblindness without specifying the cause. Harland and Cuskelly (2000) explored cases of moderate to profound vision and hearing impairments, often accompanied by intellectual and physical disabilities. Vert et al. (2016) specifically studied individuals with CHARGE syndrome. Watters et al. (2005) investigated both acquired and congenital forms of deafblindness. Ellis and Hodges (2013) examined Usher syndrome, distinguishing between type I, type II, and type III. Finally, Heller et al. (1999) study focused on children with deafblindness without specifiying the various types.

Several studies have adopted qualitative methods to explore siblings' experiences in the context of deafblindness. Rowan (1990), a graduate student in education, conducted one-on-one interviews with siblings to gain insights into their perspectives. Laman (2006), a doctor in special education, employed semi-structured interviews to examine adult siblings' perceptions of their involvement in the Individualized Transition Plan (ITP) of siblings with congenital deafblindness. Harland and Cuskelly (2000), two researchers in special education, conducted semi-structured interviews on sibling responsibilities, support, personal development, advocacy, and more topics. Watters et al. (2005), researchers in disability studies, conducted focus groups with individuals with deafblindness, parents/advocates, and interviews with service providers. Ellis and Hodges (2013), researchers in education, employed semi-structured and extensive interviews.

In addition to qualitative approaches, quantitative studies have examined siblings' experiences with deafblindness. Vert et al. (2016) utilized the Sibling Evaluation Questionnaire, UCLA Loneliness Scale, Network Orientation Scale, Family Hardiness Index, and Family Member Well-Being Index to assess various sibling experiences and well-being aspects. Furthermore, Heller et al. (1999) used a questionnaire to evaluate parents' perceptions of siblings' interactions with their brothers and sisters with deafblindness (Table 1).

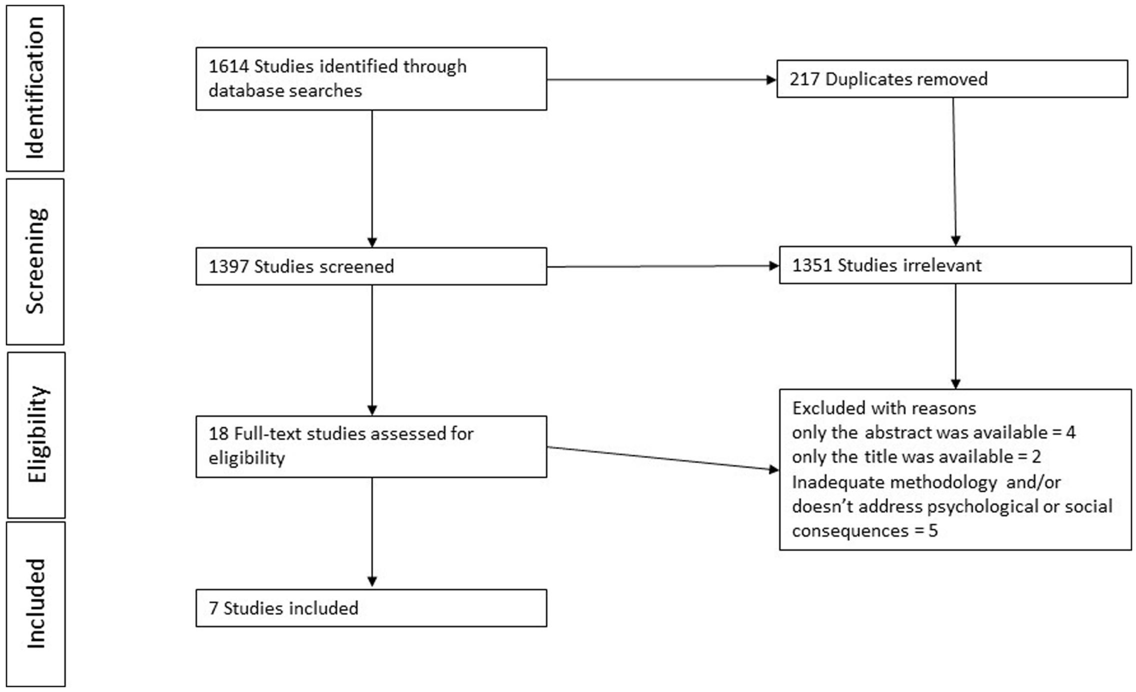

FIGURE 1
Data selection process.

TABLE 1 Characteristics of the studies.

Authors/date	Country	Type of publication	Type of study
Rowan (1990)	USA	Master dissertation in Education	Qualitative And Quantitative
Heller et al. (1999)	USA	Peer-reviewed article	Quantitative
Watters et al. (2005)	Canada	Report from the Canadian Society of deaf-Blind	Qualitative and Quantitative
Laman (2006)	USA	Dissertation in Special Education for the degree of Doctor of Education – Faculty of Texas Tech University	Qualitative
Harland and Cuskelly (2000)	Australia	Peer-reviewed article	Qualitative
Ellis and Hodges (2013)	U.K	Research report University of Birmingham	Qualitative and Quantitative
Vert et al. (2016)	USA	Peer-reviewed article	Quantitative

Quality of the study

Several limitations and biases have been identified in the examined studies by the authors of each study themselves, highlighting the importance of considering these factors when interpreting the findings. As an example, an interview may lead to the unintended procurement of socially desirable responses, especially when it is based on voluntary participation (Rowan, 1990; Harland and Cuskelly, 2000; Laman, 2006). Also, as Rowan (1990) mentioned, the understanding of questions by siblings and the author's interpretation could influence results.

Small sample sizes from specific regions limit the generalizability of the findings (Rowan, 1990; Heller et al., 1999; Harland and Cuskelly, 2000; Laman, 2006; Vert et al., 2016). Additionally, three studies did not directly assess the siblings experience but investigated people with deafblindness' perspective of their siblings' experience (Watters et al., 2005; Ellis and Hodges, 2013) or parents' perspectives (Heller et al., 1999) (Table 2).

Psychological consequences

Siblings may experience certain negative emotions such as resentment towards their parents, primarily due to the parents' reduced physical and emotional availability (Watters et al., 2005). Some siblings experienced feelings of exclusion and jealousy due to the additional parental attention given to the child with deafblindness (Watters et al., 2005). For example, in the study conducted by Ellis and Hodges (2013), Bethany (17, type 2) shared her experience of being perceived as the "favorite" among her siblings because she received more attention, which created tension with her sibling. In contrast, another study by Rowan (1990) revealed that nine out of twelve siblings did not perceive their siblings with deafblindness as more fortunate due to the attention or special treatment' received by the child with deafblindness.

TABLE 2 Characteristics of the studies.

Authors/date	Participants	Methodology	Limitations/bias
Rowan (1990)	12 siblings of deafblind children from five families in the Utah Intervener Services Program. 8 males and 4 females, ages ranging from 7 to 16 years old. Deafblind children: 2 males and 3 females, ages ranging from 1 year 10 months to 3 years 5 months. Deafblind children's conditions: failure to thrive, cortical visual impairment, profound hearing loss; cortical visual impairment, mild hearing loss; cortical visual impairment, cortical hearing impairment; severe handicaps, cortical visual impairment, cortical hearing impairment; profound hearing loss, retinopathy of prematurity, exotropia of the right eye. Additional complications: Many of the deaf-blind children had complications such as requiring oxygen and tube feeding, and some had seizures.	Qualitative: one-on-one interviews with the siblings. Quantitative: The Siblings' Perceptions of the Intervener Interview (SPII) and Taylor's Siblings' problems Questionnaire were administered.	Voluntary participation, potentially biased towards individuals with positive attitudes. Small sample size from only one state, limiting generalizability to other siblings of deaf-blind children. The assessment tools used lacked reliability and validity information. Only one interview may have resulted in socially desirable responses. Siblings' understanding of questions and the author's interpretation could influence results.
Heller et al. (1999)	36 parents of children with deafblindness. Ranging age of the children 1 to 22 years old. A sibling is living at least at home. Ranging age of the siblings (1 to 22 years old). Type of disability: Congenital deafblindess. Most participants' children have additional physical complications, such as orthopedic impairments, health impairments, and mental retardation. Most siblings were older than the child with deafblindness (61.1%). Siblings gender distribution: Almost an even distribution by gender for both the deaf-blind children and their siblings. Deaf-blindness causes: Majority of children had deaf-blindness due to multiple congenital anomalies (Charge syndrome or Rubella was the most present). Additional disabilities: 92% of children with deafblindness had additional disabilities, including orthopedic impairments, health impairments, and mental retardation.	Questionnaire to evaluate parents'perceptions of siblings'interactions with their brothers and sisters who are deafblind.	Smal sampel size. Only parents perception and not siblings's one.
Watters et al. (2005)	44 participants with deafblindness (29 females and 15 males). Age ranging from 20 to 75 years old. 42 respondents with acquired deafblindness and 2 respondents with congenital deafblindness. Focus group with parents/advocates: Date of the focus group is not available. Interviews with service providers: Data from the interviews are not available.	Quantitative: Questionnaire to gather demographics data (through telephone/emails) Qualitative: Focus groups with deafblind people or parents/advocates Service provider interviews	The reported experiences are not directly from siblings but from individuals living with deafblindness. Number of parents/advocates who participated to the focus group is unspecified. Number of service providers who participated to the interviews is unspecified Limited to Canada.

(Continued)

TABLE 2 (Continued)

Authors/date	Participants	Methodology	Limitations/bias
Laman (2006)	4 siblings, age ranging from 19 to 22 years old. Who have a sibling with congenital deafblindness (cause undefined)	Semi-structured interviews	Voluntary participation, potentially biased towards individuals with positive attitudes. Small sample size from only one state, limiting generalizability to other siblings of deaf-blind children. Only one interview may have resulted in socially desirable responses. Siblings' understanding of questions and author's interpretation could influence results.
Harland and Cuskelly (2000)	4 siblings from different South East Queensland, Australia families participated in the study. Age ranging from 21 to 30 years old. Type of disability: moderate to profound vision and hearing impairments, and in some cases, additional intellectual and physical disabilities.	Semi-structured interviews covering various topics, including responsibilities, support, development, and suggestions for improving quality of life. Grounded theory was used as the research approach.	Only one sibling per family was interviewed. Voluntary participation of siblings may introduce bias. The most supportive sibling may have been selected by the parent in families Siblings' biases towards the researcher and interpreter may affect validity. Small sample size. Cultural differences may impact the transferability of disability impact studies.
Vert et al. (2016)	29 siblings of children with charge syndrome. Age ranging from 13 to 42 years old. 14 males and 15 females. Severity of CHARGE syndrome: Majority (69%) of individuals with CHARGE were rated as moderately affected, with a minority being mildly or severely affected.	Sibling Evaluation Questionnaire (SEQ) UCLA Loneliness Scale (LS) Network Orientation Scale (NOS) Family Hardiness Index Family Member Well-Being Index	CHARGE research may not apply to other syndromes. Severity of disability affects sibling adjustment. Small sample size Lack of sibling relationship measures, SEQ validation needed. Siblings may show positive bias, influencing results.

Moreover, in Harland and Cuskelly (2000) study, some siblings expressed guilt over their inability to take a more active role in caring for their disabled sibling. They recognized the burden of care placed on their parents but felt helpless in changing the situation. In this study, one sibling expressed his guilt by saying, "I wish I could do more for Mum because she does do an awful lot." (p. 300). The same authors noticed that some siblings encounter feelings of anxiety as they grapple with understanding and managing their disabled sibling's behavior. Interacting with a sibling who has disabilities can indeed present challenges, potentially leading to anxiety and frustration for siblings (Harland and Cuskelly, 2000). Communication barriers, personal insecurities, and the uncertainty surrounding their sibling's receptiveness to communication may contribute to feelings of exclusion and insecurity (Harland and Cuskelly, 2000).

Also, siblings may experience a sense of inequity and frustration because of the family's financial problems that are caused by having a child with deafblindness (Watters et al., 2005). Additionally, in Rowan study's (1990), some siblings mentioned that they felt they bore additional responsibilities which caused frustration and a sense of unfairness. In Rowan study's, some siblings (3 out of 12) stated they put pressure on themselves to succeed academically or in other areas to compensate for the limitations of their sibling with deafblindness. This self-imposed pressure could contribute to their anxiety. Social embarrassment due to friends' reactions and questions further exacerbated the emotional toll (Rowan, 1990).

Anxiety may also arise from difficulty envisioning the future. Indeed, sibling disability can significantly impact an individual's perception of their future, as they envision a lifelong role in the life of the child with deafblindness (Harland and Cuskelly, 2000; Laman, 2006). In Laman (2006) study, all the siblings expressed concerns and anxieties regarding their future roles once their parents could no longer support the person with disabilities. Those results are supported by Harland and Cuskelly (2000) results. In Harland and Cuskelly's study, some siblings also expressed anxiety and concerns for the future. The siblings recognized the importance of future financial support. They acknowledged that their roles would need to evolve as their parents could not provide primary home-based care for their siblings. Furthermore, many siblings expressed inadequacy in not being able to assume greater responsibility for supporting their siblings. This sense of inadequacy was influenced by various factors, resulting in stress and conflict among most siblings (Harland and Cuskelly, 2000). Time constraints emerged as a concern for five out of twelve of the siblings in Rowan (1990). They expressed worries about having limited time to support their siblings with disabilities due to their numerous other commitments.

Furthermore, some siblings experienced deep distress regarding their lack of preparedness for their future responsibilities towards their sibling with deafblindness. Despite the anxiety stemming from their parents' mortality, all the siblings were willing to assume increased responsibility if necessary to support their siblings with disabilities (Harland and Cuskelly, 2000). In Harland and Cuskelly (2000) study, most siblings were expected to continue their responsibility for supporting their sibling's personal development. However, some siblings, like Phillip (a sibling interviewed by Harland and Cuskelly, 2000), doubted their ability to take on teaching responsibilities themselves.

The search for suitable accommodation for their sibling with deafblindness was identified as a significant concern for some siblings. In Harland and Cuskelly (2000) study, one sibling stated, "There is nothing for the deafblind in the area of job preparation and placement" (p. 302). Another sibling was concerned about her ability to find appropriate accommodation for her sister in the future. Some siblings also expressed anxiety about the level of care their brother or sister would receive in non-family-based supported accommodation settings (Harland and Cuskelly, 2000). Assisting with or managing their sibling's financial affairs was another responsibility that most siblings anticipated undertaking in the future (Harland and Cuskelly, 2000). Rowan (1990) stated that the worries and concerns expressed by some siblings of children with deafblindness encompassed fears, concerns, worries and anxieties about their sibling's mobility, mortality, future employment prospects, and access to education (Rowan, 1990).

Harland and Cuskelly (2000) noticed that some siblings expressed previous apprehensions about the potential of having children with disabilities themselves. One participant in their study sought genetic counseling to address this concern, and another one worried that prospective partners might mistakenly assume genetic complications associated with their relationship.

The experience of having a sibling with deafblindness can give rise to a strong need for information among siblings, leading to confusion, incomprehension, and anxiety. Understanding the specific problems faced by the child with deafblindness is another significant concern for siblings. Some siblings expressed desire to gain deeper insights into terminology, causes of the handicap, functioning abilities, caregiving techniques, preferences, and future prospects for their sibling (Rowan, 1990). They may desire to understand the nature and extent of their sibling's disability in hopes of finding a sense of clarity about what it means to be deafblind (Rowan, 1990). They questioned whether their sibling is truly deaf, blind, or both, highlighting the need for accurate information to dispel misconceptions (Rowan, 1990). In addition to seeking information, siblings recognized their responsibility as advocates for their siblings with deafblindness. They strongly desired to learn more about available services and opportunities, understanding that this knowledge is crucial for their future role. However, the lack of information leaves them uncertain about their sibling's future plans and legal matters, causing anxiety and a need for clarity (Harland and Cuskelly, 2000). Concerns about future living arrangements and the potential changes in their sibling's condition further contribute to siblings' anxiety. Some siblings believed that their sibling with deafblindness may not always live at home, while others believed their sibling's deafblindness may change over time (Harland and Cuskelly, 2000). In Heller et al. (1999) study, 67% of the parents recognized the siblings' desire to learn more about interacting and communicating with their sibling with deafblindness, emphasizing the importance of knowledge and understanding in facilitating meaningful connections.

Banta (1979) highlighted the siblings' greater distress compared to their parents as a result of ineffective coping mechanisms. As Harland and Cuskelly (2000) noticed, siblings relied heavily on their parents for help and guidance in caring for their sibling with deafblindness. They sought assistance in understanding the special attention and care required by a child with deafblindness, learning how to use specialized equipment, effectively communicate, handle emotional situations, and provide proper nourishment (Harland and Cuskelly, 2000). To cope with their worries and problems, siblings actively sought emotional support from various sources. Mothers are often their primary confidants, followed by fathers and friends (Rowan, 1990) (Table 3).

Social consequences

Siblings may experience difficulties in interaction with their deafblind brother or sister. Indeed, the presence of communication difficulties related to the nature of the disability, which affects language, makes it difficult for siblings to engage in activities with their sibling with deafblindness (Heller et al., 1999; Watters et al., 2005). Communication is an important aspect of the sibling relationship that often needs adjustments and adaptations, commonly achieved through augmentative and alternative methods of communication. In the case of siblings with a brother or sister with deafblindness, it has been observed that they mostly rely on nonsymbolic forms of communication to interact. However, there is a noticeable absence of alternative communication methods, such as tactile or visual communication boards/systems, and limited involvement from professionals in teaching siblings how to communicate with their sibling with deafblindness. Interestingly, most siblings acquired nonsymbolic communication skills independently, as reported by 62.5% of parents and 56.3% of parents of elementary school children (Heller et al., 1999). On the other hand, sign language is predominantly taught by a parent/relative or, in some instances, by a speech therapist, as seen with two older children. Among parents who reported that siblings face communication challenges consistently, those who rely on nonsymbolic forms of communication experienced the greatest difficulty. Additionally, it seems that the nature of these difficulties is influenced by the severity of the impairment and the availability of alternative communication methods (Heller et al., 1999).

In Heller et al. (1999) study, most parents (over 25%) reported that siblings have minimal interaction or deliberately try to avoid their brother or sister with deafblindness. Many parents (22.2%) and parents of elementary school children (16.7%) indicated that siblings never try to engage their brother or sister in any activity. Regarding the duration of sibling engagement, approximately 23.5% of parents and 21.7% of parents of elementary school children reported a daily involvement of 5–15 min, while 20.6% of parents and 26.1% of parents of elementary school children reported spending 30 min to an hour together. However, a notable percentage of parents (14.7%) and parents of elementary school children (21.7%) mentioned that siblings spend less than 5 min with their sibling with deeafblindness, and 17.6 and 8.7% of parents and parents of elementary school children,

TABLE 3 Results.

Authors/date	Psychological consequences	Social consequences
Rowan (1990)	Seek information on deafblindness. Feel embarrassed and perceive adverse reactions from friends. Experience frustration, anxiety, and early responsibilities with pressure to succeed. Bear additional household duties, leading to frustration and unfairness. Express guilt over limited involvement in caring for their disabled sibling. Have concerns about future roles without parental support. Recognize the importance of emotional support from parents and friends Do not perceive their sibling as fortunate or receiving special treatment.	Take on various responsibilities within the family dynamics to care for a deaf-blind individual. Caregiver and protectors. Possess knowledge and skills to assist the deaf-blind individual. Seek information on services, living arrangements, and the condition of the deafblind individual.
Heller et al. (1999)	Feel anxious about the future.	Adopt an helping role. Seek support for communication with their deafblind sibling. Limited interaction or avoidance may occur. Adjustments for participation may be lacking. Communication challenges hinder interaction. Nonsymbolic communication is common. Communication adjustments are necessary.
Watters et al. (2005)	Resentment towards parents and the deafblind child Experience negative emotions like exclusion, jealousy, and resentment. Bearing financial responsibility for their activities may cause a sense of inequity and frustration. Perceive it as unfair and straining the family's limited resources.	Face difficulties in interacting with their deafblind sibling due to communication challenges. Communication adjustments and adaptations, including augmentative and alternative communication methods.
Laman (2006)	Anxiety and uncertainty about what is expected from them in the future.	Want to be involved in their sibling's future.
Harland and Cuskelly (2000)	Interacting with a sibling with disabilities presents challenges and causes anxiety and frustration. Siblings express a need for information and understanding to support their siblings. Express guilt over their inability to take a more active role in caring Experience anxiety in managing their sibling's challenging behavior. Concerns arise about their sibling's future plans, legal matters, living arrangements, changes in condition, and employment prospects. Stress and conflict arise due to feelings of inadequacy and time constraints. Siblings feel unprepared for future responsibilities and doubt their ability to assume teaching roles. Have apprehensions about having children with disabilities themselves. Emotional support from parents and friends is recognized as essential and sought after.	Face challenges and disadvantages, Encounter attention and stares from others, limited space, and disrupted routines. Expressing dissatisfaction, they mention the interference of their deaf-blind sibling in their lives and plans. Embrace a helping role.
Ellis and Hodges (2013)	Jealousy felt by siblings towards the deafblind child. Perceive favoritism among their siblings, as they receive more attention from adults.	In need of support and advice from specialist services and networks who understood Usher syndrome better.
Vert et al. (2016)	Rate their personal well-being positively. Siblings' relationship are associated with hardiness, loneliness, and Charge knowledge.	Accept their sibling with Charge syndrome neutrally. Report less loneliness, and similar social support access. Benefit from interacting with other children with disabled siblings.

respectively, stated that no dedicated time is spent between siblings. The frequency of difficulty encountered in playing or engaging in activities together varied among respondents. The majority of parents (44.4%) and parents of elementary school children (41.7%) reported that such difficulties occurred some of the time. When questioned about the siblings' willingness to make necessary adjustments for their brother or sister with deafblindness to participate in games or activities that require visual perception, a significant percentage of parents (68.6%) and of parents of elementary school children (70.8%) indicated that siblings never made adaptations or only did so on certain occasions. Similarly, for activities requiring hearing, most parents (70.6%) and parents of elementary school children (73.9%) reported that siblings never made accommodations or only did so sometimes.

According to Rowan (1990), siblings of individuals with deafblindness play pivotal roles in family dynamics, specifically in caring for and supporting their sibling with deafblindness. In Rowan (1990), a majority (eight out of twelve) indicated that their parents expected them to take on additional responsibilities due to the presence of a sibling with deafblindness. These responsibilities span from babysitting their deafblind sibling and other siblings, assisting younger siblings with chores, bathing, feeding, changing diapers, dressing, providing tracheostomy care, engaging in playtime, attending to the child's needs during the night, cooking, and undertaking household to outdoor tasks. This underscores the heightened responsibility these siblings' experience, with two of the twelve respondents citing exclusive household roles arising from their deafblind sibling's needs (Rowan, 1990). Additionally, six of eleven siblings noted their caregiving role was more extensive than their peers' involvement with their own siblings. However, ten out of twelve siblings perceive their parents' rules as fair, indicating a balanced approach to managing family dynamics. Concerning knowledge and skills, six out of twelve siblings reported being adequately equipped to assist their sibling with deafblindness (Rowan, 1990).

In Heller et al. (1999) study, parents characterized the relationship between siblings and the child with deafblindness as "helping." The study revealed that while mothers were the primary caregivers, siblings played a significant albeit secondary role in supporting their brother or sister with visual and hearing impairments. Harland and Cuskelly (2000) found in their study that most siblings provided practical assistance to their brother or sister with deafblindness (e.g., in mobility, recreational activities, and respite care). In some cases, they even took on the role of being a parent for two weeks to give their parents a break (Harland and Cuskelly, 2000). Most siblings offered practical assistance by reinforcing or teaching their brother or sister with deafblindness new skills. One common responsibility shouldered by adult siblings was maintaining regular contact with their siblings with disabilities. In fact, adult siblings maintained regular contact and emotional support, reinforcing or teaching new skills *regardless* of geographic proximity (Harland and Cuskelly, 2000).

Siblings of children with deafblindness encounter various challenges and disadvantages in their everyday lives. Public attention when accompanying their sibling with deafblindness and crowded living conditions due to special equipment are among the drawbacks (Rowan, 1990). Frequent hospital visits further add to the siblings' inconvenience and disruption of their personal routine. Another disadvantage is the impact on their participation in family activities, as their sibling's condition may require adjustments or prevent them from fully engaging in shared experiences (Rowan, 1990). In Rowan (1990), nine out of twelve siblings expressed dissatisfaction with how their brother or sister with deafblindness interfered in their lives. This dissatisfaction was often associated with the inability to implement family plans and activities due to their sibling's needs (i.e., a parent staying home with the child with deafblindness rather than joining the rest of a family on an outing). Furthermore, four out of ten siblings shared their discontent with their sibling's impact on their personal plans and activities. These concerns range from being unable to visit friends' houses or host them at their homes, feeling obligated to be involved with the care of their sibling with deafblindness, and having their activities disrupted due to the responsibility of looking after their other siblings. To address the limited participation in activities, it is recommended, within the literature, to modify the activities to accommodate the vision and hearing loss of the child with deafblindness. This modification can enhance siblings' ability to participate in and foster meaningful interaction. However, it is worth noting that siblings may lack information on how to effectively modify or identify suitable activities for their siblings with deafblindness (Rowan, 1990).

Rowan (1990) also notes the generally positive and protective attitudes of siblings towards their sibling with deafblindness. Notably, in Rowan (1990) all siblings demonstrated a favorable attitude and actively discouraged any teasing directed towards their sibling with deafblindness. Despite the challenges they may encounter, siblings cited genuine enjoyment/happiness stemming from their relationship with their brother/sister with deafblindness and are committed to safeguarding their well-being when necessary. Among the surveyed, six out of the twelve siblings mentioned that their friends visit their home and engage in various activities, such as playing pat-a-cake, throwing a ball, holding hands, and communicating with the child with deafblindness. On the contrary, six siblings reported that their friends do not come to their homes to interact with their siblings with deafblindness. When explaining their sibling's condition to friends, five out of the twelve siblings expressed no difficulty conveying the necessary information. Feelings of rejection were minimal (eight out of twelve), with most embracing their sibling's condition openly, showing unconditional love and acceptance. Several stated they did not wish for their deafblind sibling to be absent, highlighting the depth of their emotional bond (Rowan, 1990).

However, Vert et al. (2016) study found a correlation between siblings' level of difficulty in engaging with their sibling with deafblindness and their willingness to include them in activities with friends or attend support groups. This suggests the potential benefit of interaction with peers who also have siblings with disabilities (Table 3).

Discussion

While studies have explored the experiences of siblings of individuals with disabilities, the literature on siblings of those with deafblindness remains notably sparse. The existing research does, however, illuminate the unique psychological and social challenges these siblings face.

To summarize, the psychological consequences experienced by siblings of deafblind individuals can include a range of negative emotions such as resentment, jealousy, frustration, guilt, anxiety, and feelings of inadequacy. Siblings may feel resentment towards their parents due to reduced attention and availability, as well as jealousy towards the additional parental attention given to the deafblind child. They may also feel excluded and experience a sense of inequity and frustration. Siblings often express guilt over their inability to take a more active role in caring for their disabled sibling and may experience anxiety when managing their sibling's challenging behavior. Communication barriers, personal insecurities, and uncertainty about their sibling's receptiveness to communication can contribute to feelings of exclusion and insecurity. Siblings may also experience anxiety when envisioning the future, particularly regarding their own future roles, once their parents can no longer provide support for the person with deafblindness.

Social consequences for siblings of individuals with deafblindness can include difficulties in interacting with their siblings due to communication challenges related to the disability. Siblings often rely on nonsymbolic forms of communication to interact. Still, there may be a lack of alternative communication methods and limited involvement from professionals in teaching siblings how to communicate with their siblings with deafblindness. Siblings also take on significant responsibilities within the family dynamics, such as caregiving tasks and support for their sibling with deafblindness which impact the time they have for their social activities. In the context of family activities/outings, siblings may experience disadvantages, such as attention and stares from others, routines disruptions, and limited household space. Siblings may also encounter difficulties forming and maintaining relationships outside the family due to their sibling's condition. Some siblings may actively avoid or have minimal interaction with their deafblind sibling.

Similar experiences have been observed in siblings of children with other types of disabilities. For example, some studies on individuals with undefined physical or chronic disabilities demonstrated that siblings of children with disabilities can experience distress, unhappiness and resentment (Lamarche, 1985; Fisman et al., 2000; Scelles, 2011; Giallo et al., 2012; Hallberg, 2013).

Also, siblings of children with other disabilities (undefined physical or intellectual disabilities) can feel ashamed of how others perceive them and struggle with social inclusion (Lamarche, 1985; Dayan and Scelles, 2017).

Siblings of children with other disabilities (hemiparesis and undefined physical or intellectual disabilities) may exhibit premature and hyper-protective behaviors, feel responsible for the family's well-being, and seek partners who understand their role (Seligman, 1983; Hannah and Midlarsky, 1985; Scelles, 2004; Gardou, 2012; Dufreche Rastello, 2020).

In addition, siblings of children with other disabilities (undefined chronic illness) tend to have higher anxiety levels stemming from uncertainty in interacting with the disabled sibling (Heiney et al., 1990; Martinez et al., 2022).

Finally, siblings of children with hemiparesis express the same lack of information about the disability (Dufreche Rastello, 2020), as for siblings of individuals with deafblindness. Research indicates that siblings of children with hemiparesis desire more information but fear the implications it may have on themselves (Dufreche Rastello, 2020). Limited contact with professionals and guilt often prevents these siblings from asking questions that might equip them with the information they desire, such as questions that address the nature of their siblings disability (Smith and Perry, 2005).

In contrast, some aspects present in the siblings of children with disabilities were not reported in those of children with deafblindness. Negative emotions such as anger are commonly experienced by siblings of children with other disabilities (undefined physical or intellectual disabilities) (Seligman, 1983; Hallberg, 2013). These emotions, however, were not reported in siblings of children with deafblindness within our research. Siblings of children with intellectual disabilities may feel threatened by the possibility of death, disability, or contamination (McHale and Gamble, 1989). Also, guilt can inhibit aggressive feelings and lead to depressive symptoms for siblings of children with hemiparesis or physical or mentaldisabilities (Meynckens-Fourez, 1995; Griot et al., 2010; Dufreche Rastello, 2020). Feelings of loneliness, lack of social support, and withdrawal are also common challenges faced by siblings of children with facial paralysis or other undefined physical disabilities (Crocker, 1981; Pogossian, 2003; Guyard, 2012). These issues were not identified in our study. Differences in rules and increased sibling rivalry can lead to aggression and behavioral problems for some siblings of children with facial paralysis or other undefined physical disabilities (Breslau et al., 1981; Crocker, 1981; Lamarche, 1985; Meynckens-Fourez, 1995). Siblings of children with hemiparesis may exhibit behavioral issues to gain attention or wish to be disabled themselves (Dufreche Rastello, 2020). However, behavioral problems for siblings of individuals with deafblindness were not identified.

It is unclear if these aspects are absent, not investigated, or not expressed by the siblings in interviews due to modesty or fear. Also, Smith and Perry (2005) noticed that limited contact with professionals and guilt often prevent siblings of children with a disability from asking questions. Additionally, the desire to appear positive to the interviewer may have led to the concealment of information, such as anger, self- or hetero-aggressive behaviors, social difficulties, and feelings of loneliness. It is also possible that these aspects are less prevalent because the research primarily focused on more well-known syndromes such as Usher and CHARGE, for which more information and support groups are available, potentially introducing a representativeness bias.

For example, in Vert et al. (2016) study, siblings of people with deafblindness reported experiencing somewhat lower levels of loneliness, which contradicts earlier research suggesting higher levels of loneliness among siblings of children with intellectual disabilities (Rossiter and Sharpe, 2001). One possible explanation for this discrepancy is that families with a child with CHARGE syndrome have easy access to support systems through the Internet and various networks. While not many participants actually reported that they attended a CHARGE conference or a support group, the availability of these networks may have helped alleviate any potential feelings of loneliness associated with having a sibling with a disability.

Other factors influencing the results including socioeconomic status, past attendance at a sibling support group, parent stress, family time and routines, family problem-solving and communication, and family hardiness. However, these factors were rarely presented in the selected studies, and thus, we could not examine their detailed impact.

The self-esteem of siblings of children with deafblindness was not explored in our study/ Considering the psychological and social challenges siblings of children with deafblindness are facing, it would be interesting to investigate the consequences on their self-esteem. Previous research suggests that self-esteem in siblings of disabled children (with pervasive developmental disorder) is similar to that of other children (Ferrari et al., 1988). Positive effects such as compassion, empathy, and resilience can develop through the sibling bond (Powell et al., 1985; Metzger, 2005; Scelles, 2008; Griot et al., 2010; Von Benedek, 2013). According to Mchale and Gamble (1989) siblings of children with autism may also acquire responsibility and develop good self-esteem (McHale and Gamble, 1989). Also, for siblings of children with a pervasive developmental disorder or hemiparesis, social behaviors, skills, and creative abilities can be enhanced (Ferrari et al., 1988; Dufreche Rastello, 2020). The lack of results on these variables may be explained by the fact that research initially focused on negative consequences, given the relatively unexplored nature of the field due to the rarity of the disability.

Limitations

Our research is predicated upon the foundation of preceding studies, which are not without their limitations (e.g., sample size, methodology, and generalizability of results). It is recognized that these limitations can limit our own research. One prominent limitation and bias observed in the antecedent studies is the use of the voluntary participation method. This method has the propensity to introduce bias in the resultant data by attracting individuals with positive attitudes or specific interests, thereby potentially impinging upon the representativeness of the sample and the generalizability of the findings.

Sample size is another limitation— posing challenges in extrapolating the findings to encompass the experience of other siblings of children with deafblindness. It is thus imperative to consider this limitation and its consequential impact when interpreting the findings.

Additionally, relying solely on a single interview with each participant is another significant limitation of the antecedent research and our subsequent use/interpretation of these studies. This approach can influence participant responses, engendering socially desirable answers that may deviate from genuine responses. Consequently, the validity of the results and the comprehension of sibling relationships may be compromised.

Cultural differences can substantially influence the results and impede the transferability of studies concerning the impact of disabilities on families across different cultures. We must recognize and account for these cultural disparities when interpreting and applying the findings.

Furthermore, the inability to access texts not available in English and French presents an additional limitation in our research. This constraint may engender incomplete information and potentially result in overlooking pertinent insights.

It is important to note that specific texts could only be partially accessed in certain cases, with only the title or abstract being available. This further curtails our access to comprehensive information and a holistic understanding of those studies. The absence of full-text access may have omitted valuable details and insights.

The limited number of selected studies and the inability to access texts in other languages underscores the necessity for future research endeavors characterized by enhanced methodologies and augmented sample sizes (that ideally encompass greater diversity). Such endeavors will surmount these limitations and engender more robust, dependable, and generalizable results within the domain of deafblindness research.

Recommendations

Practical implications

In light of the challenges some siblings face, we recommend providing psychotherapeutic spaces[2] to address their needs (Rowan, 1990; Ellis and Hodges, 2013). The research we examined underscores, either explicitly or implicitly, the importance of support and care for siblings in (1) understanding the condition of deafblindness better, (2) expressing their emotions, and (3) acquiring information (Rowan, 1990; Ellis and Hodges, 2013). However, such therapeutic spaces for siblings remain limited, potentially due to a lack of awareness among parents and institutions (Plumridge et al., 2011).

To meet these needs, we recommend employing the Taylor's Siblings Questionnaire to identify the concerns of siblings of children with deafblindness and tailor interventions accordingly (Rowan, 1990). Professionals like psychologists and psychometricians can facilitate sibling groups and offer a safe environment for siblings to express themselves and develop a deeper understanding of disability-related issues (Pitman and Matthe, 2004; Dufreche Rastello, 2020). Participation in sibling groups has demonstrated numerous benefits, including reduced anxiety, enhanced self-esteem, and improved communication within the family (Heiney et al., 1990; Smith and Perry, 2005; Scelles et al., 2007; Plumridge et al., 2011; Scelles, 2011).

Professionals working with families should acknowledge the impact of disability on both siblings and parents, adopting a comprehensive approach to address their needs (Vert et al., 2016). Support programs should actively involve siblings in crucial educational and transitional meetings and provide sibling workshops to equip them with knowledge and skills (Laman, 2006).

Research implications

Future studies should incorporate diverse data sources to ensure reliable data collection (Vert et al., 2016). Further exploration of the role of interveners in supporting siblings and the implementation of larger-scale studies will contribute to a deeper understanding of their needs (Vert et al., 2016).

In the context of deafblindness, long-term studies focusing on interventions targeting psychological well-being, orientation and mobility, independence development, transition to adulthood, and sibling relationships are warranted (Rowan, 1990). Additional research is needed to delve into the positive aspects, coping resources, and resilience developed by siblings (Rowan, 1990).

To enhance the generalizability of research, it is recommended to include families from diverse geographical regions and incorporate the perspectives of siblings and children with deafblindness (Rowan, 1990). Conducting longitudinal studies on sibling relationships over time would provide valuable insights, particularly for children with deafblindness. This study highlights the importance of addressing limited interaction or avoidance between siblings and their brother/sister with deafblindness by providing tailored information and support for sibling relationships (Rowan, 1990).

Furthermore, future research should focus on larger and more diverse samples, employ improved methodologies, and directly incorporate siblings' perspectives to ensure more reliable and generalizable results in the field of deafblindness research.

Author contributions

MA did the mini-review. RP leads the research project of which this mini-review is a contribution and coordinated MA's work during the selection process and participated in the corrections. ND corrected the text and provided perspectives. All authors contributed to the article and approved the submitted version.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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[2] A therapeutic space is a specially designed environment that supports healing and therapy. It can be a physical or virtual space where individuals engage in therapeutic activities to improve their well-being. It promotes safety, comfort, and relaxation, and may include tools and materials to enhance the therapeutic experience.

Publisher's note

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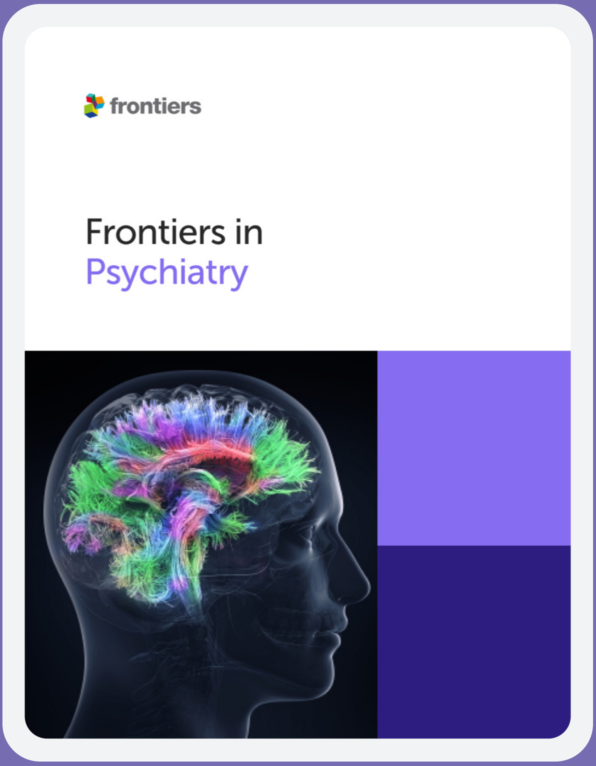

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