# Minimally invasive pediatric surgery: How to improve and overcome limitations

Edited by

Edoardo Bindi, Giuseppe Lauriti, Giovanni Cobellis and Gabriele Lisi

**Published in** Frontiers in Pediatrics Frontiers in Surgery





#### FRONTIERS EBOOK COPYRIGHT STATEMENT

The copyright in the text of individual articles in this ebook is the property of their respective authors or their respective institutions or funders. The copyright in graphics and images within each article may be subject to copyright of other parties. In both cases this is subject to a license granted to Frontiers.

The compilation of articles constituting this ebook is the property of Frontiers.

Each article within this ebook, and the ebook itself, are published under the most recent version of the Creative Commons CC-BY licence. The version current at the date of publication of this ebook is CC-BY 4.0. If the CC-BY licence is updated, the licence granted by Frontiers is automatically updated to the new version.

When exercising any right under the CC-BY licence, Frontiers must be attributed as the original publisher of the article or ebook, as applicable.

Authors have the responsibility of ensuring that any graphics or other materials which are the property of others may be included in the CC-BY licence, but this should be checked before relying on the CC-BY licence to reproduce those materials. Any copyright notices relating to those materials must be complied with.

Copyright and source

acknowledgement notices may not be removed and must be displayed in any copy, derivative work or partial copy which includes the elements in question.

All copyright, and all rights therein, are protected by national and international copyright laws. The above represents a summary only. For further information please read Frontiers' Conditions for Website Use and Copyright Statement, and the applicable CC-BY licence.

ISSN 1664-8714 ISBN 978-2-8325-5233-9 DOI 10.3389/978-2-8325-5233-9

#### **About Frontiers**

Frontiers is more than just an open access publisher of scholarly articles: it is a pioneering approach to the world of academia, radically improving the way scholarly research is managed. The grand vision of Frontiers is a world where all people have an equal opportunity to seek, share and generate knowledge. Frontiers provides immediate and permanent online open access to all its publications, but this alone is not enough to realize our grand goals.

#### Frontiers journal series

The Frontiers journal series is a multi-tier and interdisciplinary set of openaccess, online journals, promising a paradigm shift from the current review, selection and dissemination processes in academic publishing. All Frontiers journals are driven by researchers for researchers; therefore, they constitute a service to the scholarly community. At the same time, the *Frontiers journal series* operates on a revolutionary invention, the tiered publishing system, initially addressing specific communities of scholars, and gradually climbing up to broader public understanding, thus serving the interests of the lay society, too.

#### Dedication to quality

Each Frontiers article is a landmark of the highest quality, thanks to genuinely collaborative interactions between authors and review editors, who include some of the world's best academicians. Research must be certified by peers before entering a stream of knowledge that may eventually reach the public - and shape society; therefore, Frontiers only applies the most rigorous and unbiased reviews. Frontiers revolutionizes research publishing by freely delivering the most outstanding research, evaluated with no bias from both the academic and social point of view. By applying the most advanced information technologies, Frontiers is catapulting scholarly publishing into a new generation.

#### What are Frontiers Research Topics?

Frontiers Research Topics are very popular trademarks of the *Frontiers journals series*: they are collections of at least ten articles, all centered on a particular subject. With their unique mix of varied contributions from Original Research to Review Articles, Frontiers Research Topics unify the most influential researchers, the latest key findings and historical advances in a hot research area.

Find out more on how to host your own Frontiers Research Topic or contribute to one as an author by contacting the Frontiers editorial office: frontiersin.org/about/contact

#### Minimally invasive pediatric surgery: How to improve and overcome limitations

#### **Topic editors**

Edoardo Bindi — Salesi Hospital Foundation, Italy Giuseppe Lauriti — G. d'Annunzio University of Chieti and Pescara, Italy Giovanni Cobellis — Marche Polytechnic University, Italy Gabriele Lisi — University of Studies G. d'Annunzio Chieti and Pescara, Italy

#### Citation

Bindi, E., Lauriti, G., Cobellis, G., Lisi, G., eds. (2024). *Minimally invasive pediatric surgery: How to improve and overcome limitations*. Lausanne: Frontiers Media SA. doi: 10.3389/978-2-8325-5233-9

#### 🐉 frontiers | Research Topics

### Table of contents

05 Editorial: Minimally invasive pediatric surgery: how to improve and overcome limitations

Edoardo Bindi, Gabriele Lisi, Giuseppe Lauriti and Giovanni Cobellis

### 08 Case report: Minimally invasive removal of a dislodged thoracoamniotic shunt with an integral cystoscope in a preterm infant

Lichao Zhu, Yanze Wang, Honghao Song, Xiaoqing Wang, Mingang Zhang and Feng Guo

13 Case report: Pediatric floating elbow fracture with monteggia-equivalent lesion, ipsilateral humeral shaft fracture, and radial nerve injury: a unique case and favorable treatment outcomes

Li Ju, Mengqiu Xu and Gang Lin

19 Case report: Laparoscopic nissen-sleeve gastrectomy in a young adult with incidental finding of Morgagni-Larrey hernia

> Rossella Palma, Francesco Angrisani, Antonella Santonicola, Paola Iovino, Vittorio Maria Ormando, Roberta Maselli and Luigi Angrisani

- 23 Minimally invasive management of combined esophageal atresia with tracheoesophageal fistula and duodenal atresia: a comprehensive case report Eunyoung Jung
- 28 Comparative assessment of fully laparoscopic Duhamel-Z with minimal rectorectal dissection vs. laparoscopy-assisted Duhamel-Z with blunt manual rectorectal dissection for total colonic aganglionosis

Go Miyano, Hisae lida, Yu Ebata, Eri Abe, Haruki Kato, Takafumi Mikami, Junya Ishii, Geoffrey J. Lane, Atsuyuki Yamataka and Tadaharu Okazaki

34 Remote cadaveric minimally invasive surgical training

Go Miyano, Makoto Takahashi, Takamasa Suzuki, Hisae Iida, Eri Abe, Haruki Kato, Shiho Yoshida, Geoffrey J. Lane, Koichiro Ichimura, Kazuhiro Sakamoto, Atsuyuki Yamataka and Tadaharu Okazaki

40 Single-port-plus-one robot-assisted laparoscopic modified Lich-Gregoir direct nipple ureteral extravesical reimplantation in children with a primary obstructive megaureter

Yufeng He, Shan Lin, Xinru Xu, Shaohua He, Huihuang Xu, Guangxu You, Jianglong Chen and Di Xu

48 Inanimate 3D printed model for thoracoscopic repair of esophageal atresia with tracheoesophageal fistula

Petra Zahradniková, Jozef Babala, Rebeka Pechanová, Martin Smrek, Pavol Vitovič, Miroslava Laurovičová, Tomáš Bernát and Barbora Nedomová

- 57 Percutaneous transthoracic catheter drainage prior to surgery in treating neonates with congenital macrocystic lung malformation presenting with respiratory distress Taozhen He, Xiaoyan Sun, Dengke Luo, Shiyi Dai, Miao Yuan, Gang Yang, Kaisheng Cheng and Chang Xu
- 66 Preoperative anxiety management in children. Benefits of humanoid robots: an experimental study Ilaria Franconi, Andrea Faragalli, Giulia Palego, Samuele Canonici, Ludovica Gatti, Alessandro Simonini, Edoardo Bindi, Giovanni Cobellis and Flavia Carle
- 71 To use a simple hernia needle for single-port laparoscopic percutaneous inguinal hernia repair in children: a 5-year experience study

Li Zhang, Rui Zhang, Jianfa Zhang, Hailong Hu, Zihan Chen, Yanxiang Fu and Saihua Li

- Outcomes of olecranon fractures in adolescents: comparison of tension band wiring and Herbert screw fixations
  Weiwei Yang, Xintao Zhang, Dong Sun, Shaobin Jin, Junfei Chen and Yang Li
- 84 Case Report: Robotic pylorus-preserving pancreatoduodenectomy for periampullary rhabdomyosarcoma in a 3-year-old patient Zijian Liang, Menglong Lan, Xiaogang Xu, Fei Liu, Boyuan Tao, Xinxing Wang and Jixiao Zeng

Check for updates

#### **OPEN ACCESS**

EDITED AND REVIEWED BY Dennis Paul Orgill, Harvard Medical School, United States

\*CORRESPONDENCE Edoardo Bindi 🖂 edo.bindi88@hotmail.it

RECEIVED 10 June 2024 ACCEPTED 08 July 2024 PUBLISHED 17 July 2024

#### CITATION

Bindi E, Lisi G, Lauriti G and Cobellis G (2024) Editorial: Minimally invasive pediatric surgery: how to improve and overcome limitations. Front. Surg. 11:1446901. doi: 10.3389/fsurg.2024.1446901

#### COPYRIGHT

© 2024 Bindi, Lisi, Lauriti and Cobellis. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

# Editorial: Minimally invasive pediatric surgery: how to improve and overcome limitations

Edoardo Bindi<sup>1,2\*</sup>, Gabriele Lisi<sup>3</sup>, Giuseppe Lauriti<sup>3</sup> and Giovanni Cobellis<sup>1,2</sup>

<sup>1</sup>Department of Pediatric Surgery, Azienda Ospedaliero Universitaria Ospedali Riuniti, Ancona, Italy, <sup>2</sup>Department of Clinical Specialty Science and Dentistry, Marche Polytechnic University, Ancona, Italy, <sup>3</sup>Department of Medicine and Science of Aging, University of Studies G. D'Annunzio Chieti and Pescara, Chieti, Italy

#### KEYWORDS

minimally invasive surgery, robotic surgery, laparoscopy, thoracoscopy, pediatric surgery

#### Editorial on the Research Topic

Minimally invasive pediatric surgery: how to improve and overcome limitations

The evolution of minimally invasive pediatric surgery is one of modern medicine's most exciting and progressive chapters. This practice, which originated in the development of laparoscopic surgery in adults in the 1980s, has gained a prominent position in the care of pediatric patients because of its many advantages over more invasive traditional surgical approaches. Minimally invasive pediatric surgery began to take shape in the 1990s, with the adoption of laparoscopic techniques for simple surgeries such as appendectomy and cholecystectomy. An important milestone was set in 1995 when van der Zee performed the first laparoscopic CDH repair (1). Since then, it has seen a wide expansion in techniques and applications, ranging from gastrointestinal to urological and thoracic surgeries. After that, surgical techniques have undergone continuous improvements, up to unimaginable goals, such as performing robotic-assisted surgical procedures on patients under the age of one. Technological advances have played a crucial role in the evolution of minimally invasive pediatric surgery (2). Miniaturization of surgical instruments and improvements in imaging technologies have made operating safe even in the smallest patients possible. Robotics and augmented reality systems are now frequently integrated to improve the precision and effectiveness of surgery. Robotassisted surgery, in particular, has enabled a further decrease in incision size and an increase in precision, thanks to robotic arms that eliminate the natural tremor of the human hand and allow extremely controlled and delicate movements. The evolution of techniques and knowledge has made it possible to overcome barriers, operating with minimally invasive techniques on small patients with complex pathologies (3, 4). The clinical results of pediatric minimally invasive surgery have been extraordinary. Several studies have shown that children undergoing minimally invasive procedures experience less postoperative pain, have lower risks of infection, and enjoy faster convalescence than those treated with traditional techniques (5, 6). In addition, the reduced visual impact of scars contributes positively to young patients' psychological acceptance of surgical treatment.

The goal of this special issue was to collect publications that could best describe and summarize not only the evolution of minimally invasive pediatric surgery but also set goals for future developments.

Robot-assisted surgery has made it easier for pediatric surgeons to perform essential reconstructive-type procedures. One of the most significant examples is ureterovesical junction surgery, in which the robotic approach demonstrates its advantages. In their work, He et al. described how the technique of modified Lich-Gregoir direct nipple ureteral extravesical reimplantation can help maintain the physiological direction of the ureter and at the same time enhance the effectiveness of antireflux in robotic surgery. In addition, design of a single-port-plus-one wound can produce a cosmetic appearance by concentrating and hiding the wound around the umbilicus. This modified reimplantation procedure has the potential to become a promising technique in the robot-assisted treatment of primary obstructive megaureter.

Robot-assisted surgery is also catching on in the field of oncology surgery, in selected cases, showing the same good results as in other areas. Liang et al. described a case of a 3-yearold patient with a periampullary rhabdomyosarcoma who underwent a robotic pylorus-preserving pancreatoduodenectomy. With the assistance of a modern robotic system, they performed an R0 resection and a reconstruction with end-to-end pancreatojejunostomy, end-to-side hepaticojejunostomy, and duodenojejunostomy, without fatal complications such as pancreatic fistula or leak. The case reported demonstrates that also this kind of procedure in pediatric patients is safe and effective without intra- or postoperative complications.

An important part of minimally invasive surgery remains the domain of techniques such as laparoscopy and thoracoscopy. In this special issue, we have collected interesting experiences from this point of view. Laparoscopic herniorrhaphy in pediatric patients has become a routine procedure, often performed in day surgery. Zhang et al. reported a large series of 848 patients undergoing single-port laparoscopic herniorrhaphy. They described satisfactory results with no cases of conversion and 2 patients presenting with recurrence. They stated that this intervention presents numerous benefits, including the utilization of uncomplicated instruments, straightforward operation, a clear curative impact, minimal tissue damage, rapid recovery, and the absence of scarring. Another interesting example is that of Jung, who reports the case of a newborn presented with a rare combination of esophageal atresia with tracheoesophageal fistula and duodenal atresia, which was successfully managed using minimally invasive surgical techniques. The neonate underwent a thoracoscopic ligation of the tracheoesophageal fistula (TEF) and a laparoscopic duodeno-duodenostomy on the same day, resulting in stabilized vital signs. Ten days after the initial operation, a thoracoscopic esophago-esophagostomy was successfully performed. This report details a recent successful experience with a two-stage operation conducted without gastrostomy and utilizing minimally invasive surgical techniques. This approach underscores the evolving potential for neonatal treatment strategies in managing such complex cases.

Talking about minimally invasive surgery also means addressing how to enable surgeons, starting with residents, to acquire a progressive learning curve to make them autonomous even in the most complex cases. In this sense, the development of training programs involving faithful anatomical models becomes essential to ensure the development of the basic skills needed to approach this type of procedure. In this special issue, we have collected two original types of research dealing with different aspects of the same topic. Zahradnikova et al. created a 3D printed model for thoracoscopic repair of esophageal atresia with tracheoesophageal fistula and created a program in which 18 participants with different surgical experiences practiced. The results emphasized that this type of model proved useful as a training tool, partly because of its realism. The authors stated that due to its reusability, and suitability for individual participants, this model holds promise as a training tool for thoracoscopic procedures among surgeons. Miyano et al., on the other hand, described a cadaver teaching program to evaluate the usefulness of remote teaching. The results of their study show, that although inferior to hands-on practice, remote observation of minimally invasive procedures provides good results and can be particularly useful for training programs in resource-limited settings.

Lastly, when talking about pediatric minimally invasive surgery, and addressing the topic of what its developments might be, the focus on patients cannot be lost sight of. Pediatric patients require different management and attention than adults, and to neglect this is to disregard the overall health of patients. From this point of view, one of the main aspects is the management of the patient's preoperative anxiety. Franconi et al., designed a study, using a humanoid robot that would interact with patients, accompanying them as they entered the operating room. The purpose of this study was to determine whether the use of a humanoid robot could reduce preoperative anxiety levels in children. This work had the interesting result of showing that a non-pharmacological intervention like a humanoid robot reduces anxiety in children during the pre-operative time and it might be an attractive solution to optimize perioperative care in children.

In conclusion, we can say that the success of this Special Issue, in addition to the important number of articles collected, also lies in the fact that it has shown how minimally invasive surgery, even in pediatric settings, is a field where important milestones have been achieved, without ceasing to look toward future goals of further development.

#### Author contributions

EB: Writing – original draft, Writing – review & editing. GaL: Visualization, Writing – review & editing. GiL: Visualization, Writing – review & editing. GC: Supervision, Validation, Visualization, Writing – review & editing.

#### 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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated

#### References

1. van der Zee DC, Bax NM. Laparoscopic repair of congenital diaphragmatic hernia in a 6-month-old child. *Surg Endosc.* (1995) 9(9):1001–3. doi: 10.1007/ BF00188460

2. Esposito C, Masieri L, Castagnetti M, Pelizzo G, De Gennaro M, Lisi G, et al. Current status of pediatric robot-assisted surgery in Italy: epidemiologic national survey and future directions. *J Laparoendosc Adv Surg Tech A*. (2023) 33(6):610-4. doi: 10.1089/lap.2019.0516

3. Bindi E, Todesco C, Nino F, Torino G, Gentilucci G, Cobellis G. Robotic surgery: is there a possibility of increasing its application in pediatric settings? A single-center experience. *Children (Basel).* (2022) 9(7):1021. doi: 10.3390/children9071021

organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

4. Cobellis G, Bindi E. Pyeloplasty in children with ureteropelvic junction obstruction and associated kidney anomalies: can a robotic approach make surgery easier? *Children (Basel)*. (2023) 10(9):1448. doi: 10.3390/children10091448

5. Bindi E, Nino F, Bollettini T, Chiarella E, Cobellis G. Robot-assisted excision of partially obstructing ureteral fibroepithelial polyp in a child: a case report and review of the literature. J Pediatr Surg Case Rep. (2022) 84:102393. doi: 10.1016/j.epsc.2022.102393

6. Pierangeli F, Bindi E, Cruccetti A, Nino F, Gentilucci G, Cobellis G. Roboticassisted excision of a giant ureteral stump in a child: case report and nonsystematic review of the literature. *J Pediatr Surg Case Rep.* (2023) 93:102623. doi: 10.1016/j.epsc.2023.102623 Check for updates

#### **OPEN ACCESS**

EDITED BY Giuseppe Lauriti, G. d'Annunzio University of Chieti and Pescara, Italy REVIEWED BY

Francesco Morini, Meyer Children's Hospital, Italy Jun Fujishiro, The University of Tokyo, Japan

\*CORRESPONDENCE Feng Guo Suofeng@sdfmu.edu.cn

RECEIVED 05 May 2023 ACCEPTED 15 June 2023 PUBLISHED 27 June 2023

#### CITATION

Zhu L, Wang Y, Song H, Wang X, Zhang M and Guo F (2023) Case report: Minimally invasive removal of a dislodged thoracoamniotic shunt with an integral cystoscope in a preterm infant. Front. Pediatr. 11:1217667. doi: 10.3389/fped.2023.1217667

COPYRIGHT

© 2023 Zhu, Wang, Song, Wang, Zhang and Guo. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

#### Case report: Minimally invasive removal of a dislodged thoracoamniotic shunt with an integral cystoscope in a preterm infant

Lichao Zhu<sup>1,2</sup>, Yanze Wang<sup>2</sup>, Honghao Song<sup>1</sup>, Xiaoqing Wang<sup>2</sup>, Mingang Zhang<sup>2</sup> and Feng Guo<sup>1,2\*</sup>

<sup>1</sup>Department of Pediatric Surgery, Shandong Provincial Hospital, Shandong University, Jinan, China, <sup>2</sup>Department of Pediatric Surgery, Shandong Provincial Hospital Affiliated to Shandong First Medical University, Jinan, China

**Introduction:** Fetal pleural effusion is a rare condition that is associated with significant mortality. Although the insertion of fetal thoracoamniotic shunts can improve perinatal outcomes, there are several associated complications, such as intrathoracic dislodgement of the shunts. The optimal neonatal treatment for retained shunts remains uncertain.

**Case Description:** A male infant was born at 32 weeks of gestation. He had antenatal hydrothorax that was detected at 27 weeks of gestation and was managed by intrauterine thoracoamniotic shunting. However, the shunt catheter dislodged into the fetal chest, which caused reaccumulated pleural effusion and respiratory distress requiring ventilatory support after birth. After the patient's condition stabilized, minimally invasive removal of the retained catheter was performed on day 17 of life using an integral pediatric cystoscope via a 3-mm thoracic incision. The procedure took approximately 5 min. The postoperative course was uneventful, and the patient, who was discharged 39 days postnatally, is thriving at the 6-month follow-up.

**Conclusions:** We present a novel and effective approach to the management of an intrathoracic shunt using an integral cystoscope. This approach may offer a valuable alternative to traditional thoracoscopy in the neonatal period.

#### KEYWORDS

fetal pleural effusion, thoracoamniotic shunt dislodgement, cystoscope, thoracoscopy, case report

#### 1. Introduction

Fetal pleural effusion is a rare condition that occurs in 1 in 10,000-15,000 pregnancies. Massive pleural effusions can cause significant compression of the lungs and heart, leading to hemodynamic instability and nonimmune fetal hydrops, a potentially fatal condition if left untreated (1). Prenatal treatment typically involves the placement of fetal thoracoamniotic shunt(s) (TAS) that can significantly improve perinatal outcomes and survival rates in cases of large fetal pleural effusions (2–6). However, shunt dislodgement occurs in 5.4%–20% of cases (3, 6–9). Some previous studies suggested early elective removal of the shunt, primarily due to concerns regarding the risk of infection. In most cases, these shunts are extracted via thoracoscopy or a small skin incision (8, 10–12). Here, we detail

our experience of removing a dislodged TAS from the chest using an "all in one" pediatric cystoscope via a 3-mm thoracic incision within 5 min.

#### 2. Case description

A 31-year-old woman, gravida 5 para 3, was referred at 28 weeks of gestation after the detection of fetal pleural effusion during this pregnancy at 27 weeks. An ultrasound examination revealed left fetal pleural effusion with mediastinal shift to the right. Amniocentesis and ultrasound-guided intrauterine thoracocentesis were performed. However, the pleural effusion reaccumulated the following day. Under local anesthesia and ultrasound guidance, a TAS, which is a double-pigtail catheter, was placed without complications at 30 weeks of gestation (**Figure 1**). The ultrasound scan 24 h after the procedure showed resolution of both the pleural effusion reaccumulated once again and this time, the whole TAS was clearly detected within the fetal chest but the intra-amniotic segment was invisible, indicating intrathoracic migration (**Figure 2A**).

At 32 weeks of gestation, a male infant weighing 2,300 g was delivered vaginally following spontaneous onset of labor. The infant's respiratory condition was unstable, and chest x-ray revealed massive left pleural effusion with dislodgement of the thoracoamniotic shunt (Figure 2B). The preterm infant was intubated, and mechanical ventilation was started. Under ultrasound guidance, a 6 Fr pigtail chest tube was inserted into the left pleural cavity and initially drained 90 ml of pleural fluid.

After evaluating the patient's condition and the position of the shunt, which had migrated to the left upper thorax (Figure 2C), minimally invasive removal of the retained double-pigtail catheter was scheduled on day 17 postnatally. The patient was placed in the right lateral decubitus position, and general anesthesia was induced. A monitor was placed over the patient's head. Instead of traditional thoracoscopy that typically requires optic and operating ports, we opted for an integral cystoscope (circumference 7.9 Fr, Compact Peadiatric Cystoscope, Olympus) with a united telescope and 4.2 Fr working channel to minimize the incision and reduce injury to the chest wall. A 3-mm incision was made overlying the fifth intercostal space in the mid-axillary line, and the cystoscope was inserted directly into the pleural cavity without trocar insertion (Figure 3A). Carbon dioxide insufflation of 4 mmHg was achieved through one irrigation channel of the cystoscope to improve visibility. However, massive pleural effusion and fibrous adhesions hampered identification of the TAS. We then gently performed blunt adhesiolysis using the rigid cystoscope as a thoracoscopic instrument and aspirated pleural effusion through another irrigation channel of the cystoscope. After careful inspection of the thoracic cavity, we located the shunt catheter behind the left upper lobe and extracted it using an endoscopic grasping forceps inserted through the 4.2 Fr working channel of the cystoscope (Figure 3B).





#### FIGURE 2

Prenatal ultrasound screening and postnatal chest radiography. (A) The ultrasound examination identified reaccumulated pleural effusion and clearly visualized the entire double-pigtail shunt (red arrows) located within the fetal chest, while the intra-amniotic segment remained invisible. (B) The chest x-ray displayed a massive left pleural effusion with dislodgement of the thoracoamniotic shunt. (C) The chest x-ray showed that the dislodged shunt had migrated to the left upper thorax.

The cystoscope was then removed, and a 10 Fr chest drainage tube was placed at the same incision. The operative time from insertion to removal of the cystoscope was approximately 5 min (Supplementary Video S1). The postoperative course was uneventful. The ultrasound scan confirmed no re-accumulation of the left pleural effusion, and the chest drains were removed on postoperative day 12. The patient, who was discharged 39 days postnatally, is thriving at the 6-month follow-up.

# including preterm birth, fetal constriction bands of the limbs, rib fractures, traumatic hemothorax, and shunt obstruction (3, 6, 9, 13, 14). Shunt dislodgement into the amniotic fluid, maternal peritoneal cavity, or fetal chest has also been reported in up to 20% of cases (3, 6–9). The mechanisms of shunt migration are not fully understood, but may include chest wall growth, respiratory movement, resolution of skin edema as hydrops improves, and technical difficulties encountered with hydropic fetuses or shunt insertion by less experienced operators (7, 15).

#### 3. Discussion

Although the insertion of fetal TAS has been shown to improve fetal outcomes, there are several associated complications,

The management of shunts that have migrated into the thoracic cavity presents a dilemma, and the optimal neonatal treatment remains uncertain. Some studies have advocated for a conservative approach to the management of intrathoracic fetal chest shunts without surgical removal, as their follow-up did not reveal



#### FIGURE 3

Operative images. (A) The cystoscope, which has a 4.2 Fr straight channel (red arrow) and two irrigation channels (yellow arrowhead), was inserted into the pleural cavity via a 3-mm incision without trocar insertion. (B) An endoscopic grasping forceps was inserted through the 4.2 Fr channel and grasped the shunt.

significant complications (9, 15). However, others have recommended early elective removal to minimize the risks of infection, the shunt's proximity to mediastinal structures, and fibrous change that may complicate shunt removal (8, 10–12). There was a case reported by Blanch et al. where an abnormal position of a retained TAS led to strangulation of the pulmonary hilum, resulting in neonatal death (16). Therefore, we should be vigilant of the catastrophic consequences of dislodged shunts and recognize the need for early surgical intervention, especially when a retained shunt is located in the mediastinum near the lung hilum (12).

Regarding the ideal timing of intervention for intrathoracic dislodged shunts, most authors suggest removing them sooner rather than later, once the patient's condition has stabilized (8, 10-12). A similar strategy was applied in our patient, in which the operation was performed on day 17 of life for initial stabilization. Compared with thoracotomy, thoracoscopic removal of retained TAS has several benefits, such as smaller incisions, clearer visualization of the thoracic cavity, less wound pain, fewer major wound complications, and faster recovery (11, 12). However, traditional thoracoscopy necessitates at least two incisions, measured 3-5 mm in length, for a camera port and a working port. Muta et al. reported their experience of removing TAS catheters with a 2.7-mm scope inserted into a 5-mm trocar to observe the thoracic cavity (10). Once the catheter was identified, a 3-mm forceps was inserted into the 5-mm trocar from the side of the scope to grasp the catheter. They believed that thoracoscopic removal with intraoperative radiography can help reduce their operation time, which averages  $35.25 \pm 30.49$  min. Macchini et al. reported two newborns with intrathoracic dislodgement of TAS, where they inserted two 3-mm trocars and performed thoracoscopic removal of the shunts, with an operation time of 30 and 35 min, respectively (12). In our case, we confirmed the position of the dislodged TAS through preoperative x-ray. Intraoperative radiography is not necessary to perform routinely, as it may not provide more valuable information about the shunt location. We then chose an integral cystoscope, a one-piece instrument with a combined telescope, sheath, and working channels. The Olympus compact paediatric cystoscope (Product Number: A37026A), equipped with a 7-degree direction of view, a 4.2 Fr straight channel, and two irrigation channels, was the preferred choice. Its small outer diameter of 7.9 Fr makes one 3mm incision adequate. Two irrigation channels can serve for carbon dioxide insufflation and pleural effusion suction. To locate the shunt, a rigid outer tube of the cystoscope can serve as a thoracoscopic instrument to lyse adhesions by the blunt technique under close monitoring. Once the shunt is identified, the 4.2 Fr straight channel of the cystoscope allows for the passage of an endoscopic forceps to grasp the catheter. An integral cystoscope is thus deemed valuable for removal of dislodged TAS. Additionally, pediatric rigid bronchoscopes, with diameters of 3-7 mm and lengths of 20-50 cm (17), may serve as a viable alternative for removing the dislodged TAS. However, at our institution, bronchoscopy is typically performed by pulmonologists and we do not have bronchoscopes readily available in the operating room. Therefore, we opted to use a cystoscope instead of a bronchoscope to remove the dislodged TAS.

As previously reported, severe intrathoracic adhesions can make it challenging to locate dislodged TAS in certain cases (10). Although we did not encounter this situation, the use of fluoroscopic radiography during surgery to detect the position of the TAS catheter could be beneficial, as described by Muta et al. These authors suggest that thoracoscopic removal with fluoroscopic radiography can help to reduce the operation time and wound length (10).

#### 4. Conclusions

We present a novel approach to management of intrathoracic TAS with an integral cystoscope in the neonatal period. This procedure is safe, effective, and minimally invasive, and has the potential to improve clinical outcomes and reduce mortality. This integral cystoscope may offer a valuable alternative to traditional thoracoscope for patients with dislodged TAS.

#### 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**

Written informed consent was obtained from the minor(s)' legal guardian/next of kin for the publication of any potentially identifiable images or data included in this article.

#### Author contributions

FG contributed to the design of the case presentation, critically reviewed and revised the manuscript. LZ and FG contributed to the design of the case presentation, drafted the initial manuscript. All authors contributed to the article and approved the submitted version.

#### Funding

This study was supported by the Youth Science Foundation of Shandong First Medical University (grant number 202201-066) for XW and the Science and Technology Development Plan Project of Shandong Province, China (grant number 2009GG20002036) for MZ.

#### Acknowledgments

We would like to thank the patient and his parents for their support and cooperation in publishing this study.

#### 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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or

#### References

1. Longaker MT, Laberge JM, Dansereau J, Langer JC, Crombleholme TM, Callen PW, et al. Primary fetal hydrothorax: natural history and management. *J Pediatr Surg.* (1989) 24(6):573–6. doi: 10.1016/s0022-3468(89)80509-3

2. Wada S, Jwa SC, Yumoto Y, Takahashi Y, Ishii K, Usui N, et al. The prognostic factors and outcomes of primary fetal hydrothorax with the effects of fetal intervention. *Prenat Diagn*. (2017) 37(2):184–92. doi: 10.1002/pd.4989

3. Peranteau WH, Adzick NS, Boelig MM, Flake AW, Hedrick HL, Howell LJ, et al. Thoracoamniotic shunts for the management of fetal lung lesions and pleural effusions: a single-institution review and predictors of survival in 75 cases. *J Pediatr Surg.* (2015) 50(2):301–5. doi: 10.1016/j.jpedsurg.2014.11.019

4. Jeong BD, Won HS, Lee MY, Shim JY, Lee PR, Kim A. Perinatal outcomes of fetal pleural effusion following thoracoamniotic shunting. *Prenat Diagn*. (2015) 35 (13):1365–70. doi: 10.1002/pd.4709

5. Pellegrinelli JM, Kohler A, Kohler M, Weingertner AS, Favre R. Prenatal management and thoracoamniotic shunting in primary fetal pleural effusions: a single centre experience. *Prenat Diagn*. (2012) 32(5):467-71. doi: 10.1002/pd. 3840

6. Yinon Y, Grisaru-Granovsky S, Chaddha V, Windrim R, Seaward PG, Kelly EN, et al. Perinatal outcome following fetal chest shunt insertion for pleural effusion. *Ultrasound Obstet Gynecol.* (2010) 36(1):58–64. doi: 10.1002/uog.7507

7. Abbasi N, Windrim R, Keunen J, Seaward PGR, Van Mieghem T, Kelly EN, et al. Perinatal outcome in fetuses with dislodged thoraco-amniotic shunts. *Fetal Diagn Ther.* (2021) 48(6):430–9. doi: 10.1159/000515694

8. Inoue S, Odaka A, Baba K, Kunikata T, Sobajima H, Tamura M. Thoracoscopyassisted removal of a thoracoamniotic shunt double-basket catheter dislodged into the fetal thoracic cavity: report of three cases. *Surg Today.* (2014) 44(4):761–6. doi: 10. 1007/s00595-013-0565-x claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

#### Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fped.2023. 1217667/full#supplementary-material.

Video 1.

An unedited video showing the entire procedure from the insertion to the removal of the cystoscope.

9. Sepulveda W, Galindo A, Sosa A, Diaz L, Flores X, de la Fuente P. Intrathoracic dislodgement of pleuro-amniotic shunt. Three case reports with long -term follow-up. *Fetal Diagn Ther.* (2005) 20(2):102–5. doi: 10.1159/000082431

10. Muta Y, Odaka A, Inoue S, Takeuchi Y, Beck Y. Thoracoscopic removal with fluoroscopic radiographic guidance of thoracoamniotic shunting catheters in newborns. *Surg Today.* (2022) 52(10):1504–8. doi: 10.1007/s00595-022-02535-2

11. Sham GTW, Chung PHY, Chan IMC, Leung WC, Wong KKY. Thoracoscopic removal of a displaced thoracoamniotic shunt in a newborn with antenatal pleural effusion-a case report. *Transl Pediatr.* (2020) 9(5):702–6.doi: 10.21037/tp-20-74

12. Macchini F, Gentilino V, Morandi A, Leva E. Thoracoscopic removal of retained thoracoamniotic shunt catheters in newborns. *J Laparoendosc Adv Surg Tech A*. (2014) 24(11):827–9. doi: 10.1089/lap.2014.0133

13. Merchant AM, Peranteau W, Wilson RD, Johnson MP, Bebbington MW, Hedrick HL, et al. Postnatal chest wall deformities after fetal thoracoamniotic shunting for congenital cystic adenomatoid malformation. *Fetal Diagn Ther.* (2007) 22(6):435–9. doi: 10.1159/000106350

14. Brown R, Nicolaides K. Constriction band of the arm following insertion of a pleuro-amniotic shunt. *Ultrasound Obstet Gynecol.* (2000) 15(5):439–40. doi: 10. 1046/j.1469-0705.2000.00114.x

15. Alkazaleh F, Saleem M, Badran E. Intrathoracic displacement of pleuroamniotic shunt after successful in utero treatment of fetal hydrops secondary to hydrothorax. Case report and review of the literature. *Fetal Diagn Ther.* (2009) 25(1):40–3. doi: 10.1159/000196374

16. Blanch G, Walkinshaw SA, Hawdon JM, Weindling AM, van Velzen D, Rodeck CH. Internalization of pleuroamniotic shunt causing neonatal demise. *Fetal Diagn Ther.* (1996) 11(1):32–6. doi: 10.1159/000264276

17. Soyer T. The role bronchoscopy in the diagnosis of airway disease in children. J Thorac Dis. (2016) 8(11):3420–6.doi: 10.21037/jtd.2016.11.87 Check for updates

#### **OPEN ACCESS**

EDITED BY Edoardo Bindi, Salesi Hospital Foundation, Italy

REVIEWED BY daniel Robert schlatterer, WellStar Health System, United States Gergo Jozsa, University of Pécs, Hungary

\*CORRESPONDENCE Gang Lin Image: Second Secon

RECEIVED 09 May 2023 ACCEPTED 05 July 2023 PUBLISHED 17 July 2023

#### CITATION

Ju L, Xu M and Lin G (2023) Case report: Pediatric floating elbow fracture with monteggia-equivalent lesion, ipsilateral humeral shaft fracture, and radial nerve injury: a unique case and favorable treatment outcomes. Front. Pediatr. 11:1219518. doi: 10.3389/fped.2023.1219518

#### COPYRIGHT

© 2023 Ju, Xu and Lin. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

#### Case report: Pediatric floating elbow fracture with monteggia-equivalent lesion, ipsilateral humeral shaft fracture, and radial nerve injury: a unique case and favorable treatment outcomes

#### Li Ju<sup>1</sup>, Mengqiu Xu<sup>2</sup> and Gang Lin<sup>1\*</sup>

<sup>1</sup>Department of Pediatric Orthopedics, Children's Hospital of Nanjing Medical University, Nanjing, China, <sup>2</sup>Nanjing Medical University, Nanjing, China

This case report presents a rare and intricate pediatric floating elbow fracture involving a Monteggia-equivalent fracture, ipsilateral humeral shaft fracture, and radial nerve injury. The unique mechanism of injury highlights the importance of increased awareness and parental education for accident prevention. Elastic intramedullary nailing was employed for both humeral shaft and forearm fractures, leading to favorable outcomes. Despite the severity of the fractures and radial nerve injury, the prognosis was positive, with nerve function restoration and satisfactory functional recovery. However, the development of avascular necrosis of the radial head remains a challenge, emphasizing the need for further research to better understand and manage these uncommon and complex injuries.

#### KEYWORDS

pediatric floating elbow, monteggia-Equivalent lesion, radial nerve injury, avascular necrosis of the radial head, elastic intramedullary nailing

#### Introduction

Floating elbow fractures in the pediatric population constitute a scarce injury encompassing a simultaneous humeral fracture and ipsilateral forearm fracture. Predominantly, research targets supracondylar humeral fractures in conjunction with forearm fractures (1). Pediatric humeral shaft fractures represent a mere 1%–3% of all fractures in children and less than 10% of all humeral fractures within this demographic (2). Monteggia fractures, severe elbow joint fractures in pediatric patients, pose considerable management challenges and are frequently accompanied by complications. Jose Luis Bado's 1962 classification system continues to be extensively utilized for Monteggia fracture categorization (3). Nonetheless, numerous complex fractures evade classification within this system, and are subsequently designated as Monteggia-equivalent fractures. The heterogeneous nature of these fractures renders treatment arduous, and a unified classification method remains elusive.

In this case report, we describe a unique floating elbow fracture in a child, involving an exceptionally complex injury with a concomitant Monteggia-equivalent fracture, ipsilateral humeral shaft fracture, and radial nerve injury. To our knowledge, this is the first

reported case of such a complex injury, with no existing literature on its management. Despite the challenges, appropriate management led to favorable outcomes, offering valuable insights into managing this rare and complex condition.

#### Case report

We present the case of a 2-year and 11-month-old boy who sustained high-energy injuries from a unique traffic accident. Physical examination revealed no open wounds but significant upper limb swelling. Vascular and neurological examination showed normal radial artery pulsation and symptoms of radial nerve injury, characterized by limited thumb extension. x-ray images revealed a right humeral shaft fracture, right ulnar and radial shaft fractures, and a lateral dislocation of the radius (Bado Type IV Monteggia fracture). Closer examination identified a radial neck fracture. Computed tomography (CT) scans confirmed four fractures in the right upper limb (Figure 1).

The patient was admitted to the Surgical Intensive Care Unit (SICU) following the injury. After ensuring the patient regained consciousness and receiving confirmation from the neurosurgical team that his condition was stable, surgery was performed under general anesthesia on the third day. Humeral shaft and ulnar and radial shaft fractures were fixed with closed reduction and elastic intramedullary nailing. Reduction of the radial head fracture (Monteggia-equivalent lesion) proved challenging. The Kocher approach was employed to expose and successfully reduce the radial head. An elastic intramedullary nail used to fix the radial shaft also supported the radial head. Notably, nerve exploration was not conducted during surgery. The patient was immobilized with a plaster sling and monitored for complications, such as compartment syndrome. At the four-week postoperative follow-up, the plaster sling was removed, and functional exercises began. Radial nerve function was restored two months after surgery. Six months later, internal fixation was removed (Figure 2). Follow-up evaluations 1.5 years post-surgery revealed largely restored upper limb function, with normal flexion and extension movements but some limitation in rotation (Figure 3).

#### Discussion

Stanitski and Micheli introduced the term "floating elbow" to describe a forearm and humerus fracture in the same limb (4). Pediatric humeral shaft fracture incidence rates range from 12 to 30 cases per 100,000 individuals, accounting for approximately 20% of all humeral fractures in children (2). Monteggia fracture equivalent incidence rates remain undetermined due to subtype variety and overall rarity. Our case report presents a rare Monteggia equivalent lesion (PMEL) involving a double forearm fracture and a radial neck fracture, which is prone to misdiagnosis as a radial head dislocation on x-rays. PMEL is often overlooked and mismanaged due to its unclear definition and diverse presentation. Xu et al. suggest defining PMEL as an ulnar fracture at any level combined with a proximal radial fracture (5). They classify PMEL into three groups based on the radiocapitellar joint status: stable, unstable, and dislocated. However, our case does not fit into any of these groups due to the unusual forearm bone fracture pattern. The combination of humeral shaft fractures and Monteggia fracture equivalents in the same limb presents a unique challenge to clinicians, emphasizing the need for further research to better understand the clinical presentation, treatment, and long-term outcomes of these complex injuries.





FIGURE 2

x-ray images of the child one day after surgery (A,B); x-ray images of the child after one year, following the removal of the internal fixation, showing avascular necrosis of the radial head (C,D)

The mechanism of injury in this case is unusual, as it did not involve a fall from a height, a sports-related injury, or a typical traffic accident. The incident occurred when the child accidentally activated an unlocked electric scooter by twisting the handlebar without adult supervision, causing sudden acceleration. This resulted in the child being carried along for several meters, ultimately leading to severe fractures. Our case is similar to a report wherein a child sustained a comparably severe floating elbow fracture after inserting his hand into a washing machine (6). Based on our radiographic findings, the primary cause of such severe fractures was a forceful twisting motion, which could lead to multiple upper limb fractures, nerve damage, and even potential avulsion injuries in the soft tissue. Fortunately, our case did not involve severe soft tissue injuries, and the patient had a good postoperative recovery. This incident emphasizes the imperative to augment awareness regarding such safety hazards and necessitates the reinforcement of parental education strategies to mitigate the prevalence of analogous accidents.

Metaizeau's introduction of elastic intramedullary nailing in the 1980s made it one of the standard methods for treating pediatric long bone fractures (7). The flexible intramedullary nailing (FIN) system is commonly used to treat humeral shaft fractures, stabilizing the fracture by applying a three-point balanced force within the medullary cavity (2, 8). Similarly, the elastic intramedullary nail has been widely recognized for treating forearm fractures due to its advantages, including minimal trauma, simple operation, preservation of blood supply, fewer complications, and faster recovery (2, 9). Our case presented a significant challenge, as the patient had two fractures in the radius. We performed a closed reduction of the midshaft radius fracture during surgery, but the proximal radius fracture was a Monteggia equivalent lesion and was displaced, with the distal fragment inserted into the muscle. Therefore, an open reduction was required. Fixation after reduction was also challenging. While some authors have used Kirschner wires, screws, or plates for fixation (10), we opted to use a single intramedullary nail to fix both the midshaft radius fracture and the radial head fracture, considering the age of our patient who was under 3 years old. This approach minimized disruption to the blood supply of the radial head and avoided damaging the growth plate of the radial head and potential needle tract infections.



Radial nerve injury is the most common nerve injury associated with humeral shaft fractures, with an incidence rate of 7% to 17% in adults (11). However, the incidence rate is significantly lower in children and has a better prognosis. In our case, radial nerve injury was also present, but the main manifestation was damage to the posterior interosseous nerve (PIN), evidenced by restricted extension of the thumb and no wrist drop. Literature reports suggest that the incidence of PIN injury in Monteggia fractures is 3.1% to 31.4%, making it the most common complication of this type of fracture (12). A retrospective study of a series of pediatric Monteggia fracture cases recommended waiting for six months after the radial nerve injury before intervention (13). Most research series report that nerve injuries after floating elbow injury are resolved (14). In our case, despite severe injury to the posterior interosseous nerve (PIN), we did not perform any special management for the radial nerve injury. As is common with most supracondylar fractures with associated nerve injuries, we did not employ surgical exploration or electrostimulation, especially considering the presence of implants (metal) in the area. Fortunately, the child's radial nerve function showed signs of recovery approximately two months after the surgery. This favorable outcome aligns with the findings reported in Baghdadi's systematic review. It is worth noting that the recovery of nerve function in pediatric cases can vary, and in our case, the restoration of radial nerve function occurred at around three months post-operation.

10.3389/fped.2023.1219518

During the 1.5-year follow-up after surgery, our case developed avascular necrosis of the radial head. D'Souza et al. reported a higher incidence of radial head avascular necrosis than previously thought, with a frequency of 10% to 20% in their patients, of which approximately 70% underwent open reduction surgery before the onset of avascular necrosis (15). In patients who underwent open reduction, the overall incidence rate of radial head avascular necrosis was 25%. Jones and Esah, as well as Newman, found that patients with radial head avascular necrosis had poorer functional recovery (16, 17). Our case was consistent with these findings, where the child underwent open reduction surgery for the radial head and had normal flexion-extension function but significant limitation in rotation function 1.5 years after the operation. Despite the presence of three fractures in the child's forearm, favorable healing was observed post-surgery. Consequently, it is posited that the restricted rotational functionality of the child's forearm is primarily attributable to the avascular necrosis of the radial head. Nevertheless, given the patient's young age, long-term monitoring is warranted to ascertain the potential for revascularization.

#### Conclusion

This case report highlights a unique and complex floating elbow fracture featuring a Monteggia-equivalent fracture, ipsilateral humeral shaft fracture, and associated radial nerve injury. The case emphasizes diagnostic and management challenges, calling for further research to better understand injury mechanisms, clinical presentations, treatments, and longterm outcomes. Our findings indicate that elastic intramedullary nailing for both humeral shaft and forearm fractures may lead to satisfactory results. Additionally, despite severe fractures, the prognosis for accompanying nerve injuries is generally positive, with immediate surgical intervention potentially unnecessary. However, addressing avascular necrosis of the radial head remains a significant challenge, especially in pediatric patients,

#### References

1. Malheiros DS, Barbara GH, Mafalda LG, Madureira JL Jr., Braga GF, Terra DL. Floating elbow in children: a descriptive study of 31 cases attended in a reference center for pediatric trauma. *Rev Bras Ortop.* (2011) 46(5):500–4. doi: 10.1016/S2255-4971(15)30403-1

2. Wiktor Ł, Tomaszewski R. Humeral shaft fractures in children incidence, management and treatment effects. *Ortop Traumatol Rehabil.* (2022) 24(4):251-61. doi: 10.5604/01.3001.0015.9986

3. Bado JL. The monteggia lesion. Clin Orthop Relat Res. (1967) 50:71-86.

4. Stanitski CL, Micheli LJ. Simultaneous ipsilateral fractures of the arm and forearm in children. *Clin Orthop Relat Res.* (1980) 153:218–22. doi: 10.1097/00003086-198011000-00031

5. Xu L, Ye W, Li H, Xu J, Zhu W, Zhen Z, et al. Monteggia equivalent lesion in children: a narrative review. *World J Pediatr Surg.* (2021) 4(3):e000283. doi: 10. 1136/wjps-2021-000283

6. Albattat MY, Alhathloul H, Almohammed Saleh M, Althabit F. Pediatric floating elbow caused by a novel mechanism: a case report. *Cureus*. (2022) 14(9):e29124. doi: 10.7759/cureus.29124

due to limited functional exercise efficacy, necessitating the development of more effective approaches.

#### 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/s.

#### Ethics statement

Written informed consent was obtained from the minor(s)' legal guardian/next of kin for the publication of any potentially identifiable images or data included in this article.

#### Author contributions

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.

#### Publisher's note

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

7. Metaizeau JP, Ligier JN. Surgical treatment of fractures of the long bones in children. Interference between osteosynthesis and the physiological processes of consolidation. Therapeutic indications. *J Chir (Paris)*. (1984) 121 (8-9):527-37.

8. Kelly DM. Flexible intramedullary nailing of pediatric humeral fractures: indications, techniques, and tips. *J Pediatr Orthop.* (2016) 36(Suppl 1):S49–55. doi: 10.1097/bp0.00000000000755

9. Khuntia S, Swaroop S, Patro BP, Sahu S. Paediatric long bone fractures managed with elastic intramedullary nails: a retrospective study of 30 patients. *Cureus*. (2020) 12(4):e7847. doi: 10.7759/cureus.7847

10. Waters PM, Stewart SL. Radial neck fracture nonunion in children. J Pediatr Orthop. (2001) 21(5):570-6.

11. Schwab TR, Stillhard PF, Schibli S, Furrer M, Sommer C. Radial nerve palsy in humeral shaft fractures with internal fixation: analysis of management and outcome. *Eur J Trauma Emerg Surg.* (2018) 44(2):235–43. doi: 10.1007/s00068-017-0775-9 12. Li H, Cai QX, Shen PQ, Chen T, Zhang ZM, Zhao L. Posterior interosseous nerve entrapment after monteggia fracture-dislocation in children. *Chin J Traumatol.* (2013) 16(3):131–5.

13. Olney BW, Menelaus MB. Monteggia and equivalent lesions in childhood. J Pediatr Orthop. (1989) 9(2):219-23. doi: 10.1097/01241398-198903000-00021

14. Baghdadi S. Pediatric floating elbow injuries are not as problematic as they were once thought to be: a systematic review. *J Pediatr Orthop*. (2020) 40(8):380–6. doi: 10. 1097/bpo.000000000001573

15. Dougall AJ. Severe fracture of the neck of the radius in children. J R Coll Surg Edinb. (1969) 14(4):220–5.

16. Jeffery CC. Fractures of the head of the radius in children. J Bone Joint Surg Br. (1950) 32-b(3):314–24. doi: 10.1302/0301-620x.32b3.314

17. Nenopoulos SP, Beslikas TA, Gigis JP. Long-term follow-up of combined fractures of the proximal radius and ulna during childhood. *J Pediatr Orthop B.* (2009) 18(5):252–60. doi: 10.1097/bpb.0b013e32 83153f7e

Check for updates

#### **OPEN ACCESS**

EDITED BY Giovanni Cobellis, Marche Polytechnic University, Italy

REVIEWED BY Edoardo Bindi, Salesi Hospital Foundation, Italy Peter Vasas, Doncaster Royal Infirmary, United Kingdom

\*CORRESPONDENCE Rossella Palma Image: rossellapalma89@gmail.com Francesco Angrisani Image: chiccoangri@gmail.com

RECEIVED 23 May 2023 ACCEPTED 27 June 2023 PUBLISHED 21 July 2023

#### CITATION

Palma R, Angrisani F, Santonicola A, Iovino P, Ormando VM, Maselli R and Angrisani L (2023) Case report: Laparoscopic nissen-sleeve gastrectomy in a young adult with incidental finding of Morgagni-Larrey hernia. Front. Surg. 10:1227567.

doi: 10.3389/fsurg.2023.1227567

#### COPYRIGHT

© 2023 Palma, Angrisani, Santonicola, Iovino, Ormando, Maselli and Angrisani. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

#### Case report: Laparoscopic nissen-sleeve gastrectomy in a young adult with incidental finding of Morgagni-Larrey hernia

Rossella Palma<sup>1\*</sup>, Francesco Angrisani<sup>2\*</sup>, Antonella Santonicola<sup>3</sup>, Paola Iovino<sup>3</sup>, Vittorio Maria Ormando<sup>4</sup>, Roberta Maselli<sup>5,6</sup> and Luigi Angrisani<sup>7</sup>

<sup>1</sup>Department of Surgical Sciences, "Sapienza" University of Rome, Rome, Italy, <sup>2</sup>Humanitas University, Milan, Italy, <sup>3</sup>Department of Medicine, Surgery and Dentistry "Scuola Medica Salernitana", University of Salerno, Salerno, Italy, <sup>4</sup>Gastroenterology and Endoscopy Unit, AORN San Giuseppe Moscati, Avellino, Italy, <sup>5</sup>Endoscopy Unit, Humanitas Clinical and Research Center IRCCS, Rozzano, Italy, <sup>6</sup>Department of Biomedical Sciences, Humanitas University, Milan, Italy, <sup>7</sup>Department of Public Health, "Federico II" University of Naples, Italy

Laparoscopic sleeve Gastrectomy (LSG) is the most performed bariatric procedure worldwide but it is associated with an increased incidence of de-novo or recurrent GERD. Recently a new technique consisting in LSG with associated fundoplication has been described. Morgagni-Larrey hernia is very rare and there is a lack of evidences on its correct surgical treatment. There are only rare cases of a MLH incidental diagnosis in patients submitted to bariatric surgery. We present our experience of Morgagni-Larrey Hernia (MLH) incidentally found intraoperatively in a patient with Gastroesophageal Reflux Disease (GERD) with Hiatal Hernia (HH) undergoing a bariatric surgical procedure.

#### KEYWORDS

obesity, diaphragmatic hernia, Morgagni-Larrey hernia, hiatal hernia, sleeve gastrectomy

#### Introduction

Laparoscopic sleeve Gastrectomy (LSG) is the most performed bariatric procedure worldwide (1). It is known that LSG is associated with an increased incidence of de-novo or recurrent GERD (2) and Barrett's Esophagus (BE) (3). Roux-en-Y Gastric Bypass (RYGB) is considered the first choice in patients affected by GERD and Hiatal Hernia. Several studies investigated the effectiveness of LSG with associated HH repair (HHR), although the long-term results are contradictory and HH and GERD symptoms recurrence have been described (4, 5).

Recently a new technique consisting in LSG with associated fundoplication has been described (6). To the best of our knowledge, there are only few anecdotal cases of intraoperative finding and concomitant repair of a Morgagni-Larrey diaphragmatic hernia during a bariatric procedure (7, 8).

#### **Case description**

In our experience, a 18-year-old male patient with grade III obesity ( $BMI = 40.76 \text{ Kg/m}^2$ ) and GERD symptoms was referred to our center to receive bariatric surgery. The patient previously placed an intragastric balloon with an initial weight loss of 30 kg, however by

two years more than 80% of lost weight was regained. The preoperative upper gastrointestinal endoscopy (UGIE) showed a three centimeters grade 4 HH according to Hill Classification. HH was confirmed on X-ray esophagus and no other defects were preoperatively diagnosed. The patient presented typical GERD symptoms that severely impacted his quality of life. We decided to perform a LSG with associated Nissen fundoplication and HHR.

During surgery a defect in the anterior, parasternal portion of the diaphragm was immediately recognized (Figure 1). The diaphragmatic hernia sac appeared free of content and the choice was to perform the planned bariatric procedure without repairing the defect (Figure 2). No perioperative complications were recorded and the patient was discharged 2 days later. At the 6 months follow up visit, the BMI was  $31.14 \text{ kg/m}^2$  with a TWL of 23.6%. Furthermore, the patient didn't report GERD symptoms, dyspepsia, dyspnea and abdominal or chest pain.

#### Discussion

Morgagni-Larrey hernia (MLH) represents the most common congenital defect in the anterior, parasternal portion of the diaphragm. This hernia is very rare with a reported incidence ranging from 3% to 5% of all diaphragmatic hernias and it is most frequently diagnosed in infants and children. However, the hernia can remain undiagnosed until adulthood, due to a vague clinical presentation (9). The patients' symptoms are generally



#### FIGURE 1 Intraoperative finding of a Morgagni-Larrey hernia.



unspecific and range from dyspnea to abdominal or chest pain and constipation. However, these are mostly dependent on hernia sac content with small or large intestine, as well as on the hernia size with compressive symptoms such as dyspnea, fatigue or exercise intolerance.

The exact prevalence of MLH in adult patients remains unclear and the knowledge about diagnostic methods, imaging techniques and treatment modalities is only based on single surgeon's experiences and anecdotal reports. In our clinical practice, over the last thirty years, only one case of pre-operative finding of Morgagni-Larrey hernia was found in a lean patient with epigastric pain and subocclusive symptoms (10) which underwent hernia reduction and direct suture repair. No other similar cases have been observed in our bariatric experience.

Although several studies report the successful repair of MLH in infants. According to a recent review with meta-analysis the recurrence and complication rates are comparable between mini invasive approaches and open repairs in patients with mean age of 17-months. Anyway the use of patch appeared to confer additional benefit in reducing recurrence (11). Recently has been also described the laparoscopy-assisted transabdominal repair of MLH using loop suture, with successful results. Leaving the hernia sac apparently does not increase the recurrence rate (12).

The indication and operative strategies of Morgagni-Larrey hernia in adults remain controversial. In a large retrospective single-center study on adult patients with MLH, surgery was performed safely with mesh reinforcement after primary closure (13).

The possible described surgical treatment options of an extrahiatal diaphragmatic hernia were:

- primary suture
- primary suture with mesh reinforcement
- mesh interposition without primary closure

In conclusion the evidences about the diagnostic pathway and surgical options in adult patients are very low, especially when incidentally discovered during a bariatric procedure. The symptoms are generally unspecific and could be overlapping to other clinical conditions, especially in case of a concomitant hiatal hernia in an obese patient.

Although MLH could be safely repaired with a laparoscopic access taking into account the surgeon experience and patient's symptoms, in our case the extra-hiatal hernia was incidentally diagnosed. The evidence of an empty sac, the lack of specific symptoms associated with the MLH, and the absence of a

#### References

1. Angrisani L, Santonicola A, Iovino P, Ramos A, Shikora S, Kow L. Bariatric surgery survey 2018: similarities and disparities among the 5 IFSO chapters. *Obes Surg.* (2021) 31(5):1937–48. doi: 10.1007/s11695-020-05207-7

2. Himpens J, Dobbeleir J, Peeters G. Long-term results of laparoscopic sleeve gastrectomy for obesity. *Ann Surg.* (2010) 252(2):319–24. doi: 10.1097/SLA. 0b013e3181e90b31

3. Genco A, Soricelli E, Casella G, Maselli R, Castagneto-Gissey L, Di Lorenzo L, et al. Gastroesophageal reflux disease and Barrett's esophagus after laparoscopic sleeve gastrectomy: a possible, underestimated long-term complication. *Surg Obes Relat Dis.* (2017) 13(4):568–74. doi: 10.1016/j.soard.2016.11.029 specific surgical informed consent (being found incidentally) have led the surgeon to prefer a conservative approach, leaving intact the MLH, to give priority to the treatment of obesity and GERD.

#### Data availability statement

The data that support the finding of this study are available from the corresponding author upon reasonable request.

#### **Ethics statement**

Written informed consent was obtained from the individual(s) for the publication of any identifiable images or data included in this article.

#### Author contributions

Conceptualization, RP, FA, LA, PI, AS, RM; writing—original draft, RP, FA VO; writing—review & editing, RP, FA, LA, AS, PI, VO and RM. All authors have read and agreed to the published version of the manuscript. 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.

#### Publisher's note

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

 Lewis K, Callaway K, Argetsinger S, Wallace J, Arterburn DE, Zhang F, et al. Concurrent hiatal hernia repair and bariatric surgery: outcomes after sleeve gastrectomy and roux-en-Y gastric bypass. *Surg Obes Relat Dis.* (2021) 17(1):72–80. doi: 10.1016/j.soard.2020.08.035

5. Angrisani L, Santonicola A, Borrelli V, Iovino P. Sleeve gastrectomy with concomitant hiatal hernia repair in obese patients: long-term results on gastroesophageal reflux disease. *Surg Obes Relat Dis.* (2020) 16(9):1171–7. doi: 10. 1016/j.soard.2020.04.049

6. Nocca D, Galtier F, Taleb S, Picot MC, Jaussent A, Silvestri M, et al. Peri-operative morbidity of nissen sleeve gastrectomy: prospective evaluation of a cohort of 365

patients, beyond the learning curve. Obes Surg. (2022) 32(7):1-7. doi: 10.1007/s11695-022-06066-0

7. Debi R, Oflazoglu K, Schouten R, Smeets S. Laparoscopic repair of an incidental Morgagni hernia during roux-en-Y gastric bypass in A severe obese adult: a case report frontiers. *Frontiers in Medical Case Reports.* (2021) 2(1):1–6. doi: 10.47746/FMCR.2021.2104

8. Alqahtani A, Aljohani E, Almadi F, Billa S. Laparoscopic sleeve gastrectomy with concomitant Morgagni hernia repair. *J Surg Case Rep.* (2019) 7:1–2. doi: 10.1093/jscr/rjz204

9. Horton JD, Hofmann LJ, Hetz SP. Presentation and management of Morgagni hernias in adults: a review of 298 cases. *Surg Endosc.* (2008) 22:1413–20. doi: 10. 1007/s00464-008-9754-x

10. Angrisani L, Lorenzo M, Santoro T, Sodano A, Tesauro B. Hernia of foramen of Morgagni in adult: case report of laparoscopic repair. *JSLS*. (2000) 4(2):177–81.

11. Tan YW, Banerjee D, Cross KM, De Coppi P, GOSH team, Blackburn SC, et al. Morgagni hernia repair in children over two decades: institutional experience, systematic review, and meta-analysis of 296 patients. *J Pediatr Surg.* (2018). 10:1883–9. doi: 10.1016/j.jpedsurg.2018.04.009

12. Karadag CA, Erginel B, Yildiz A, Kaba M, Demir M, Sever N. Laparoscopic repair of morgagni hernia in children. J Pediatr Surg. (2023) S0022-3468 (23):00073–8. doi: 10.1016/j.jpedsurg.2023.01.045

13. Oppelt PU, Askevold I, Bender F, Liese J, Padberg W, Hecker A, et al. Morgagnilarrey diaphragmatic hernia repair in adult patients: a retrospective single-center experience. *Hernia*. (2021) 25:479–89. doi: 10.1007/s10029-020-02147-0 Check for updates

#### **OPEN ACCESS**

EDITED BY Giuseppe Lauriti, G. d'Annunzio University of Chieti and Pescara, Italy REVIEWED BY

Vincenzo Davide Catania, Sant'Orsola-Malpighi Polyclinic, Italy Jun Fujishiro, The University of Tokyo, Japan

\*CORRESPONDENCE Eunyoung Jung Seyjung@dsmc.or.kr

RECEIVED 04 July 2023 ACCEPTED 18 August 2023 PUBLISHED 08 September 2023

#### CITATION

Jung E (2023) Minimally invasive management of combined esophageal atresia with tracheoesophageal fistula and duodenal atresia: a comprehensive case report. Front. Pediatr. 11:1252660. doi: 10.3389/fped.2023.1252660

#### COPYRIGHT

© 2023 Jung. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

#### Minimally invasive management of combined esophageal atresia with tracheoesophageal fistula and duodenal atresia: a comprehensive case report

#### Eunyoung Jung\*

Division of Pediatric Surgery, Department of Surgery, Keimyung University School of Medicine, Dongsan Medical Center, Daegu, Republic of Korea

A newborn presented with a rare combination of esophageal atresia with tracheoesophageal fistula (EA/TEF) and duodenal atresia (DA), which was successfully managed using minimally invasive surgical techniques. The patient was a 1-day-old male for whom passing a feeding tube was infeasible and who had a double bubble sign on radiography. The neonate underwent a thoracoscopic ligation of the tracheoesophageal fistula (TEF) and a laparoscopic duodeno-duodenostomy on the same day, resulting in stabilized vital signs. Ten days after the initial operation, a thoracoscopic esophago-esophagostomy was successfully performed. The patient demonstrated full feeding capability and normal weight gain after the surgeries. The co-occurrence of EA/TEF and DA is a rare and complex anomaly. This case indicates that minimally invasive techniques can effectively manage this condition.

#### KEYWORDS

esophageal atresia, tracheoesophageal fistula, duodenal atresia, laparoscopy, thoracoscopy, minimally invasive surgical procedures

#### 1. Introduction

Esophageal atresia (EA) and duodenal atresia (DA) are rare congenital anomalies that occur independently. Their simultaneous manifestation is even rarer, accounting for approximately 2% of cases (1, 2). This dual anomaly presents unique challenges, instigating debates over surgical strategy, including the potential requirement for routine gastrostomy (3–6). The existing literature has mainly focused on cases managed with traditional open surgical techniques (6–9). However, this report aims to contribute a novel perspective, detailing a case of a newborn treated successfully for EA, tracheoesophageal fistula (TEF), and DA via staged, minimally invasive procedures with percutaneous gastrostomy. This manuscript was prepared following the CARE guidelines (https://www.care-statement.org).

#### 2. Case description

A 1-day-old male neonate was referred to the department after a failed attempt to pass a feeding tube shortly after birth. He was born via a planned cesarean section at a gestational

Abbreviations

DA, duodenal atresia; EA, esophageal atresia; TEF, tracheoesophageal fistula.

age of 36 weeks and 3 days; the baby had a birth weight of 2,260 g. Polyhydramnios was diagnosed during the mother's pregnancy; however, the prenatal ultrasonography did not indicate the double bubble sign. The neonate's APGAR scores were 8 and 9 at 1 and 5 min, respectively. Immediate postnatal chest and abdominal radiographs revealed a coiled feeding tube in the esophageal pouch and a double bubble sign (Figure 1). These findings highly suggested the diagnoses of EA/TEF and DA. Although the newborn exhibited stable vital signs at birth, notable abdominal distension was observed without distal smallor large-bowel gas. With the onset of worsening abdominal distension and tachypnea, the team decided to operate on the day of birth. Before general anesthesia, gastric decompression was performed via percutaneous puncture with an 18G angiocatheter (Figure 2) in the operating room. With the patient supine, the distended stomach can be readily identified via visualization and percussion. This allows for the straightforward advancement of the catheter by percutaneous puncture without the need for imaging guidance. The catheter was kept in place



#### FIGURE 1

Chest and abdominal radiographs obtained post-orogastric tube insertion demonstrated a coiled orogastric tube (indicated by the yellow arrow) and the double bubble sign (indicated by the orange arrow).



FIGURE 2 Before general anesthesia, a percutaneous 18G angiocatheter was placed in the stomach without imaging guidance, serving as a temporary gastrostomy.

until the completion of the thoracoscopic TEF ligation. Considering the preoperative echocardiography confirming the aortic arch's location on the left side, the surgical approach was from the right side, with the infant placed in a semi-prone position. The initial procedure involved thoracoscopic ligation of the TEF (Figure 3) using the conventional three-trocar technique. The operative time for thoracoscopic TEF ligation was 20 min. Following the TEF ligation, the infant's position was adjusted to supine for the succeeding procedure. A 5-mm umbilical port and two additional ports were inserted in the right and left abdomen. The angiocatheter for temporary gastrostomy was removed under the guidance of a laparoscope after decompression of the stomach and duodenum to facilitate good surgical view in laparoscopic DA repair. A laparoscopic duodeno-duodenostomy was conducted to manage type III DA. The time for the laparoscopic DA repair was 90 min. The patient demonstrated a preoperative venous CO2 level of 48.4 mmHg. The level was reduced to 39.0 mmHg immediately during the postoperative phase. During the intraoperative phase, the endtidal carbon dioxide (ET<sub>CO2</sub>) level was consistently regulated at a mean of 32 mmHg (range: 30-36) during the thoracoscopic TEF Additionally, throughout the ligation. laparoscopic duodenostomy, the ET<sub>CO2</sub> level was sustained at an average of 39 mmHg (range: 29-47). Postoperative vital signs, including respiration and arterial saturation, were stable. On postoperative day 10, with the patient's weight improved to 2,440 g, a thoracoscopic esophago-esophagostomy was performed via the



FIGURE 3 A singular 5-mm metal clip was used to close the tracheoesophageal fistula (TEF)

previous trocar sites on the chest. The operative time was 140 min. Esophagography on postoperative day 5 showed no evidence of leakage or stricture. Consequently, feeding was initiated, and the patient was discharged on postoperative day 12, displaying full feeding capability. The patient's weight was 2.54 kg at discharge. At one month of life, he was 3.6 kg with full feeding. At 4 months, he was 7.3 kg that was normal growth with 60th percentile and without signs of esophageal stricture or reflux.

#### 3. Discussion

The concurrent presentation of EA/TEF and DA is a challenging medical condition due to its infrequent occurrence. While the prenatal detection rates of EA and DA individually are over 70% (10, 11), their simultaneous detection is rare. Therefore, swift and precise diagnosis is crucial when both anomalies are identified postnatally, as seen in this case. Nevertheless, the rarity of these co-existing malformations means that comprehensive studies addressing optimal treatment strategies are limited, with most available information derived from case reports or series (3, 4). The primary surgical considerations include the necessity for gastrostomy (4) and the timing for addressing both anomalies – either through single- or multistage surgical procedures (3, 8).

Single-stage surgery might seem ideal; however, no definitive evidence suggests superior survival outcomes compared to multistage surgery. Conversely, hasty surgical interventions without carefully considering the neonate's overall condition could lead to decreased survival rates (3, 5). Table 1 summarizes previously reported cases of combined EA with TEF and DA. The surgical management options and survivals are variable, and all the reported cases were performed using open thoracotomy and laparotomy. Ideally, performing surgeries for EA with TEF and DA concurrently without the need for a gastrostomy seems optimal. Particularly, if one can utilize thoracoscopy and laparoscopy for the surgery, it would be even more advantageous. If the patient's condition is stable and the procedure is conducted by a well-experienced surgeon, combining the surgeries in one session seems best. However, to date, there are no reports on spontaneous repair via minimally invasive surgery. Moreover, spontaneous repairs performed thoracotomy and laparotomy were reported to have a higher mortality compared to multistage repairs (3). Therefore, a tailored management plan that fits the patient's situation is essential. For multistage procedures, most literature indicates that surgery for esophagoesophagostomy, with or without gastrostomy, is usually conducted first (5-9). Although no literature explicitly states the reason, one can infer that for open thoracotomy, since one needs to make an incision for esophageal atresia with tracheoesophageal fistula ligation anyway, performing esophagoesophagostomy directly after closing the thoracotomy, instead of conducting another laparotomy for duodeno-duodenostomy, seems more practical. However, in some cases, there are concerns about complete duodenal obstruction due to duodenal atresia, leading to the implementation of a gastrostomy (8). In our case, since the duodenal atresia surgery was performed as a first stage operation, there was an effect of gastric decompression through duodeno-duodenostomy, eliminating the need to maintain a gastrostomy post-surgery.

In this case, persistent abdominal distension and tachypnea soon manifested despite the patient's initial stability. In instances

	Period	Cases (N)	Mean GA <sup>1</sup> (range)	Mean BW <sup>2</sup> (range)	Type C EA (N)	Gastrostomy	Simultaneous repair	Staged repair	EE first <sup>3</sup>	Mortality
Lee et al. (7) <sup>a</sup>	1989-2006	7	35 (32-40)	1,871 (1,100-2,600)	7/7 (100%)	3/7 (43%)	5/7 (71%)	1 (14%)	1/1	29%
Nabzdyk et al. (8)	2010-2012	3	34 (33–35)	1,822 (1,388-2,153)	3/3 (100%)	3/3 (100%)	0/3 (0%)	3/3 (100%)	3/3	0%
Fragoso et al. (12) <sup>b</sup>	1965-2012	10	35 (32–37)	2,240 (1,660-3,120)	10/10 (100%)	8/10 (80%)	8/10 (80%)	2/10 (20%)	1/2	50%
Miscia et al. (5) <sup>c</sup>	2000-2019	5	35 (33–36)	1,911 (1,350-2,365)	4/5 (80%)	1/5 (20%)	4/5 (80%)	1/5 (20%)	1/1	0%
Cao et al. (9)	2015-2018	4	39 (38–40)	2,748 (2,310-3,400)	4/4 (100%)	1/4 (25%)	3/4 (75%)	1/4 (25%)	1/1	0%
Ein et al. (3) <sup>d</sup>	1971-2000	24	35 (30–39)	2,100 (1,130-3,450)	17/24 (71%)	19/24 (80%)	7/24 (29%)	16/24 (67%)	unknown	25%
Dave et al. (6) <sup>a</sup>	1974-2003	10	36 (34-40)	2,317 (1,835-2,830)	9/10 (90%)	6/10 (60%)	4/10 (40%)	5/10 (50%)	5/5	10%
Spitz et al. (4) <sup>e</sup>	1964-1978	18	36 (30-42)	2,000 (1,200-2,800)	16/18 (89%)	5/18 (28%)	4/18 (22%)	8/18 (44%)	3/8	67%

TABLE 1 Summary of all studies for the type and surgeries for combined esophageal atresia and duodenal atresia.

GA, gestational age, weeks; BW, birth weight, gram; EE first, esophago-esophagostomy as a first operation in staged repaired patients. <sup>a</sup>One case expired without treatment.

<sup>b</sup>No information for mortality cases, only analyzed 10 survival case among 20 combined EA and DA.

<sup>c</sup>One case of staged repair is due to delayed diagnosis of duodenal web.

<sup>d</sup>Mortality is 12% In type C patients, and 57% in pure EA patients.

<sup>e</sup>Four cases were not treated.

Juna

of TEF without DA, stomach decompression might not be essential as stomach gas can transit through the distal gastrointestinal tract. However, with complete obstruction due to DA, continuous gastric distension could lead to irreversible lung damage from gastric aspiration via the TEF, necessitating emergency TEF ligation.

Although some studies advocate DA correction first before TEF ligation, if air entry into the stomach through TEF can be controlled with appropriate management of tidal volume, such delicate control is often challenging. Furthermore, some reports highlight the risk of gastric perforation during mechanical ventilation (13, 14). Therefore, beginning with TEF ligation can help stabilize the patient's airway. Similarly, Spitz et al. (4) recommended the creation of a wide gastrostomy following EA/ TEF repair.

The sequence of operations addressing EA/TEF and DA remains undetermined. While other reports performed fluoroscopy-guided percutaneous gastrostomy (15), an 18G angiocatheter for stomach decompression was utilized as an alternative to initial gastrostomy without imaging guidance in the current case, facilitating uncomplicated thoracoscopic TEF ligation. This shortens the procedure time for TEF ligation and minimizes the gastrostomy scar. This strategy prevented severe gastric dilatation and aspiration from excessive tidal volume during intraoperative ventilation, providing a clear surgical field view for the subsequent DA operation.

In situations where the patient's condition could rapidly deteriorate if a time-consuming laparoscopic duodenoduodenostomy is performed first, it is deemed safe to perform TEF ligation as soon as possible.

With advancements in neonatal surgery, anesthesia, and care, primary simultaneous repair has become an option. However, definitive evidence suggesting its superiority over staged operations is lacking. Ein et al. (3) reported a 75% survival rate in 24 cases of combined EA/DA over 30 years, with multistage operations unexpectedly showing better survival rates than single-stage procedures.

While single-stage surgeries may seem advantageous, the extended durations of anesthesia and surgery pose significant safety risks for neonates. Therefore, insisting on single-stage surgery without considering the potential negative impact on the newborn's outcome is unwarranted.

In conclusion, the co-occurrence of EA/TEF and DA is rare, necessitating a treatment approach that adapts to the individual patient and the resources available at the treating center. This report details a recent successful experience with a two-stage operation conducted without gastrostomy and utilizing minimally invasive surgical techniques. This approach underscores the evolving potential for neonatal treatment strategies in managing such complex cases.

#### Data availability statement

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

#### **Ethics statement**

Written informed consent was obtained from the minor(s)' legal guardian/next of kin for the publication of any potentially identifiable images or data included in this article.

#### Author contributions

EJ conceptualized and designed the study, drafted the initial manuscript, and revised it. EJ also performed all procedures and operations described in this paper. The author approved the final manuscript as submitted. The author meets the current ICMJE criteria for authorship.

#### 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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated

#### References

1. Stoll C, Alembik Y, Dott B, Roth MP. Associated anomalies in cases with esophageal atresia. Am J Med Genet A. (2017) 173(8):2139–57. doi: 10.1002/ajmg.a. 38303

2. Galarreta CI, Vaida F, Bird LM. Patterns of malformation associated with esophageal atresia/tracheoesophageal fistula: a retrospective single center study. Am J Med Genet A. (2020) 182(6):1351-63. doi: 10.1002/ajmg.a.61582

3. Ein SH, Palder SB, Filler RM. Babies with esophageal and duodenal atresia: a 30year review of a multifaceted problem. *J Surg.* (2006) 41(3):530-2. doi: 10.1016/j. jpedsurg.2005.11.061

4. Spitz L, Ali M, Brereton RJ. Combined esophageal and duodenal atresia: experience of 18 patients. *J Pediatr Surg.* (1981) 16(1):4–7. doi: 10.1016/s0022-3468 (81)80105-4

5. Miscia ME, Lauriti G, Di Renzo D, Riccio A, Lisi G, Lelli Chiesa P. Esophageal atresia and associated duodenal atresia: a cohort study and review of the literature. *Eur J Pediatr Surg.* (2021) 31(5):445–51. doi: 10.1055/s-0040-1716884

6. Dave S, Shi EC. The management of combined oesophageal and duodenal atresia. *Pediatr Surg Int.* (2004) 20(9):689-91. doi: 10.1007/s00383-004-1274-8

7. Lee YM, Nam SH, Kim DY, Kim SC, Kim IK. Clinical experience with esophageal atresia combined with duodenal atresia. *J Korean Assoc Pediatr Surg.* (2008) 14 (1):21–6. doi: 10.13029/jkaps.2008.14.1.21

8. Nabzdyk CS, Chiu B, Jackson CC, Chwals WJ. Management of patients with combined tracheoesophageal fistula, esophageal atresia, and duodenal atresia. *Int J Surg Case Rep.* (2014) 5(12):1288–91. doi: 10.1016/j.ijscr.2013.09.016

organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

#### Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fped.2023. 1252660/full#supplementary-material.

9. Cao ZP, Li QF, Liu SQ, Niu JH, Zhao JR, Chen YJ, et al. Surgical management of newborns with combined tracheoesophageal fistula, esophageal atresia, and duodenal obstruction. *Chin Med J.* (2019) 132(6):726–30. doi: 10.1097/CM9. 00000000000102

10. Saalabian K, Friedmacher F, Theilen TM, Keese D, Rolle U, Gfroerer S. Prenatal detection of congenital duodenal obstruction-impact on postnatal care. *Children*. (2022) 9(2):160. doi: 10.3390/children9020160

11. Pardy C, D'Antonio F, Khalil A, Giuliani S. Prenatal detection of esophageal atresia: a systematic review and meta-analysis. *Acta Obstet Gynecol Scand.* (2019) 98 (6):689–99. doi: 10.1111/aogs.13536

12. Fragoso AC, Ortiz R, Hernandez F, Olivares P, Martinez L, Tovar JA. Defective upper gastrointestinal function after repair of combined esophageal and duodenal atresia. *J Pediatrc Surg.* (2015) 50(4):531–4. doi: 10.1016/j.jpedsurg.2014.05.029

13. Maoate K, Myers NA, Beasley SW. Gastric perforation in infants with oesophageal atresia and distal tracheo-oesophageal fistula. *Pediatr Surg Int.* (1999) 15(1):24–7. doi: 10.1007/s003830050504

14. Jones TB, Kirchner SG, Lee FA, Heller RM. Stomach rupture associated with esophageal atresia, tracheoesophageal fistula, and ventilatory assistance. *AJR Am J Roentgenol.* (1980) 134(4):675–7. doi: 10.2214/ajr.134.4.675

15. Catalano P, Virardi L, Implatini A, Grimaldi SA, Enrico Papale AN, Tina LG, et al. Percutaneous radiologic gastrostomy as bridge to definitive surgery in a very preterm infant with combined esophageal and duodenal atresia. *J Pediatric Surg Case Rep.* (2021) 64:101704. doi: 10.1016/j.epsc.2020.101704

Check for updates

#### **OPEN ACCESS**

EDITED BY Giuseppe Lauriti, G. d'Annunzio University of Chieti and Pescara, Italy

#### REVIEWED BY

Sherif Mohamed Shehata, Tanta University, Egypt Shun Onishi, Kagoshima University, Japan

\*CORRESPONDENCE Go Miyano 🖾 go@juntendo.ac.jp

RECEIVED 10 July 2023 ACCEPTED 11 September 2023 PUBLISHED 04 October 2023

#### CITATION

Miyano G, lida H, Ebata Y, Abe E, Kato H, Mikami T, Ishii J, Lane GJ, Yamataka A and Okazaki T (2023) Comparative assessment of fully laparoscopic Duhamel-Z with minimal rectorectal dissection vs. laparoscopy-assisted Duhamel-Z with blunt manual rectorectal dissection for total colonic aganglionosis. Front. Pediatr. 11:1255899. doi: 10.3389/fped.2023.1255899

#### COPYRIGHT

© 2023 Miyano, lida, Ebata, Abe, Kato, Mikami, Ishii, Lane, Yamataka and Okazaki. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms. Comparative assessment of fully laparoscopic Duhamel-Z with minimal rectorectal dissection vs. laparoscopy-assisted Duhamel-Z with blunt manual rectorectal dissection for total colonic aganglionosis

Go Miyano<sup>1\*</sup>, Hisae Iida<sup>1</sup>, Yu Ebata<sup>1</sup>, Eri Abe<sup>1</sup>, Haruki Kato<sup>1</sup>, Takafumi Mikami<sup>1</sup>, Junya Ishii<sup>1</sup>, Geoffrey J. Lane<sup>2</sup>, Atsuyuki Yamataka<sup>2</sup> and Tadaharu Okazaki<sup>1</sup>

<sup>1</sup>Pediatric Surgery, Juntendo University Urayasu Hospital, Chiba, Japan, <sup>2</sup>Pediatric General & Urogenital Surgery, Juntendo University School of Medicine, Chiba, Japan

**Aims:** Early postoperative outcome (EPO) was compared between fully laparoscopic Duhamel-Z (F-Dz) and laparoscopy-assisted Duhamel-Z (A-Dz) anastomoses performed for total colonic aganglionosis (TCA).

**Methods:** EPO was assessed quarterly for the first year after F-Dz/A-Dz using a continence evaluation score (CES) based on stool frequency (motions/day) and stool consistency (0 = liquid, 1 = soft, 2 = formed), presence of anal erosion (0 = severe, 1 = moderate, 2 = mild), and incidence of enterocolitis.

Surgical technique involved taking the ileostomy down, dissecting the colon laparoscopically, and preparing the pull-through ileum through the stoma wound. In F-Dz (n = 3), a working port (SILS trocar) was inserted, and laparoscopic retrorectal dissection with forceps used to create a retrorectal tunnel from the peritoneal reflection extending downward as narrow as possible along the posterior wall of the rectum to prevent lateral nerve injury and preserve vascularity. After completing the tunnel, the ileum was pulled-through from an incision on the anorectal line and a Z-shaped ileorectal side-to-side anastomosis performed without a blind pouch. In A-Dz (n = 11), the retrorectal pull-through route was created through a Pfannenstiel incision using blunt manual (finger) dissection along the anterior surface of the sacrum.

**Results:** Subject backgrounds were similar. Mean quarterly data were: frequency (F-Dz: 4.67, 4.67, 4.67, 3.33) vs. (A-Dz: 7.27, 7.09, 6.18, 5.36) p < .05; consistency (F-Dz: 0.33, 0.67, 0.67, 0.67) vs. (A-Dz: 0.27, 0.45, 0.70, 0.73) p = ns; anal erosion (F-Dz: 0.33, 0.33, 0.33, 0.67) vs. (A-Dz: 0.18, 0.36, 0.45, 0.64) p = ns; and enterocolitis (F-Dz: 1 episode in 1/3 cases or 33.3%) vs. (A-Dz: 7 episodes in 6/11 cases or 54.5%) p = ns.

Conclusions: Overall, EPO after F-Dz was better than after A-Dz.

#### KEYWORDS

total colonic aganlionosis, duhamel-Z, laparoscopy, retrorectal dissection, continence

#### Introduction

Surgical intervention for total colonic aganglionosis (TCA) has been reported and modified over the years (1-4) without consensus for a treatment of choice being reached. The Duhamel procedure gained favor because there was less pelvic dissection and a segment of aganglionic rectum was used as a reservoir to reduce the frequency of defecation, improving bowel continence. Ikeda (5) modified the Duhamel procedure by introducing a Z-shaped ileorectal side-to-side anastomosis in 1967 that eliminated the rectal reservoir and dividing the colorectal septum, completely. In 2017, the first author reported gradual improvement in postoperative bowel continence in TCA patients treated by laparoscopy-assisted Duhamel-Z anastomosis (A-Dz) over time using a comprehensive continence evaluation score (CES) (6).

Here, early postoperative outcome (EPO determined from selected CES criteria considered most likely to influence quality of life such as frequency/consistency of motions, presence of anal erosion, and incidence of enterocolitis) after fully laparoscopic Duhamel-Z anastomosis (F-Dz) and A-Dz were compared in TCA patients.

#### Methods

A-Dz was performed from 2009 to 2019 and F-Dz since 2020. Cases in this series were consecutive. Surgically, for both F-Dz and A-Dz, four 5-mm ports were used to dissect the entire colon beginning from the peritoneal reflection at the sigmoid colon progressing proximally to the ileostomy site following the bowel wall closely using conventional laparoscopic techniques. After the ileostomy was taken down under laparoscopic control (7), pull-through ileum was prepared by dissecting the ileal mesentery through the abdominal ileostomy wound.

For F-Dz, a Free Access<sup>TM</sup> working port/platform (Top Corporation, Tokyo, Japan) was placed in the abdominal wound. Laparoscopic retrorectal dissection was performed using forceps extending downward to create a retrorectal tunnel. From the peritoneal reflection, dissection was performed as close as possible to the posterior wall of the rectum to prevent lateral nerve injury and preserve lateral vascularity (Figure 1). When laparoscopic dissection/tunneling reached the anorectal line (ARL), an incision was made on the ARL taking care to protect the surgical anal canal, and the ileum was pulled-through from this incision through the narrow retrorectal tunnel without additional dissection (Figure 2). The rectal stump was resected 2 cm above the peritoneal reflection, and a transverse incision was made on the anterior wall of the ganglionic ileum at the level of the proximal rectal end. The posterior wall of the upper rectum and lower edge of the incised anterior wall of the ileum were then anastomosed very tightly using interrupted sutures. The posterior wall of the rectum was incised on the ARL and the pulled-through ileum was anastomosed to the anus circumferentially in a single layer using interrupted 5-0 monofilament sutures. A 60 mm long Tri-Staple<sup>TM</sup> technology surgical stapler was inserted through the anus to divide the posterior rectal wall and anterior ileal wall. Finally, the anterior wall of the upper rectum and the upper edge of the incised anterior wall of the normal ileum were anastomosed in two layers to complete the Z-shaped ileorectal side-to-side anastomosis without a blind pouch (Figure 3).

For A-Dz, after the ileostomy was taken down, a Pfannennstiel incision was used to create an extensive retrorectal pull-through route using finger dissection along the anterior surface of the sacrum and perform a Z-shaped ileorectal side-to-side anastomosis as an open procedure. A-Dz was then completed using the same technique described for F-Dz.

Data were collected for demographics, clinical presentation, presence of associated anomalies, surgical management, and perioperative complications. EPO assessment was performed



#### FIGURE 1

Laparoscopic retrorectal dissection. Retrorectal dissection was essentially performed bluntly and as narrowly as possible, along the posterior wall of the rectum to prevent lateral nerve injury and preserve lateral vascularity in F-Dz. F-Dz: fully laparoscopic Duhamel-Z, A-Dz: laparoscopy-assisted Duhamel-Z.



quarterly for one year by specialist nursing staff with wound, ostomy, and continence (WOC) care certification; CES was determined from frequency (motions per day) and consistency (scored using: 0 = watery, 1 = soft, and 2 = formed, to calculate a consistency score) data by WOC staff; severity of anal erosion determined by WOC staff using: 0 = severe (requiring constant corticosteroid administration), 1 = moderate (requiring intermittent corticosteroid administration), 2 = mild (requiring emollients only); and the incidence of enterocolitis. Data were compared between F-Dz (n = 3) and A-Dz (n = 11). Data for A-Dz were obtained from a previously published report by the same authors.

Data were analyzed using standard statistical methods with the software Statcel-2 (OMS Publishing Inc., Saitama, Japan). Demographics were compared using Bonferroni corrected *post hoc* tests. EPO determined from selected CES criteria were compared using the Student's *t*-test. For all statistical tests, p < .05 was used to determine significance. Methodology and ethics were in accordance with the Helsinki Declaration (2013).

#### Results

F-Dz cases were 2 females and 1 male; A-Dz cases were 5 females and 6 males. Mean age and mean weight were 7.6 months old and 7.1 kg, respectively in F-Dz and 10.2 months and 8.4 kg in A-Dz; differences were not statistically significant (p = ns for age; p = ns for weight). Mean lengths of aganglionic

ileum resected (24.0 cm in F-Dz vs. 19.5 cm in A-Dz) were statistically similar (p = ns). Mean operative times were 5.9 h in F-Dz vs. 6.2 h in A-Dz; differences were not statistically significant (p = ns). There were no perioperative complications in either group.

No subjects were constipated during the immediate postoperative period. Mean frequencies of stools/day at 3, 6, 9 and 12 months after F-Dz were: 4.67  $\pm$  0.5, 4.67  $\pm$  01.1, 4.67  $\pm$  0.5, and  $3.33 \pm 0.5$ , respectively; after A-Dz were:  $7.27 \pm 1.4$ ,  $7.09 \pm$ 1.6,  $6.18 \pm 1.3$ , and  $5.36 \pm 1.4$ , respectively; p < .05 at 3, 6, and 12 months (Figure 4). Mean consistency scores at 3, 6, 9, and 12 months after F-Dz were:  $0.33 \pm 0.5$ ,  $0.67 \pm 0.5$ ,  $0.67 \pm 0.5$ , and  $0.67 \pm 0.5$ , respectively; after A-Dz were:  $0.27 \pm 0.4$ ,  $0.45 \pm 0.5$ ,  $0.70 \pm 0.6$ , and  $0.73 \pm 0.6$ , respectively; p = ns throughout the study period (Figure 5). Mean erosion scores at 3, 6, 9, and 12 months after F-Dz were:  $0.33 \pm 0.5$ ,  $0.33 \pm 0.5$ ,  $0.33 \pm 0.5$ , and  $0.67 \pm 0.5$ , respectively; after A-Dz were:  $0.18 \pm 0.4$ ,  $0.36 \pm 0.5$ ,  $0.45 \pm 0.5$ , and  $0.64 \pm 0.5$ , respectively; p = ns throughout the study period (Figure 6). Enterocolitis occurred in 1/3 F-Dz cases (33.3%) and there were 7 episodes in 6/11 A-DZ cases (54.5%); p = ns.

#### Discussion

Duhamel-Z is indicated primarily for rectosigmoid Hirschsprung disease. When performed as an open procedure, retrorectal dissection is performed transanally, using the fingers



#### FIGURE 3

Z-shaped ileorectal side-to-side anastomosis without a blind reservoir. The incised posterior wall of the rectum and pulled-through ileum were anastomosed at the anus, and a surgical stapler used to divide the posterior rectal wall and anterior ileal wall. Finally, the anterior wall of the upper rectum and the upper edge of the incised anterior wall of the normal ileum were anastomosed to complete the Z-shaped ileorectal side-to-side anastomosis without a blind reservoir.





FIGURE 5

Consistency of stools in both groups. Consistency of stools was not statistically different between F-Dz and A-Dz.



through a transverse incision on the posterior rectal wall to extend full-thickness upward along the surface of the sacrum, followed by creating a wide retrorectal tunnel between the anal side and the peritoneal cavity (8). However, when performed laparoscopically for TCA, retrorectal dissection is performed laparoscopically. A report detailing transanal retrorectal dissection during laparoscopic Duhamel-Z with the direction of dissection extending upward from a full-thickness hemicircumferential incision on the posterior wall of the rectum required the retrorectal tunnel to be wide enough to allow the folded rectum to be grasped and pulled-down through it (9). Thus, a feature of F-Dz used in the current report is that local trauma during dissection was minimized and lateral rectal ligaments preserved by using peanut gauze swabs for dissection in view of the density of neurovascular and venous plexuses on the pelvic surface of the sacrum. Similarly, electrocautery was not used for hemostasis to prevent compromising the rectal blood supply and presacral innervation (10) that could potentially disrupt postoperative bowel function.

F-Dz was also considered less invasive because laparoscopy ensured more precise and meticulous dissection of the posterior wall, preventing more extensive dissection of the sacrum associated with manual (finger) dissection or open dissection during A-Dz. As a result, better control of defecation reflected by the lower frequency of stools after F-Dz may be related to minimizing injury associated with manual dissection of the sacral surface or preventing the creation of a narrow pull-through route by limited dissection. The relatively younger age of F-Dz patients may be implicated in stool frequency results although age differences between groups in this study were not statistically significant. Since publishing the report from which A-Dz data was obtained, longer follow-up has shown further gradual improvement in overall CES over time, emphasizing that the anorectum has reserve for functional resolution after surgical intervention.

Major limitations of this study are the small sample size and short duration of follow-up. The F-Dz group had surgery at a relatively younger but not statistically significant age which could influence postoperative fecal continence, an issue that could not be confirmed because of the small series and short follow-up. However, despite the small sample size and short follow-up period for F-Dz in this study, improvement in EPO after F-Dz was considered worth reporting for its potential for enhancing the treatment of TCA and improving the quality of life of postoperative TCA patients. Since extending the follow-up period and increasing the sample size will allow a more comprehensive evaluation of surgery in the long-term, outcome after F-Dz is being closely monitored and will be reported when sufficient data is available.

#### 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 Juntendo University School of Medicine. The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

#### Author contributions

GM: Conceptualization, Formal Analysis, Funding acquisition, Methodology, Visualization, Writing – original draft, Writing – review & editing. HI: Data curation, Writing – review & editing. YE: Data curation, Writing – review & editing. EA: Data curation, Writing – review & editing. HK: Data curation, Writing – review & editing. TM: Data curation, Writing – review & editing. JI: Data curation, Writing – review & editing. GL: Validation, Writing – review & editing. AY: Resources, Writing – review & editing. TO: Supervision, Validation, Writing – review & editing.

#### Funding

The author(s) declare that no financial support was received for the research, authorship, and/or publication of this article.

#### 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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated

#### References

1. Martin LW. Surgical management of total colonic aganglionosis. *Ann Surg.* (1972) 176(3):343–6. doi: 10.1097/00000658-197209000-00010

2. Kimura K, Nishijima E, Muraji T, Tsugawa Y, Matsumoto Y. A new surgical approach to extensive aganglionosis. *J Pediatr Surg.* (1981) 16(6):840–3. doi: 10. 1016/s0022-3468(81)80830-5

3. Marquez TT, Acton RD, Hess DJ, Duval S, Saltzman DA. Comprehensive review of procedures for total colonic aganglionosis. *J Pediatr Surg.* (2009) 44(1):257--265. doi: 10.1016/j.jpedsurg.2008.10.055

4. Bischoff A, Levitt MA, Pena A. Total colonic aganglionosis: a surgical challenge. How to avoid complications? *Pediatr Surg Int*. (2011) 27(10):1047–52. doi: 10.1007/ s00383-011-2960-y

5. Ikeda K. New techniques in the surgical treatment of hirschsprung's disease. Surgery. (1967) 61(4):503-8.

6. Miyano G, Nakamura H, Seo S, Sueyoshi R, Okawada M, Doi T, et al. Laparoscopy-assisted Duhamel-Z anastomosis for total colonic aganglionosis:

organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

#### Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fped.2023. 1255899/full#supplementary-material

#### SUPPLEMENTARY VIDEO 1

Retrorectal dissection toward the perineum. Retrorectal dissection was performed as close as possible to the posterior wall of the rectum, and when dissection/tunneling reached the anorectal line, the laparoscopic surgeon can see the ARL being pressed by a finger from outside the body.

outcome assessed by fecal continence evaluation. J Laparoendosc Adv Surg Tech A. (2017) 27(3):302–5. doi: 10.1089/lap.2016.0216

 Miyano G, Yanai T, Okazaki T, Kobayashi H, Lane GJ, Yamataka A. Laparoscopyassisted stoma closure. J Laparoendosc Adv Surg Tech A. (2007) 17(3):395–8. doi: 10. 1089/lap.2006.0074

8. Urushihara N, Fukumoto K, Fukuzawa H, Sugiyama A, Watanabe K, Mitsunaga M, et al. Outcome of laparoscopic modified Duhamel procedure with Z-shaped anastomosis for Hirschsprung's disease. *Surg Endosc.* (2012) 26(5):1325–31. doi: 10. 1007/s00464-011-2031-4

9. Xi Z, Cao GQ, Tang ST, Chang XP, Li S, Yang L, et al. Laparoscopic-assisted Duhamel procedure with ex-anal rectal transection for total colonic aganglionosis. *J Pediatr Surg.* (2018) 53(3):531–6. doi: 10.1016/j.jpedsurg.2017.06.009

10. Miyano G, Yamada S, Murakami H, Lane GJ, Yamataka A. Case report: gross persistent rectal prolapse. A case treated without mesh using deep retrorectal dissection/suturing. *Front Pediatr.* (2022) 18(10):900081. doi: 10.3389/fped.2022. 900081

Check for updates

#### **OPEN ACCESS**

EDITED BY Gabriele Lisi, University of Studies G. d'Annunzio Chieti and Pescara, Italy

REVIEWED BY Caio Vinicius Suartz, University of São Paulo, Brazil Satoshi Obata, Fukuoka University School of Medicine and Hospital, Japan

\*CORRESPONDENCE Go Miyano 🖂 go@juntendo.ac.jp

RECEIVED 10 July 2023 ACCEPTED 14 September 2023 PUBLISHED 09 October 2023

#### CITATION

Miyano G, Takahashi M, Suzuki T, Iida H, Abe E, Kato H, Yoshida S, Lane GJ, Ichimura K, Sakamoto K, Yamataka A and Okazaki T (2023) Remote cadaveric minimally invasive surgical training. Eront Pediatr 11:1255882

doi: 10.3389/fped.2023.1255882

#### COPYRIGHT

© 2023 Miyano, Takahashi, Suzuki, lida, Abe, Kato, Yoshida, Lane, Ichimura, Sakamoto, Yamataka and Okazaki. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

## Remote cadaveric minimally invasive surgical training

Go Miyano<sup>1\*</sup>, Makoto Takahashi<sup>2</sup>, Takamasa Suzuki<sup>1</sup>, Hisae Iida<sup>1</sup>, Eri Abe<sup>1</sup>, Haruki Kato<sup>1</sup>, Shiho Yoshida<sup>3</sup>, Geoffrey J. Lane<sup>3</sup>, Koichiro Ichimura<sup>4</sup>, Kazuhiro Sakamoto<sup>2</sup>, Atsuyuki Yamataka<sup>3</sup> and Tadaharu Okazaki<sup>1</sup>

<sup>1</sup>Pediatric Surgery, Juntendo University Urayasu Hospital, Chiba, Japan, <sup>2</sup>Coloproctological Surgery, Juntendo University School of Medicine, Tokyo, Japan, <sup>3</sup>Pediatric General & Urogenital Surgery, Juntendo University School of Medicine, Tokyo, Japan, <sup>4</sup>Anatomy and Life Structure, Juntendo University School of Medicine, Tokyo, Japan

**Objective:** The aim of the study is to discuss the efficacy of live vs. remote cadaver surgical training (CST) for minimally invasive surgery (MIS).

Methods: A cohort of 30 interns in their first and second years of training were divided into three groups: live observers (n = 12), live participants (n = 6), and remote observers: (n = 12). The interns had the opportunity to either observe or actively participate in two different surgical procedures, namely, laparoscopic lower anterior resection, performed by a colorectal surgical team, and laparoscopic fundoplication, performed by a pediatric surgical team. The procedures were conducted either at a base center or at a remote center affiliated with the institute. Some of the interns interacted directly with the surgical teams at the base center, and others interacted indirectly with the surgical teams from the remote center. All interns were administered questionnaires before and after completion of the CST in order to assess their understanding of various aspects related to the operating room layout/ instruments (called "design"), accessing the surgical field (called "field"), understanding of anatomic relations (called "anatomy"), their skill of dissection (called "dissection"), ability to resolve procedural/technical problems (called "troubleshooting"), and their skill in planning surgery (called "planning") according to their confidence to operate using the following scale: 1 = not confident to operate independently; 4 = confident to operate with a more senior trainee; 7 = confident to operate with a peer; and 10 = confident to operate with a less experienced trainee. A p < 0.05 was considered statistically significant.

**Results:** All scores improved after CST at both the base and remote centers. The following significant increases were observed: for remote observers: "field" (2.67 $\rightarrow$ 4.92; *p* < .01), "anatomy" (3.58 $\rightarrow$ 5.75; *p* < .01), "dissection" (3.08 $\rightarrow$ 4.33; *p* = .01), and "planning" (3.08 $\rightarrow$ 4.33; *p* < .01); for live observers: "design" (3.75 $\rightarrow$ 6.17; *p* < .01), "field" (2.83 $\rightarrow$ 5.17; *p* < .01), "anatomy" (3.67 $\rightarrow$ 5.58; *p* < .01), "dissection" (3.17 $\rightarrow$ 4.58; *p* < .01), "troubleshooting" (2.33 $\rightarrow$ 3.67; *p* < .01), and "planning" (2.92 $\rightarrow$ 4.25; *p* < .01), "anatomy" (3.67 $\rightarrow$ 5.67; *p* < .01), "dissection" (2.83 $\rightarrow$ 6.83; *p* < .01), "anatomy" (3.67 $\rightarrow$ 5.67; *p* < .01), "dissection" (2.83 $\rightarrow$ 6.17; *p* < .01), "troubleshooting" (2.17 $\rightarrow$ 4.17; *p* < .01), "dissection" (2.83 $\rightarrow$ 4.67; *p* < .01). Understanding of "design" improved significantly after CST in live observers compared with remote observers (*p* < .01). Understanding of "field and "dissection" improved significantly after CST in live observers, 10 participants (83.3%) reported that interacting with surgical teams was easy because they were not on-site.

**Conclusions:** Although all the responses were subjective and the respondents were aware that observation was inferior to hands-on experience, the results from both centers were equivalent, suggesting that remote learning could potentially be viable when resources are limited.

KEYWORDS

remote education, minimally invasive surgery, cadaver surgical training, pediatric surgery, general surgery

#### Introduction

The postgraduate training program in Japan was revised in 2004. This revision affected graduates of accredited medical schools who had completed 6-year-long courses and successfully passed the national medical registration examination. The previous system, which involved immediate commencement of specialty training after graduation, was replaced with an internship system where all graduates are required to spend the first 2 years after graduation rotating through multiple departments. While the aim of this revision was to train doctors with more general experience and skills, the duration of surgical training was limited to a period of 6 months, which was very different from the former system in which prospective surgeons would immediately commence on their surgical training after graduation.

As a result, surgical trainees under the new system must commence surgical training with only limited exposure to surgery. In recent years, surgical training with training boxes and laboratory animals has increased, resulting in a distinct trend away from direct, hands-on management with some interns having little access to experiencing surgery. One of the primary challenges with utilizing training models and laboratory animals is their physical difference to actual patients; the variety of patients cannot be reproduced adequately with animals, and of course, there are differences in anatomic relations (1-3). Thus, options for improving exposure to surgery are decreasing while the COVID-19 pandemic and social distancing rules have also limited access to in-person training activities and prevented attendance at "live" teaching sessions. As a result, medical education has been forced to evolve rapidly to a virtual format (4) facilitated by improved data transmission. While surgical procedures are often video recorded for educational purposes, such as utilization at congresses/conferences and live digital learning events (5), and some hospitals even broadcasted operations to waiting rooms so relatives can observe the surgery, such options have not been generally applied for routine teaching purposes due to patient safety and ethical concerns associated with live surgery broadcasts (6).

Cadaver surgical training (CST) is an integral part of understanding and learning anatomy and a time-tested technique for obtaining valuable understanding of the structure and textures of the body required for confident surgical intervention. CST would be most beneficial as a "live" experience, but virtual or remote CST may provide exposure that would otherwise be unattainable and could prove to be valuable after COVID-19 related restrictions, such as social distancing and ease (7). Remote CST has been reported for plastic surgery (7), and its potential for minimally invasive surgery (MIS) training is being considered.

In order to provide surgical trainees with exposure to improve their education and understanding of anatomy and aspects of planning and logistics essential for successful surgery, a previous experience with CST and laparoscopy/thoracoscopy (8) was applied to determine the impressions of first and second year interns faced with increasingly limited opportunities for surgical experience exacerbated by COVID-19 restrictions. In this study, groups of interns either observed or participated in CST procedures directly at a base center (live observers/live participants) or observed the same procedures from a remote center at an affiliated institute (remote observers). In addition, their perceived confidence to operate was assessed by questionnaires administered before and after CST. Their responses were considered likely to be useful for assessing the potential of remote education as a viable alternative to direct teaching methods as a means for improving the efficiency of medical education.

#### Methods

The Juntendo University CST Center was established in 2019, rendering it accessible to the 18 specialty surgical departments at Juntendo University Medical School Hospital, as well as the Department of Anatomy at Juntendo University Medical School. The center is supported financially by the Japanese Ministry of Health, Labour, and Welfare, for the effective use of donated bodies for the advancement of science (8).

The CST Center located at Juntendo University Hospital (base center) was connected to a remote center at Juntendo University Urayasu Hospital (Figure 1) by a data transmission system established with a "KAKENHI" grant (grant number: 22K02835) from the Japan Society for the Promotion of Science and Kawano Masanori Memorial Public Interest Incorporated Foundation for Promotion of Pediatrics. Juntendo University utilized its own transmission network to offer on-site video and audio broadcast services using highly secure connections to prevent information leakage.

When the CST program was first established at Juntendo, the cadavers were embalmed using a saturated salt solution. For the current study, only cadavers preserved using Thiel's method (9) were used. A total of 30 first and second year interns either observed or participated in a laparoscopic lower anterior


FIGURE 1

Remote CST observation. First and second year interns participated as observers; they were allowed to ask questions freely during their CST session.

resection performed by a colorectal surgical team and a laparoscopic fundoplication performed by a pediatric surgical team directly at the base center (live observers = 12, live participants = 6) or observed the same procedures from the remote center (remote observers = 12). Live participants had the opportunity to alternate roles as the main surgeon, scope surgeon, and assistant surgeon. The two operations used for CST were chosen based on their frequent utilization in clinical practice, involving a variety of maneuvers. All interns were allowed to interact freely with the surgical teams directly at the base center and indirectly at the remote center. All interns involved at both centers were administered a questionnaire before and after their CST session that asked regarding their impressions of six criteria: operating room layout/logistics, including an introduction of trocars, trocar selection, and trocar insertion (called "design"), accessing/establishing the surgical field (called "field"), understanding of anatomic relations (called "anatomy"), understanding of basic dissection techniques (called "dissection"), dealing with procedural/technical problems (called "troubleshooting"), and how to plan surgery (called "planning"). Questionnaires assessed how confident interns with no hands-on experience of surgery would feel about operating, based on the understanding derived from their CST session. The responses were scored as follows: 1 = not confident to operate independently; 4 = confident to operate with a more senior trainee; 7 = confident to operate with a peer; 10 = confident to operate with a less experienced trainee. A sample questionnaire is presented in Table 1.

Data were analyzed using standard statistical methods with the software Statcel-2 (OMS Publishing Inc., Saitama, Japan). Technical background of interns was compared using Bonferroni corrected *post hoc* tests. The changes of the score from the questionnaire were compared using the Student's *t*-test. For all statistical tests, p < .05 was used to determine significance. Juntendo University institutional review board approval was obtained for this study (2019173). Methodology and ethics were in accordance with the Helsinki Declaration (2013).

#### TABLE 1 Perceived self-confidence.

	Pre-CST	Post-CST
1. Operating theater layout (including trocar selection)		
2. Accessing the surgical field		
3. Understanding anatomy		
4. How to dissect		
5. Solving problems		
6. Planning surgery		

1. Not confident at all; 4. Confident if with someone senior; 7. Confident with peer; 10. Confident to supervise someone junior.

#### Results

The technical backgrounds of the first and second year interns are shown in Table 2. There were no significant differences observed among the live observers, live participants, and remote observers. While the scores at both centers increased for all criteria after CST, a significant improvement was reported by the remote observers for the following: "field"  $(2.67 \pm 0.9 \rightarrow 4.92 \pm 0.9;$ p < .01), "anatomy"  $(3.58 \pm 0.7 \rightarrow 5.75 \pm 0.9; p < .01)$ , "dissection"  $(3.08 \pm 0.9 \rightarrow 4.33 \pm 0.9; p = .01)$ , and "planning"  $(3.08 \pm$  $1.0 \rightarrow 4.33 \pm 1.1$ ; p < .01); by live observers for all criteria: "design"  $(3.75 \pm 1.2 \rightarrow 6.17 \pm 1.1; p < .01)$ , "field"  $(2.83 \pm 1.1 \rightarrow 5.17 \pm 0.9;$ p < .01), "anatomy" (3.67 ± 0.7  $\rightarrow$  5.58 ± 0.7; p < .01), "dissection"  $(3.17 \pm 1.1 \rightarrow 4.58 \pm 0.9; p < .01)$ , "troubleshooting"  $(2.33 \pm$  $0.6 \rightarrow 3.67 \pm 0.9$ ; p < .01), and "planning"  $(2.92 \pm 1.0 \rightarrow 4.25 \pm 0.7)$ ; p < .01). Only "design" was significantly higher when the rates of increase between live observers and remote observers were compared (p < .01). Figure 2 shows the results for the live observers and remote observers. Interestingly, 10 participants out of the 12 (83.3%) remote observers returned the questionnaires with equivalent results to the live observers; seven out of the 12 remote observers (58.3%) were interested in attending another remote CST session.

For the live participants, significant increases were observed in the scores after CST for "design" ( $3.83 \pm 1.7 \rightarrow 6.33 \pm 1.0$ ; p = .02), "field" ( $2.83 \pm 0.7 \rightarrow 6.83 \pm 0.7$ ; p < .01), "anatomy" ( $3.67 \pm 0.8 \rightarrow 5.67 \pm 0.8$ ; p < .01), "dissection" ( $2.83 \pm 1.1 \rightarrow 6.17 \pm 1.3$ ; p < .01), "troubleshooting" ( $2.17 \pm 0.7 \rightarrow 4.17 \pm 0.7$ ; p < .01), and "planning" ( $2.83 \pm 0.7 \rightarrow 4.67 \pm 0.5$ ; p < .01). "Field" and "dissection" increased significantly in the live participants, reflecting the benefit of hands-on experience (p = .01, p = .03, respectively). Figure 3 shows the results for the live observers and live participants.

#### Discussion

The recent report (8) on CST for MIS training examined its relevance and the extent to which CST was considered realistic. The findings of this study were applied when designing the current study to establish the goals for training and how to assess the sense of achievement after CST at both the base and remote centers. The impressions of CST at both centers were comparable, with the remote observers particularly supportive of

#### TABLE 2 Overall dexterity of interns.

		Live observers	Live participants	Remote observers	<i>p</i> -Value
		( <i>n</i> = 12)	( <i>n</i> = 6)	( <i>n</i> = 12)	
Surgical field of interest	Gastrointestinal	3	2	4	ns
	Cardiac	1	1	1	
	Hepatobiliary	2	0	1	
	Pediatric	3	2	3	
	Others	3	1	3	
Type of workplace	University hospital	7	3	8	ns
	General hospital	2	1	1	
	Private clinic	1	0	0	
	Others	2	2	3	
Exercise experience	Team sports	8	5	9	ns
	Personal exercise	2	1	2	
	None	2	0	1	
Musical instrumental experience	Member of a group	1	0	1	ns
	Personal pleasure	3	2	4	
	None	8	4	7	
Video game experience	Regular player	2	2	3	ns
	Occasional player	8	3	7	
	None	2	1	2	



their remote learning experience. McIntyre et al. (10) compared the subjects observing surgery in an operating theater with subjects watching a live broadcast of the same surgery; the group engaged in live broadcast asked four times more questions than the group in the operating theater, and more of their questions were answered. There was a similar trend in this study with the

remote observers interacting more during CST than the live observers/live participants, possibly related to not being physically close to the surgeons teaching the CST session on-site. As all interns considered CST as a valuable learning experience, easier interaction would be advantageous and a possible reason why the remote observers found their experience so worthwhile



and actively chose "remote" as the format for a repeat CST session. In other words, a virtual cadaver experience provided exposure and an opportunity to learn that was less stressful than being with the instructors directly. The remote observers felt more relaxed due to their physical separation from the senior staff, which allowed them to focus on their observation and express their curiosity.

Another advantage of the remote center that may be relevant is that surgery may have been easier to watch because images were transmitted directly from the operation site. This was hinted at in a report on cadaveric plastic surgery training (7) and could be particularly relevant for MIS involving both thoracoscopy and laparoscopy. This is due to the fact that the live observers, live participants, and the teaching surgeons essentially share the same monitor in the operating room, while the remote observers can observe using several monitors, if available, and engage in open discussions regarding their observations. In fact, remote learning could potentially offer advantages over live observation in an operating room, particularly in situations when several trainees are clamoring for an opportunity to observe.

This simple study identified the potential value of remote education by providing data reflecting the appreciation and satisfaction experienced after a remote CST session. The findings suggest that remote education may be a valid modality for learning, although it is difficult to make specific conclusions regarding the potential value of remote education for learning physical skills due to the subjective nature of this study. Further research is required to determine how effective CST and remote CST are for preparing more experienced surgical trainees with previous experience and exposure to operating rooms and surgical procedures. Of particular interest would be assessing whether CST or remote CST influenced the progress of surgical training.

Based on the data obtained in the present study, while the results for the remote observers and live observers/live participants were similar, there were notable differences and discrepancies in certain criteria, indicating the existence of potential areas for improvement in order to enhance the effectiveness of CST as a learning experience. By identifying areas requiring more effort, the focus of planning for future medical education can be adjusted to overcome shortcomings and potentially include supplementary topics such as experimental surgery or instruction in surgical techniques using models.

As a baseline study for assessing the factors related to a successful remote learning, the favorable reaction of the remote observers would suggest that further research is worthwhile. In addition, the current study could be considered as a trial of the potential of remote learning. With the utilization of Juntendo's existing facilities and the development of a dedicated transmission system tailored for remote teaching, the same training can be conducted with enhanced clarity and broader technical input using more advanced facilities and has the potential for application anywhere, even internationally, with well-renowned surgeons hosting teaching sessions in real time. The legitimate concerns regarding security of information transfer and privacy require the involvement of expert technicians. However, the potential for expanding surgical training from the traditional "see one, do one, teach one" approach to a global interface using remote learning presents an

exciting opportunity for every surgeon, no matter how wellrenowned. While the scope of the current study is small, the education strategy that could develop based on the data presented could contribute to reducing gaps in surgical education through collaboration among different centers or institutes without any restrictions based on distance.

## 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 Juntendo University School of Medicine. The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

#### Author contributions

GM: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Visualization, Writing – original draft, Writing – review & editing. MT: Data curation, Methodology, Resources, Writing – review & editing. TS: Data curation, Resources, Writing – review & editing. HI: Data curation, Writing – review & editing. EA: Data curation Writing – review & editing. HK: Resources, Writing – review & editing. SY: Methodology, Writing

## References

1. Zimmerman H, Latifi R, Dehdashti B, Ong E, Jie T, Galvani C, et al. Intensive laparoscopic training course for surgical residents: program description, initial results, and requirements. *Surg Endosc.* (2011) 25(11):3636–41. doi: 10.1007/s00464-011-1770-6

2. La Torre M, Caruso C. The animal model in advanced laparoscopy resident training. *Surg Laparosc Endosc Percutan Tech.* (2013) 23(3):271–5. doi: 10.1097/SLE. 0b013e31828b895b

3. Luks FI, Peers KH, Deprest JA, Lerut TE. Gasless laparoscopy in infants: the rabbit model. *J Pediatr Surg.* (1995) 30(8):1206–8. doi: 10.1016/0022-3468(95) 90023-3

4. Ahmed H, Mohammed A, Elghazaly H. COVID-19 and medical education. Lancet Infect Dis. (2020) 20(7):777-8. doi: 10.1016/S1473-3099(20)30226-7

5. Artibani W, Ficarra V, Challacombe BJ, Abbou CC, Bedke J, Boscolo-Berto R, et al. EAU policy on live surgery events. *Eur Urol.* (2014) 66(1):87–97. doi: 10.1016/ j.eururo.2014.01.028

original draft. GL: Formal analysis, Validation, Writing – review & editing. KI: Project administration, Resources, Supervision, Validation, Writing – review & editing. KS: Supervision, Writing – review & editing. AY: Supervision, Writing – review & editing. TO: Supervision, Writing – review & editing.

### Funding

The author(s) declare financial support was received for the research, authorship, and/or publication of this article.

The authors declare that this study received funding JSPS KAKENHI (grant number JP22K02835) and Kawano Masanori Memorial Public Interest Incorporated Foundation for Promotion of Pediatrics.

# 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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

6. Duty B, Okhunov Z, Friedlander J, Okeke Z, Smith A. Live surgical demonstrations: an old, but increasingly controversial practice. *Urology*. (2012) 79 (5):1185.e7-11. doi: 10.1016/j.urology.2011.12.037

7. Lisiecki JL, Johnson SP, Grant D, Chung KC. Learners responses to a virtual cadaver dissection nerve course in the COVID era: a survey study. *Arch Plast Surg.* (2022) 49(5):676–82. doi: 10.1055/s-0042-1756351

8. Yoshida S, Miyano G, Tanaka M, Ikegami M, Kato H, Seo S, et al. Cadaver training for minimally invasive pediatric surgery: a preliminary report. J Laparoendosc Adv Surg Tech A. (2021) 31(12):1485–90. doi: 10.1089/lap.2021.0333

9. Thiel W. Supplement to the conservation of an entire cadaver according to W. Thiel. *Ann Anat.* (2002) 184(3):267–9. doi: 10.1016/S0940-9602(02) 80121-2

10. Brunckhorst O, Challacombe B, Abboudi H, Khan MS, Dasgupta P, Ahmed K. Systematic review of live surgical demonstrations and their effectiveness on training. *Br J Surg.* (2014) 101(13):1637–43. doi: 10.1002/bjs.9635

Check for updates

#### **OPEN ACCESS**

EDITED BY Giuseppe Lauriti, G. d'Annunzio University of Chieti and Pescara, Italy REVIEWED BY

Giovanni Cobellis, Marche Polytechnic University, Italy Miguel Alfedo Castellan, University of Miami, United States

\*CORRESPONDENCE

Di Xu ⊠ fjsleyk@163.com

IJSIEYK@105.COM

 $^{\dagger}\mbox{These}$  authors have contributed equally to this work

RECEIVED 12 June 2023 ACCEPTED 21 September 2023 PUBLISHED 02 November 2023

#### CITATION

He Y, Lin S, Xu X, He S, Xu H, You G, Chen J and Xu D (2023) Single-port-plus-one robot-assisted laparoscopic modified Lich-Gregoir direct nipple ureteral extravesical reimplantation in children with a primary obstructive megaureter. Front. Pediatr. 11:1238918. doi: 10.3389/fped.2023.1238918

#### COPYRIGHT

© 2023 He, Lin, Xu, He, Xu, You, Chen and Xu. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

# Single-port-plus-one robot-assisted laparoscopic modified Lich-Gregoir direct nipple ureteral extravesical reimplantation in children with a primary obstructive megaureter

Yufeng He<sup>2,1t</sup>, Shan Lin<sup>1,2t</sup>, Xinru Xu<sup>2,1t</sup>, Shaohua He<sup>1,2</sup>, Huihuang Xu<sup>1,2</sup>, Guangxu You<sup>2,1</sup>, Jianglong Chen<sup>1,2</sup> and Di Xu<sup>1,2\*</sup>

<sup>1</sup>Department of Pediatric Surgery, Children Medical Center, Fujian Provincial Hospital, Fuzhou, China, <sup>2</sup>Shengli Clinical Medical College of Fujian Medical University, Fuzhou, China

**Purpose:** To introduce a new technique of single-port-plus-one robotic laparoscopic-modified Lich-Gregoir direct nipple ureteral extravesical reimplantation and ascertain its validity in the treatment of pediatric primary obstructive megaureter.

**Methods:** Between January 2021 and November 2021, we retrospectively analyzed the clinical data of 12 children with primary obstructive megaureter who were admitted to the Department of Pediatric Surgery of Fujian Provincial Hospital. All 12 children were treated with single-port-plus-one robotic laparoscopic Lich-Gregoir direct nipple ureteral extravesical reimplantation. Five of them were female and seven were male, including nine cases were simple obstructive type, while the remaining three cases were obstructive with reflux type. The mean age of the children was  $17.33 \pm 6.99 (10-36)$  months and the mean follow-up time was  $14.16 \pm 1.75 (12-17)$  months. Changes in preoperative and first-year postoperative parameters were compared.

**Results:** The mean operative time for all 12 children was  $123.58 \pm 10.85$  (110-145) min, with a mean internal operative time of  $101.42 \pm 0.85$  (90-120) min, a mean operative bleeding time of  $2.42 \pm 0.67$  (2-4) ml, and a mean hematuria duration of  $16.08 \pm 1.44$  (14-19) h. The mean indwelling catheterization time was  $2.25 \pm 0.45$  (2-3) days and the mean hospitalization time was  $3.83 \pm 0.39$  (3-4) days. At the postoperative first year, the ureteral diameter, calyceal diameter, and anterior-posterior renal pelvic diameter were found to be significantly smaller than at the preoperative period ( $18.83 \pm 3.21$  mm vs.  $6.83 \pm 1.27$  mm,  $13.99 \pm 3.58$  mm vs.  $3.5 \pm 2.90$  mm, and  $34.92 \pm 4.25$  mm vs.  $10.08 \pm 1.88$  mm, P < 0.001). There was a significant increase in renal cortical thickness and the percentage of differential renal function ( $3.63 \pm 1.66$  mm vs.  $5.67 \pm 1.88$  mm,  $33.75 \pm 2.77$  mm vs.  $37.50 \pm 1.31$  mm, P < 0.001). The resolution rate of obstruction was 100% and no child developed DeNovo vesicoureteral reflux.

Abbreviations

UR, ureteral reimplantation; VUR, vesicoureteral reflux; POM, primary obstructive megaureter; DRF, differential renal function; APRPD, anterior–posterior renal pelvic diameter; MIS, minimally invasive surgery; UTIs, urinary tract infections; UTDS, urinary tract dilation risk stratification; VCUG, voiding cystourethrogram; MRU, magnetic resonance urography; DMSA, dimercaptosuccinic acid.

**Conclusion:** The technique of modified Lich-Gregoir direct nipple ureteral extravesical reimplantation can help maintain the physiological direction of the ureter and at the same time enhance the effectiveness of antirefluxing in robotic surgery. The design of a single-port-plus-one wound can produce a cosmetic appearance by concentrating and hiding the wound around the umbilicus. This modified reimplantation procedure has the potential to become a promising technique in the robot-assisted treatment of primary obstructive megaureter.

#### KEYWORDS

single-port-plus-one, robotic laparoscopic surgery, primary obstructive megaureter, direct nipple, ureteral extravesical reimplantation

## Introduction

The term Primary Obstructive Megaureter (POM), which was coined by Caulk in 1923 (1), results in hindered urine evacuation and dilates the ureter and renal collecting system (2). Most will resolve over time. However, in approximately 20% of patients with uncontrolled urinary tract infection (UTI) or high-grade or progressive obstruction, ureteral reimplantation is required (3). Traditionally, open ureteral reimplantation is the gold standard for primary obstructive megaureter. In the past decade, pediatric urologists have frequently implemented minimally invasive procedures by using laparoscopic, robotic-assisted, and other devices. The Da Vinci robot-assisted laparoscopic surgery platform, which is characterized by a 6° freedom articulation, tremor filtering, and stereoscopic vision, provides distinct advantages in intracorporeal reparation and suturing (4). These advantages have expanded the role of robot-assisted surgery in complex lower urinary tract reconstructive surgeries, which are possibly time-consuming due to the narrow pelvic space (5). To promote the use of robot-assisted surgery for the treatment of obstructed megaureter and to achieve optimal outcomes, herein, we implemented a novel technique called "single-port-plus-one robotic laparoscopic-modified direct nipple Lich-Gregoir extravesical reimplantation", in an attempt to minimize technical difficulties. We reviewed our prospectively collected data and assessed the short-term safety and feasibility of the procedure.

#### Patients and methods

Between January 2021 and November 2021, we retrospectively reviewed 12 patients with POM who were admitted to the Department of Pediatric Surgery of Fujian Provincial Hospital, of whom five were females and seven were males. Their mean age was  $17.33 \pm 6.99$  (10–36) months and the mean follow-up time was  $14.16 \pm 1.75$  (12–17) months. Out of 12 patients, five are on the right side and seven are on the left side. The patients on the right are all obstructive. Among the patients on the left, four are obstructive and three are obstructive with reflux. A prenatal diagnosis was carried out in all 12 patients. Two patients experienced abdominal pain and three presented with UTIs.

All patients underwent a preoperative magnetic resonance urography (MRU), urological ultrasound, diuretic renogram (99 m Tc-DTPA), renal static imaging (99 m Tc-DMSA), and voiding cystourethrography (VCUG). All patients presented with a worsening dilatation (6) of 5 UTDP3 grade and 7 UTDP2 grade, and all of them had a deteriorating differential renal function (DRF) and obstructive curves on serial scans. A preoperative VCUG was done in all patients, and three of them had a vesicoureteral reflux (VUR), with grade 1 in two and grade 3 in one.

All patients underwent single-port-plus-one robotic laparoscopic-modified direct nipple Lich-Gregoir extravesical reimplantation. Five of them underwent ureteral tapering repair, which was performed by using Hendren's technique. Informed consent was obtained from the parents of the children, and the ethical review of the study was performed in our institution (ethics approval number: K2020-12-033).

In the postoperative second month, the patients were readmitted to the hospital for cystoscopy, observation of the ureteral orifice pattern, and removal of the double J tube. On the first year after surgery, the patients again returned for a urological ultrasound, MRU, diuretic nephrography, renal static imaging (99m Tc-DMSA), and VCUG to assess their recovery. Also, on the first year after surgery, all patients underwent a cystoscopy for an observation of the ureteral orifice, and the F3 ureter catheter was passed through the ureteral orifice to confirm no kicking in the ureter.

#### Inclusion and exclusion criteria

**Inclusion criteria:** POM patients with symptoms such as febrile UTIs or pain were included in the study (7). Asymptomatic patients with a DRF below 40% and which was associated with massive or progressive hydronephrosis, or a drop of >5% in differential function, were included.

**Exclusion criteria**: Patients with secondary giant ureters caused by ectopic ureteral opening, neurogenic bladder, posterior urethral valve, or urethral stenosis were excluded from the study (7).

#### Surgical procedures

All robot-assisted laparoscopic surgeries were performed by the same surgeon and surgical team.

After administering a successful general anesthesia, the patients were kept in the supine position, with head low and foot high, the bilateral upper limbs were placed in a "surrender" position, and the bilateral upper limbs were slightly opened. All parts of the body under pressure were padded with a sponge and fixed with bandages. Disinfecting, draping, and urethral catheterization were performed before the starting the single-port-plus-one robotic laparoscopic-modified Lich-Gregoir direct nipple ureteral extravesical reimplantation.

Although the da Vinci system Xi has four robotic arms, we used only three [three-dimensional (3D) camera arm III, operating arm II, and operating arm IV] and inserted a quadruple-channel puncture device to improve cosmetic appearance. We made a 2.5–3 cm curved incision along the edge of the umbilical cord to place a quadruple-channel puncture device, the four channels of which were used to insert an 8 mm 3D camera port III and an 8 mm operating port IV and were also used as assistant channels in a subsequent operation.

Artificial pneumoperitoneum was established with a pressure rate of  $10 \text{ mmH}_2\text{O}$  and a flow rate of 4 L/min. Another 8 mm incision was made 6 cm away from 3D camera port III on the right side of the abdomen for inserting robotic operating port II, as shown in Figures 1A,B.

Instead of using the traditional two peritoneal incisions to identify the ureter, we devised a new method of creating a "peritoneal window." We used a transverse incision to open the peritoneal layer along the surface of the posterior bladder wall, dissected the peritoneal layer from the posterior bladder wall on both sides, and then used an undamaged vessel clip to fix the peritoneal layer with the surrounding tissue. In these ways, we established a "peritoneal window," in which the vas or uterine artery could be directly identified from the ureter, as shown in **Figures 1C,D**.



#### FIGURE 1

(A,B) A 2.5–3 cm curved incision was made along the edge of the umbilical cord to place a quadruple-channel puncture device. Another 8 mm incision was made 6 cm away from 3D camera port III on the right side of the abdomen for inserting the robotic operating port II. (C,D) We established a "peritoneal window," in which the vas or uterine artery was directly identified from the ureter. (E,F) The direction of the detrusor tunnel was designed obliquely. The bladder detrusor was cut and disassembled until bulging of the bladder mucosa occurred, the length of the incision is five times the normal width of the ureter. (G,H) The overdilated ureter was cut and shaped and a double J tube was inserted into the ureter. The 1.5 cm end of the ureter was placed into the bladder. The seromuscular layer of the ureter and the bladder mucosa were sutured at 3, 6, 9, and 12 o' clock positions with 6-0 PDS. Finally, four stitches were made in each quadrant to complete the anastomosis. (I) A "down-top" method was applied to complete an intermittent incision of the cut detrusor suture with 4-0 Vicryl.

The direction of the detrusor tunnel was designed obliquely and a local electrocoagulation marker was generated. Traction sutures of the posterior bladder wall were inserted through the abdominal wall above the middle of the pubic symphysis and pulled out from the inserted position. In this way, the posterior bladder wall was pulled obliquely to the abdominal wall, and the detrusor tunnel was kept straight, which was convenient for cutting and dissecting. Inject saline into the bladder through a catheter to fill it. The bladder detrusor was cut and disassembled until bulging of the bladder mucosa occurred, the length of the incision is five times the normal width of the ureter. The outward and upward detrusor tunnel was more in line with the physiological direction of the ureter, as shown in **Figures 1E,F**.

The stricture segment of the ureter was resected, the overdilated ureter was cut and shaped as needed, and a double J tube was inserted into the ureter. Then, the 1.5 cm end of the ureter was placed into the bladder. Next, we sutured the seromuscular layer of the ureter and the bladder mucosa at 3, 6, 9, and 12 o' clock positions with 6-0 PDS, and finally, four stitches were made in each quadrant to complete the anastomosis, as shown in Figures 1G,H.

The "down-top" method was applied to complete an intermittent incision of the cut detrusor suture with 4-0 Vicryl, as shown in **Figure 1I**. Finally, the peritoneal layer of the posterior bladder wall was closed and the wound was sutured.

## Results

The mean operative time for all 12 children was  $123.58 \pm 10.85$  (110–145) min, with a mean internal operative time of  $101.42 \pm 0.85$  (90–120) min, a mean operative bleeding of  $2.42 \pm 0.67$  (2–4) ml, and a mean hematuria duration of  $16.08 \pm 1.44$  (14–19) h. The mean indwelling catheterization time was  $2.25 \pm 0.45$  (2–3) days, and the mean hospitalization time was  $3.83 \pm 0.39$  (3–4) days, as shown in Table 1.

At the postoperative first year, the ureteral diameter, calyceal diameter, and anterior–posterior renal pelvic diameter (APRPD) were found to be significantly smaller than at the preoperative period ( $18.83 \pm 3.21 \text{ mm vs.} 6.83 \pm 1.27 \text{ mm}$ ,  $13.99 \pm 3.58 \text{ mm vs.} 3.5 \pm 2.90 \text{ mm}$ , and  $34.92 \pm 4.25 \text{ mm vs.} 10.08 \pm 1.88 \text{ mm}$ , P < 0.001) (Figures 2A,B: the yellow arrow). There was a significant increase in renal cortical thickness and the DRF percentage ( $3.63 \pm 1.66 \text{ mm vs.} 5.67 \pm 1.88 \text{ mm}$ ,  $33.75 \pm 2.77 \text{ mm}$  vs.  $37.50 \pm 1.31 \text{ mm}$ , P < 0.001), as shown in Table 2.

In the postoperative period, no patient developed urinary retention, urinary extravasation, and wound infection. Two patients developed postoperative UTI treated conservatively and no one demonstrated reflux on VCUG. The resolution rate of obstruction was 100% and no patient developed DeNovo VUR.

At the postoperative second month, the patients were readmitted to the hospital for a cystoscopy for removal of the double J catheter, and the end of the ureter that was placed into the bladder was found to turn into nipples automatically, as shown in **Figure 2C**, and the wounds were cosmetic, as shown in

ion													
Complication													
Ŭ													
Hospitalization (days)	4	4	4	4	4	e	4	3	4	4	4	4	
n time													
terizatio 's)													
ng catheteri (days)	2	2	2	2	2	2	33	3	3	2	2	2	
Indwellir													
Blood loss Gross hematuria Indwelling catheterization time (ml) (h)													
oss hema (h)	15	16	18	19	17	15	16	15	14	17	16	15	
ss Gro													
3lood loss (ml)	2	ŝ	3	2	2	2	2	4	3	2	2	2	
Operation time (mins)	112	130	145	123	116	117	135	134	110	125	124	112	
Oper													
UTD grade	P3	P2	P3	P3	P3	P2	P3	P2	P2	P2	P2	P2	
The type of POM <sup>a</sup>					~		~			~			
Side .	0	0	0	0	, OR	0	, OR	0	0	OR	0	0	
_	Г	R	Г	Г	Ц	R	Г	Г	R	Г	R	R	
Age (months)	13	10	12	15	17	24	36	20	18	16	13	14	UTD, urinary tract dilation.
Sex	Boy	Girl	Boy	Boy	Girl	Boy	Girl	Boy	Girl	Boy	Boy	Girl	urinary tr
0 N	1	7	ю	4	ß	9	~	×	6	10	11	12	UTD,

Basic patient information on the modified Lich surgery

TABLE 1

source of POM typing

the

<sup>a</sup>Represents

According to the system of the American Pediatric Association for classifying the megaureter. O represents obstruction without refluxing, and QR represents obstruction with refluxing



#### FIGURE 2

(A,B) At the postoperative first year, there was a significant reduction in APRPD and ureteral diameter compared with those before the operation (yellow arrow). (C) The end of the ureter that was placed into the bladder was found to turn into nipples automatically in cystoscopy at the postoperative second month. (D) Cosmetic appearance of the wound at the postoperative second month (the scar is marked by a red arrow). (E) In the first year after surgery, the F3 ureter catheter was passed through the ureteral orifice smoothly into the ureter, confirming no kicking in the ureter.

**Figure 2D**. On the first year after surgery, all patients underwent cystoscopy for observation of the ureteral orifice again, and the F3 ureter catheter was passed through the ureteral orifice smoothly into the ureter, which confirmed no kicking in the ureter, as shown in Figure 2E. The trends of variations in the ureteral diameter, APRPD, and calyceal diameter at the preoperative and 1-year postoperative time points are shown in Figure 3. The trends of renal cortex thickness and DRF variation at the preoperative and 1-year postoperative time points are shown in Figure 4.

#### Discussion

According to the International Classification of Diseases, congenital megaureter can be classified either as obstructed and refluxing or as unobstructed and unrefluxing (7). The treatment

goal of POM is to relieve obstruction while establishing a new and effective antireflux mechanism. The primary minimally invasive surgical approaches for the correction of an obstructed megaureter that were used so far and are still used today are laparoscopic extravesical reimplantation and the pneumovesical laparoscopic approach (8, 9). Pneumovesical laparoscopic approaches such as Cohen and Leadbetter are often technically challenging and require a long curve, even for the most experienced laparoscopic surgeons. In infants and fat adolescents, they are often more challenging due to the small bladder volume and thickness of the abdominal wall (10, 11). In recent years, the robot-assisted laparoscopic platform with a stereoscopic vision and flexible arms has provided a new opportunity for the laparoscopic extravesical approach for POM.

Two robotic-assisted laparoscopic ureteral extravesical reimplantation methods for the treatment of POM have been described in recent studies. One is robotic-assisted extravesical

TABLE 2 Comparison of the preoperative parameters and 1-year postoperative parameters.

	Ureteral diameter (mm), mean ± SD	Calyceal diameter (mm), mean ± SD	APRPD (mm), mean ± SD (mm)	Renal cortex thickness (mm)	DRF %, mean ± SD
Preoperative	$18.83 \pm 3.21$	$13.99 \pm 3.58$	$34.92 \pm 4.25$	$3.63 \pm 1.66$	$33.75 \pm 2.77$
First-year postoperative	$6.83 \pm 1.27$	$3.5 \pm 2.90$	$10.08 \pm 1.88$	$5.67 \pm 1.88$	$37.50 \pm 1.31$
d	$12.00 \pm 2.52$	$10.49 \pm 4.20$	$24.83 \pm 3.19$	$2.14\pm0.60$	$3.75 \pm 1.71$
t	12.30	8.638	18.501	-12.168	-4.24
Р	<0.001	<0.001	<0.001	<0.001	<0.001



cross-trigonal ureteral reimplantation (12, 13). In 2020 (13), Neheman et al. described a novel surgical approach in the pediatric population and found that anastomosis was more ergonomic when performing a horizontal detrusorrhaphy, as opposed to a vertical one, and the submucosal tunnel was easier to extend. But the technique also had disadvantages in the form of challenges with endoscopic ureteral intubation during followup, if needed, postoperative cytoplasm, and urinary retention.

The other method is robotic-assisted extravesical Lich-Gregoir ureteral reimplantation, which is mainly used in high-grade VUR surgeries (14, 15). We reviewed the currently published literature on robot-assisted laparoscopic ureteral reimplantation (RALUR) and found that the VUR resolution rates widely ranged from 66.7% to 100%, which may be attributed to a submucosal tunnel of insufficient length (15). In this investigate, we implemented a new method by maintaining the vertical submucosal tunnel in the Lich-Gregoir technique and adding ureteral direct nipple implantation to produce an antireflux effect. As indicated by previous reports, traditional ureteral nipple implantation is not convenient to perform in adults (16, 17). Al-Shukri and Alwan (18) first reported direct nipple ureteroneocystostomy in adults with a ureteral stricture (19). In 2014, Fu et al. also used this method in robot surgeries in adult megaureters. In our new technique, we used this method in the pediatric population. In all 12 children in our study, the new ureteral orifice automatically acquired a papillary shape upon cystoscopy 2 months after surgery. At the postoperative first year, the ureteral diameter, calyceal diameter, and APRPD were found to be significantly smaller than at the preoperative period. There was a significant increase in renal



cortical thickness and DRF percentage. The resolution rate of obstruction was 100% and no child developed DeNovo VUR on VCUG. The results of the postoperative tests showed that this strategy was simple, safe, and feasible. In particular, the ureteral direct nipple implantation also shortened the anastomosis time, and the vertical submucosal tunnel remained in the physiological direction of the ureter, which was confirmed by the smooth passage of the 3F ureteral catheter upon cystoscopy at the postoperative 1-year period.

According to research by Leissner et al. of human cadaver dissection, the main portion of the pelvic plexus is located at about 1.5 cm dorsal and medial to the ureterovesical junction. The bundles of the pelvic plexus end at the distal ureter, trigone, and rectum (20). The oblique detrusor tunnel designed in the Lich-Gregoir technique may help preserve the neurovascular bundles and avoid potential voiding issues such as urinary retention during mobilization of the ureter.

Traumas experienced by children will get magnified as they grow up. That is why minimally invasive surgery has gradually become the mainstream treatment in the pediatric population in recent years. Unlike the four scattered wounds (three robotic port wounds and one assisted laparoscopic port wound) in the abdomen wall (21, 22), the wounds created by the single-siteplus-one technique are concentrated and hidden around the umbilical cord, which are more concealed when children grow up. When 5 mm robotic arms or the Da Vinci SP system are used by Chinese surgeons in the future, there is a possibility that all robotic arms are concentrated in a single-site port to realize a successful single-site robot-assisted surgery.

## Conclusion

The technique of modified Lich-Gregoir ureteral direct nipple extravesical reimplantation can help maintain the physiological direction of the ureter and at the same time enhance the effectiveness of antirefluxing in robotic surgery. The design of a single-port-plus-one wound can produce a cosmetic appearance by concentrating and hiding the wound around the umbilicus. This modified reimplantation procedure has the potential to become a promising surgical technique in the robot-assisted treatment of POM.

## Data availability statement

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

#### References

2. Pfister RC, Hendren WH. Primary megaureter in children and adults. Clinical and pathophysiologic features of 150 ureters. *Urology*. (1978) 12(2):160–76. doi: 10. 1016/0090-4295(78)90327-8

#### Ethics statement

The studies involving humans were approved by the institutional review board (IRB) of the Fujian Provincial Hospital (Ethics No: K2020-12-033). The studies were conducted in accordance with local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

# Author contributions

YH wrote the manuscript. SL and XX collected data, performed the statistical analysis, and revised the manuscript. SH, HX, and GY collected data and performed the statistical analysis. SL, XX, and DX conceptualized the study. JC and DX revised the manuscript and funded the work. DX was the primary surgeon. All authors contributed to the article and approved the submitted version.

## Funding

This work was supported by the Talent Induction Projection of Fujian Province (Grant Number: 2021YJ03), the Natural Science Foundation of Fujian Province (Grant Number: 2021J01386), the Medical Innovation Projection of Fujian Provincial Health Department Foundation (Grant Number: 2020CXB004), the Science and Technology Innovation Joint of Fujian Province (Grant Number: 2021Y9019), and the Natural Science Foundation of Fujian Province (Grant Number: 2022J05208).

## 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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

4. Bindi E, Todesco C, Nino F, Torino G, Gentilucci G, Cobellis G. Robotic surgery: is there a possibility of increasing its application in pediatric settings? A

<sup>1.</sup> Caulk JR. Megaloureter: the importance of the ureterovesical valve. J Urol. (1923) 9:315–9.

<sup>3.</sup> Zhu W, Zhou H, Cao H, Li P, Tao Y, Ma L, et al. Modified robot-assisted laparoscopic infantile ureteral reimplantation technique for obstructive megaureter. *J Pediatr Surg.* (2022) 57(12):1011–7. doi: 10.1016/j.jpedsurg.2022.05.015

single-center experience. Children (Basel). (2022) 9(7):1021. doi: 10.3390/ children9071021

5. Arlen AM, Broderick KM, Travers C, Smith EA, Elmore JM, Kirsch AJ. Outcomes of complex robot-assisted extravesical ureteral reimplantation in the pediatric population. *J Pediatr Urol.* (2016) 12(3):169.e1-6. doi: 10.1016/j.jpurol. 2015.11.007

6. Hwang J, Kim PH, Yoon HM, Song SH, Jung AY, Lee JS, et al. Application of the postnatal urinary tract dilation classification system to predict the need for surgical intervention among neonates and young infants. *Ultrasonography.* (2023) 42 (1):136–46. doi: 10.14366/usg.22035

7. Wein AJ, Kavoussi LR, Partin AW, Peters CA. Campbell-Walsh urology: 4-volume set. 11th ed. Philadelphia, PA: Elsevier Science Health Science (2016).

8. Soulier V, Scalabre AL, Lopez M, Li C-Y, Thach S, Vermersch S, et al. Laparoscopic vesicoureteral reimplantation with Lich-Gregoir approach in children: medium-term results of 159 renal units in 117 children. *World J Urol.* (2017) 35 (11):1791–8. doi: 10.1007/s00345-017-2064-y

9. Teklali Y, Robert Y, Boillot B, Overs C, Piolat C, Rabattu PY. Endoscopic management of primary obstructive megaureter in pediatrics. *J Pediatr Urol.* (2018) 14(5):382–7. doi: 10.1016/j.jpurol.2018.05.027

10. Baek M, Han DH. Transvesicoscopic Politano-Leadbetter ureteral reimplantation in children with vesicoureteral reflux: a novel surgical technique. *Investig Clin Urol.* (2019) 60(5):405–11. doi: 10.4111/icu.2019.60.5.405

11. Ansari MS, Yadav P, Arora S, Singh P, Sekhon V. Bilateral transvesicoscopic cross-trigonal ureteric reimplantation in children: surgical subtleties and a prospective summary. *Urology*. (2017) 101:67–72. doi: 10.1016/j.urology.2016.11.034

12. Rappaport YH, Kord E, Noh PH, Koucherov S, Gaber J, Shumaker A, et al. Minimally invasive dismembered extravesical cross-trigonal ureteral reimplantation for obstructed megaureter: a multi-institutional study comparing robotic and laparoscopic approaches. *Urology*. (2021) 149:211–5. doi: 10.1016/j.urology.2020.10.018

13. Neheman A, Kord E, Koucherov S, Kafka I, Gaber J, Noh PH, et al. A novel surgical technique for obstructed megaureter: robot-assisted laparoscopic

dismembered extravesical cross-trigonal ureteral reimplantation-short-term assessment. J Endourol. (2020) 34(3):249–54. doi: 10.1089/end.2019.0192

14. Akhavan A, Avery D, Lendvay TS. Robot-assisted extravesical ureteral reimplantation: outcomes and conclusions from 78 ureters. *J Pediatr Urol.* (2014) 10(5):864–8. doi: 10.1016/j.jpurol.2014.01.028

15. Grimsby GM, Dwyer ME, Jacobs MA, Ost MC, Schneck FX, Cannon GM, et al. Multi-institutional review of outcomes of robot-assisted laparoscopic extravesical ureteral reimplantation. *J Urol.* (2015) 193(5 Suppl):1791–5. doi: 10.1016/j.juro. 2014.07.128

16. Tatlişen A, Ekmekçioğlu O. Direct nipple ureteroneocystostomy in adults with primary obstructed megaureter. *J Urol.* (2005) 173(3):877–80. doi: 10.1097/01.ju. 0000152533.93716.3c

17. Chung H, Jeong BC, Kim HH. Laparoscopic ureteroneocystostomy with vesicopsoas hitch: non-refluxing ureteral reimplantation using cystoscopy-assisted submucosal tunneling. *J Endourol.* (2006) 20(9):632–8. doi: 10.1089/end.2006.20.632

18. Al-Shukri S, Alwan MH. Bilharzial strictures of the lower third of the ureter: a critical review of 560 strictures. *Br J Urol.* (1983) 55(5):477–82. doi: 10.1111/j.1464-410x.1983.tb03352.x

19. Fu W, Zhang X, Zhang Y, Zhang P, Gao J, Dong J, et al. Pure laparoscopic and robot-assisted laparoscopic reconstructive surgery in congenital megaureter: a single institution experience. *PLoS One.* (2014) 9(6):e99777. doi: 10.1371/journal.pone.0099777

20. Leissner J, Allhoff EP, Wolff W, Feja C, Höckel M, Black P, et al. The pelvic plexus and antireflux surgery: topographical findings and clinical consequences. *J Urol.* (2001) 165(5):1652–5. doi: 10.1016/S0022-5347(05)66384-7

21. Alizai NK, Dawrant MJ, Najmaldin AS. Robot-assisted resection of choledochal cysts and hepaticojejunostomy in children. *Pediatr Surg Int.* (2014) 30(3):291–4. doi: 10.1007/s00383-013-3459-5

22. Kim NY, Chang EY, Hong YJ, Park S, Kim HY, Bai S-J, et al. Retrospective assessment of the validity of robotic surgery in comparison to open surgery for pediatric choledochal cyst. *Yonsei Med J.* (2015) 56(3):737–43. doi: 10.3349/ymj. 2015.56.3.737

Check for updates

#### **OPEN ACCESS**

EDITED BY Giovanni Cobellis, Marche Polytechnic University, Italy

#### REVIEWED BY

Tetsuya Ishimaru, National Center for Child Health and Development (NCCHD), Japan Ciro Esposito, University of Naples Federico II, Italy

#### \*CORRESPONDENCE

Petra Zahradnikova pezahradnikova@gmail.com Jozef Babala jozef.babala@gmail.com

†These authors have contributed equally to this work and share first authorship

RECEIVED 31 August 2023 ACCEPTED 30 October 2023 PUBLISHED 14 November 2023

#### CITATION

Zahradniková P, Babala J, Pechanová R, Smrek M, Vitovič P, Laurovičová M, Bernát T and Nedomová B (2023) Inanimate 3D printed model for thoracoscopic repair of esophageal atresia with tracheoesophageal fistula. Front. Pediatr. 11:1286946. doi: 10.3389/fped.2023.1286946

#### COPYRIGHT

© 2023 Zahradniková, Babala, Pechanová, Smrek, Vitovič, Laurovičová, Bernát and Nedomová. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

# Inanimate 3D printed model for thoracoscopic repair of esophageal atresia with tracheoesophageal fistula

Petra Zahradniková<sup>1\*†</sup>, Jozef Babala<sup>1\*†</sup>, Rebeka Pechanová<sup>1</sup>, Martin Smrek<sup>1</sup>, Pavol Vitovič<sup>2</sup>, Miroslava Laurovičová<sup>2</sup>, Tomáš Bernát<sup>3</sup> and Barbora Nedomová<sup>4</sup>

<sup>1</sup>Department of Pediatric Surgery, Faculty of Medicine, Comenius University and National Institute of Children's Diseases, Bratislava, Slovakia, <sup>2</sup>Faculty of Medicine, Institute of Medical Education and Simulations, Comenius University, Bratislava, Slovakia, <sup>3</sup>Faculty of Medicine, Comenius University, Bratislava, Slovakia, <sup>4</sup>Department of Paediatric Anaesthesiology and Intensive Medicine, Faculty of Medicine, Comenius University and National Institute of Children's Diseases, Bratislava, Slovakia

**Background:** Thoracoscopic repair of esophageal atresia (EA) and tracheoesophageal fistula (TEF) poses significant technical challenges. This study aimed to develop an inexpensive, reusable, high-fidelity synthetic tissue model for simulating EA/TEF repairs and to assess the validity of the simulator.

**Methods:** By using 3D printing and silicone casting, we designed an inexpensive and reusable inanimate model for training in thoracoscopic EA/TEF repair. The objective was to validate the model using a 5-point Likert scale and the Objective Structured Assessment of Technical Skills (OSATS) to evaluate participants' surgical proficiency.

**Results:** A total of 18 participants (7 medical students, 4 pediatric surgery trainees, and 7 experienced surgeons), after being instructed and trained, were asked to perform TEF ligation, dissection, as well as esophageal anastomosis using six sliding knots on the EA/TEF simulator. All participants in the expert group completed the task within the 120-minute time limit, however only 4 (57%) participants from the novice/intermediate completed the task within the time limit. There was a statistically significant difference in OSATS scores for the "flow of task" (p = 0.018) and scores for the "overall MIS skills" (p = 0.010) task distinguishing between novice and intermediates and experts. The simulator demonstrated strong suitability as a training tool, indicated by a mean score of 4.66. The mean scores for the model's realism and the working environment were 4.25 and 4.5, respectively. Overall, the face validity was scored significantly lower in the expert group compared to the novice/intermediate groups (p = 0.0002).

**Conclusions:** Our study established good face and content validity of the simulator. Due to its reusability, and suitability for individual participants, our model holds promise as a training tool for thoracoscopic procedures among surgeons. However, novices and trainees struggled with advanced minimally invasive surgical procedures. Therefore, a structured and focused training curriculum in pediatric MIS is needed for optimal utilization of the available training hours.

#### KEYWORDS

oesophagus, minimally invasive surgery (MIS), thoracoscopic surgery, oesophageal atresia (EA), laparoscopic training, synthetic tissue, models, simulation

## Introduction

Pediatric surgeons have shown growing interest in minimally invasive surgery (MIS) for esophageal atresia with distal tracheoesophageal fistula (EA/TEF) since the initial successful thoracoscopic repair performed by Rothenberg in an infant with EA/TEF (1). Performing thoracoscopic repair for EA/TEF poses technical challenges and remains an unfamiliar procedure for numerous pediatric surgeons. Furthermore, the majority of infants undergoing EA/TEF repair are exceptionally small at the time of surgery, resulting in a confined and narrow surgical field that presents considerable technical difficulties for the surgeons. MIS in infants involves a challenging learning curve when it comes to suturing techniques and tissue manipulation. This learning curve often translates to extended operative times until surgeons attain full expertise in these skills (2). Specifically, suturing techniques pose significant challenges, especially in pediatric patients, because of the smaller tissue size and the restricted abdominal space for maneuvering instruments (3). On the other hand, pediatric surgeons often spend a significant portion of their careers waiting to perform certain procedures. The stakes with pediatric patients are so high that usually only the most experienced specialists are allowed to perform these procedures, creating a gap. Thus, a gap is created when experienced surgeons in the middle of their careers become frustrated while realizing they have so much further left to go. They realize they are expected to sit and wait until it is finally their turn.

Simulation has proven to be a fundamental tool in surgical education (4). In recent years, shortening of the learning curve has been demonstrated by implementing of wide range of surgical models (5-7). Due to the possibility of repeating the simulation, both the surgeons and residents have the great opportunity to learn surgical techniques, or particular steps, with the possibility of making mistakes, practice abilities and procedures in standardized and supervised situations (8). Ideally, simulation should enable the deliberate, repetitive, and participatory practice of neonatal MIS operative steps. This practice should utilize a validated model capable of identifying and correcting performance errors (9-11). Similarly, it should determine when the required skills have been acquired with a reliable degree of accuracy (12). The integration of threedimensional (3D) printing technology and imaging data from CT and MRI scans has unveiled new possibilities in crafting highfidelity laparoscopic simulators. These simulators accurately replicate, to scale, the environment encountered in neonatal surgery (10). This technology is potentially more cost-effective and avoids the ethical issues associated with using cadaveric and animal tissues. It is believed that due to current technological advances the future models will likely be both of high fidelity and low cost (12). Conversely, inanimate simulation models, especially those considered low-fidelity, are thought to lack the realism found in more sophisticated counterparts. However, research suggests that increased realism in a simulator doesn't necessarily correlate with improved learning outcomes. This prompts questions about the justification of the added costs

associated with high-fidelity simulators, especially when comparable knowledge and skill outcomes can be attained with more budget-friendly alternatives (13, 14). The objective of this study was to introduce and validate an inexpensive, reusable inanimate model designed for training in the thoracoscopic repair of EA/TEF.

#### **Methods**

#### Simulator development

The esophageal atresia model was developed using a combination of FDM (fused deposition modeling) 3D printing and casting of platinum-cured silicone. Initially, a CT scan was utilized to segment a child's spinal column along with the ribcage, scapula, and clavicles using 3DSlicer. The resulting STL file (a format for saving 3D models) was then modified in Blender3D to incorporate internal fixtures for the esophageal pouches and trachea (Figure 1). In total, the 3D-printed components consist of three models: a baseplate onto which the other two parts securely attach to prevent any unwanted movement, a ribcage model, and an internal fixator model (Figure 2). The printing process took slightly less than 24 h to complete using a Prusa i3 MK3S printer set to a layer height of 200 micrometers, with the spinal column positioned facing upwards. A total of 178 g of Prusament PLA (priced at 30 Euro per kilogram) was used, resulting in a 3D filament cost of 5.00 Euro. For crafting the silicone esophagus, an FDM printer was also employed to produce two molds made of PLA plastic-one for the distal pouch and another for the proximal one. These molds, when filled with silicone, enabled the creation of a hollow tube with a wall thickness of under 2mm. The silicone was prepared by blending two components (A and B) along with a designated silicone dye (Figure 3). Power-mesh was integrated into the silicone mixture to prevent tearing during suturing. The curing process of this silicone takes up to four hours, which meant that several days were required to complete the task and produce an adequate number of esophagus models and pouches



FIGURE 1

Process of segmentation: CT scan was utilized to segment a child's spinal column along with the ribcage, scapula, and clavicles using 3D slicer.



(Figures 4, 5). To simulate human skin, the same silicone and dye were used, poured loosely into a rectangular mold  $(25 \times 25 \text{ cm})$  with a height of 1 mm and lined with power-mesh. The bottom surface of the mold was composed of synthetic leather, which imprinted a skin-like texture onto the skin imitation (Figure 6).

#### Participants

The current study was conducted between October 2022 and August 2023 at the National Institute of Children's Diseases in Bratislava, Slovakia. All participants were anonymized and



#### FIGURE 3

For crafting the silicone esophagus, an FDM printer was also employed to produce two molds made of PLA plastic—one for the distal pouch and another for the proximal one. The diameter of the proximal esophagus was 10 mm, while the distal esophageal pouch measured 7 mm in diameter.





categorized as novice, intermediate, and expert based on their previous training and experience levels. Novice participants were defined as medical students in their fifth year of medical study. Intermediate participants were identified as pediatric surgical trainees in the 4th, 5th and 6th years of training. Expert participants were characterized by having a minimum of 15 years of experience in pediatric surgery. Prior to attempting the trial, every participant viewed brief instructional videos and accompanying written guidelines for each task. During each trial, participants were also given straightforward instructional cues. Furthermore, simple instructional cues were reiterated throughout each trial. The tasks included:

- a) Closure of TEF using a single sliding knot suture
- b) Division of TEF
- c) Opening of the upper esophageal pouch



FIGURE 5 Internal part EA/TEF model ready for training. Silicone esophagus can be changed by one person in 5 min.



d) Esophageal anastomosis involving three sliding knots on the back side and three on the front side. The anastomosis was performed intracorporeally in simulator All participants' attempts at these tasks were recorded and saved for subsequent analysis.

Throughout the procedures, SZABO-BERCI-SACKIER Laparoscopic Trainer was used. This laparoscopic trainer contains diaphragms at the typical puncture sites and a flexible endoscope holder that provides the surgeon with the ability to manipulate instruments with both hands. This trainer was borrowed for the purpose of this study from the RADIX MEDICAL Ltd. team (exclusive distributor of KARL STORZ in Slovak republic).

#### Assessments and parameters examined

After each procedure, four parameters were assessed:

- **Surgical time**: This metric measured the time taken from the entry of instruments into the simulator until the completion of the last sliding knot.
- Objective structured assessment of technical skills (OSATS): Performance during the simulated tasks was evaluated using the OSATS methodology. Each individual trial was recorded and later reviewed by three independent expert pediatric surgical consultants who were blinded to the identity and skill level of each participant. A modified OSATS scoring system was utilized to grade each trial on various aspects, including dexterity, flow of task, spatial orientation, needle holding, instrument movement, sliding knots performing, MIS skills, and overall performance. Scores ranged from 1 (inexperienced) to 5 (expert) for each aspect, resulting in a total score range of 8–40. The examiners maintained their blindness to participant identity and skill level throughout the assessment process.
- **Participants' feedback:** A Likert scale questionnaire was employed to gather participant feedback, with responses ranging from 1 to 5, addressing the following parameters:
  - Face validity
    - a) Realism of the model
    - b) Haptic feedback on materials

- Content validity: Assessing the value and suitability of the model as a training tool.
- Overall impression
  - a) Utility as a simulator
  - b) Utility for training in thoracoscopic EA/TEF repair
  - c) Utility as training and educational tools
- Quality of the anastomosis: This aspect involved a blinded analysis in each model, evaluating the number of sufficient knots, luminal passage, and anastomosis strength under tension.

Each model was reviewed by two independent expert pediatric surgical consultants who were blinded. The goal of this analysis was to assess the number of sufficient knots (the task involved creating 6 knots around EA). Luminal passage evaluation was performed using flexible endoscopy. To measure the strength of the anastomosis under tension, we employed a spring force meter, applying a tensile force of 250 g (equivalent to 2.5 N). We deemed the anastomosis successful if it could withstand a tensile force of 2.5 N (Figures 7, 8).

#### Trial study

Initially, each participant was allowed to train basic and advanced laparoscopic skills (needle holding, sliding knot suturing) according individually experience. This training was tailored to individual experience levels. Laparo Adavance (LAPARO Sp. z o.o. Poland, European Union) and Laparo Analytic (LAPARO Sp. z o.o. Poland, European Union) simulation tool was used for training (Figure 9).

Throughout the procedures, Karl Storz laparoscopic equipment and recording devices were utilized. The surgical instrument setup included a 5-mm telescope at a 30-degree angle and 3-mm instruments (grasper, needle holder, and scissors). Surgipro 5/0 (polypropylene) thread was used for the continuous sliding knot sutures for the EA/TEF repair. Standard port placement procedures were followed. Each task commenced by initiating computer video recording and inserting a 30° 5 mm laparoscopic camera into the simulator. Subsequently, participants introduced their specific tools through standardized pre-made punctures in the silicone skin cover. The model was utilized to simulate the thoracoscopic EA/TEF repair. Only the internal silicone pouches



Model prepared for analysis.



required replacement after each use, incurring a cost of approximately 5 Euros per participant. Between participants, the model could be prepared for reuse in less than 10 min.

#### Statistical analysis

Statistical analyses were performed with SPSSv25 software (IBM, NY, USA). All values were represented as mean with the standard deviation. The Mann–Whitney U test and one-way ANOVA were used to ascertain significant differences between groups. A nominal significance level of 0.05 was adopted.

#### Results

The study received approval from the hospital ethics committee. Eighteen participants were enrolled in the study, with a median age of 33.5 years (interquartile range: 20–59). Among the participants, 7 (39%) were female, and 17 (94%) were righthand dominant. The participant distribution included 7 (39%) medical students classified as novices, 4 (22%) intermediate



FIGURE 9 Basic/advanced laparoscopic training.

(pediatric surgery trainees with fewer than 20 minimally invasive surgeries of experience), and 7 (39%) experienced individuals with over 15 years of expertise in pediatric surgery. In the expert group, all participants successfully completed the task within the allocated 120-minute timeframe. However, only one participant from the novice group and 2 (50%) form the intermediate group managed to complete the task within the time limit. Notably, the time taken to complete the attempt was longer in the intermediate and novice groups compared to the expert group (p = 0.006). Five (71%) medical students managed to finish the task involving TEF closure with a single sliding knot. However, they lacked the skill to perform end-to-end anastomosis using sliding knots. In the group of inexperienced surgeons, 2 (50%) were able to complete all three tasks (**Table 1**).

All participants in the expert group managed to complete all three tasks, however, when examining the quality of the anastomosis, it was revealed that only 5 (71%) models in the expert group had a sufficient number of knots. The adequacy of anastomosis strength under tension was observed in 4 (57%) models, whereas one anastomosis was too tight for effective lumen passage.

#### Quality of the anastomosis

A total of 10 models with esophageal anastomosis were evaluated. Among these models, there were 1 novice, 2 intermediate, and 7 experienced participants. Out of the 10 models assessed, only 5 demonstrated sufficient anastomosis with 6 stitches each, and all of these models were from the expert group. In the case of the other models, either the stitches were missing before the evaluation, or they had become loosened (untied) during the assessment. The adequacy of anastomosis strength under tension was observed in 4 models (using tensile force of 2.5 N), whereas one anastomosis was too tight for effective lumen passage (using flexible endoscope). Nine models could be evaluated by passage with a flexible endoscope (luminal passage was possible).

Experienced surgeons achieved a higher total OSATS score compared to the inexperienced group, consisting of novices and intermediates (18 (15–23) vs. 15.6 (7–22), p = 0.313); however, this difference did not reach statistical significance. Notably, there was a statistically significant discrepancy in OSATS scores for the 'flow of task' (p = 0.018) and 'MIS skills' (p = 0.010) tasks, effectively distinguishing between novices/intermediates and experts. While experts tended to score higher than novices and intermediates for each task, this trend did not achieve statistical significance, likely reflecting the study's statistical power (**Table 2**).

#### Content validity

Using a 5-point Likert scale, the majority of participants expressed the view that the 3D synthetic model of EA/TEF serves as a suitable training tool (mean: 4.68). Almost all participants believed there is a role for simulation-based training and

Level of training/ expertise	Total number of participants	Mean age year (SD)	MIS procedures/ year	Completed TEF ligation and division	Completed all 3 tasks within time limit (120 min)	Mean operating time (min)
Novice	7	22 (0.95)	0	5 (71%)	1 (14%)	142 (101–188)
Intermediate	4	30 (0.95)	<20	4 (100%)	2 (50%)	93 (70-109)
Expert	7	48.5 (6.16)	>20	7 (100%)	7 (100%)	73 (57–95)

TABLE 1 Participant characteristics.

educational training (mean: 4.62). When evaluating the content validity of TEF dissection/ligation and sliding knot anastomosis between esophageal pouches, the experienced group provided realistic mean scores of 4.12 and 4.14, respectively.

#### Face validity

For assessing face validity, the experienced group responded to a series of questions aiming to rate the realism of different aspects of the simulator using a 5-point scale. The mean scores for the realism of the model and the working environment were 4.25 and 4.5, respectively. The haptics of the esophageal pouches received the lowest score (3.75). Interestingly, the experienced group scored significantly lower than the novice /trainee's group for 'TEF caliber' (3.57 vs. 4.22, p = 0.002) and 'proximal esophageal pouch caliber' (3.71 vs. 4.55, p = 0.003). Overall, the face validity was scored significantly lower in the experienced group compared to the inexperienced group (p = 0.0002). In terms of the overall impression, the highest ratings in the physical attribute's domain were for the overall impression and the usefulness of the tool as a simulator, with mean scores of 4.66 and 4.75, respectively. A comprehensive breakdown of responses from all participants can be found in Table 3.

#### Discussion

Across the globe, there is a growing adoption of MIS techniques in pediatric surgery, with these methods becoming the preferred standard for specific indications. As a result, there is an increasing demand for laparoscopic surgeons who are not only well-trained but also certified to address this expanding requirement (15, 16). MIS poses greater technical challenges compared to conventional open surgery, with a heightened risk of complications, particularly during the initial stages of

TABLE	2	OSATS	scoring	results.
INDEL	<u> </u>	OSAIS	scornig	results.

experience (17). The demand for pediatric surgical simulators appears to be increasing as a consequence of the rarity of several complex neonatal conditions (e.g., EA), and the limited opportunities the trainees have to acquire the demanded technical skills on actual neonates. Simulation can be of low or high fidelity, according to how closely it resembles real life (10). The degree of fidelity should align with the specific task and the stage of training that the participant is in (11). The combination of three-dimensional (3D) printing technology and imaging data from CT and MRI scans has created new possibilities for developing high-fidelity laparoscopic simulators. These simulators can accurately replicate, to scale, the environment encountered in neonatal surgery (12). Nair et al. created and validated with using 3D modeling and printing high fidelity model with the exactly size and shape of neonatal chest. They demonstrated construct validity for two basic "foundation" tasks: ring transfer and needle pass. They concluded, that complex tasks such as intracorporeal suturing will be needed for training and assessing more experienced surgeons (18). Our study showcased that in more intricate tasks, such as intracorporeal suturing involving sliding knots, the capability to distinguish between medical students and trainees, as well as between trainees and experts, was particularly effective. It's plausible that surgeons in training already possess good spatial awareness and experience in minimally invasive surgeries; however, they may lack proficiency in intracorporeal suturing. Simulation offers the advantage of being conducted in a low-risk setting outside of the operating theater, eliminating any potential harm to patient safety. Participants can utilize validated models to practice specific tasks or the entire procedure in a deliberate and repetitive fashion within a secure and cost-effective learning environment (19). Assessing surgical skills in an objective manner presents challenges. Previous research has documented validated simulator metrics, encompassing factors such as time elapsed, the overall count of movements, and the cumulative path length. These metrics were subject to objective evaluation through specialized

		Novices $(n = 7)$	Intermediate $(n = 4)$	Expert ( <i>n</i> = 7)	<i>p</i> -value
Dexterity	Median (range)	2 (1-3)	2.75 (2-3)	2.3 (2-3)	0.209
Flow of task	Median (range)	1.86 (1-3)	3.25 (3-4)	2.57 (2-4)	0.018
Spatial orientation	Median (range)	2.23 (1-4)	3 (2-4)	3.14 (2-4)	0.230
Needle holding	Median (range)	2.14 (1-3)	2.5 (1-4)	2.71 (2-4)	0.579
Instruments movement	Median (range)	1.85 (1-3)	2.75 (2-3)	2.71 (2-4)	0.100
Sliding knots	Median (range)	1.57 (1-3)	2 (1-3)	1.71 (1-3)	0.684
MIS skills	Median (range)	1.42 (1-3)	2.5 (2-3)	2.57 (2-3)	0.010
Overall score	Median (range)	13 (7–22)	18.75 (15-22)	18 (15-23)	0.074

	Novices/intermediate (n = 11)	Expert ( $n = 7$ )	Overall ( <i>n</i> = 18)	<i>p</i> -value
Face validity (mean score [SD])	4.44 (0.61)	3.8 (0.79)	4.16 (0.79)	0.0002
Realism of the model (mean score [SD])	4.33 (0.4)	4.14 (0.57)	4.25 (0.83)	0.916
3D neonatal thorax model (working environment) (mean score [SD])	4.66 (0.51)	4.28 (0.95)	4.5 (0.71)	0.07
Proximal esophageal pouch caliber (mean score [SD])	4.55 (0.69)	3.71 (0.75)	4.18 (0.78)	0.003
Distal esophageal pouch caliber (mean score [SD])	4.44 (0.70)	3.57 (0.53)	4.06 (0.78)	0.035
TEF caliber (mean score [SD])	4.22 (1.15)	3.28 (0.48)	3.81 (1.05)	0.002
Haptic feedback on materials (mean score [SD])	4.13 (1.03)	3.77 (0.75)	3.97 (0.93)	0.05
General appearance (mean score [SD])	4.33 (1.07)	4 (0.75)	4.17 (0.94)	0.015
3D neonatal thorax model (working environment) (mean score [SD])	4.33 (0.84)	4.14 (0.89)	4.25 (0.84)	0.285
Proximal esophageal pouch (mean score [SD])	3.88 (1.23)	3.57 (0.78)	3.75 (1.04)	0.035
Distal esophageal pouch caliber (mean score [SD])	3.88 (1.10)	3.57 (0.78)	3.75 (0.97)	0.003
TEF caliber (mean score [SD])	4.22 (0.82)	3.57 (0.53)	3.93 (0.79)	0.836
Content validity (mean score [SD])	4.52 (0.89)	4.28 (0.78)	4.41 (0.84)	0.003
Value and suitable as a training tool (mean score [SD])	4.77 (0.67)	4.57 (0.54)	4.68 (0.66)	0.264
TEF dissection and ligation (mean score [SD])	4.33 (0.96)	4.12 (0.89)	4.25 (0.91)	0.090
EA anastomosis (mean score [SD])	4.44 (1.05)	4.14 (0.89)	4.31 (0.97)	0.039
Overall impression (mean score [SD])	4.70 (0.31)	4.62 (0.53)	4.66 (0.43)	0.07
Useful as simulator (mean score [SD])	4.88 (0.31)	4.57 (0.54)	4.75 (0.44)	0.061
Useful as thoracoscopic EA with TEF training tool (mean score [SD])	4.66 (0.51)	4.57 (0.53)	4.63 (0.51)	1.000
Useful as training and educational tool (mean score [SD])	4.55 (0.51)	4.71 (0.48)	4.62 (0.49)	0.529

TABLE 3 Model validation: results from the Likert Scale Survey (n = 17).

systems integrated within a simulator environment (20, 21). These metrics are valuable and can be readily evaluated through simulators. However, these assessments are cumulative in nature, aiding trainees in developing a tangible comprehension of their technical skills (22). Subjective validation methods involve gathering participant opinions, whereas objective measures are commonly employed in experimental studies. Subjective methods typically include face, content, referent, and expert validity, requiring participants to complete surveys about their experience with the model. On the other hand, objective approaches encompass construct, discriminative, concurrent, criterion, and predictive validity (23). It is widely acknowledged that simulators must undergo validation prior to their successful integration into educational initiatives. Wells et al. documented the development of a fully synthetic simulator for Thoracoscopic Esophageal Atresia/Tracheo-Esophageal Fistula. They noted that with each design iteration, the model's fidelity and validity improved. In their final version of the simulator, the recorded average scores were as follows: value as a training tool (4.8), relevance (4.6), physical attributes (4.5), realism of material (4.25), realism experience (4.17), and ability to perform tasks (3.77) (24). "We employed a commonly used 5-point Likert scale for the validation of our model. The calculated mean overall impression score was 4.6, indicating a generally positive perception across all participants. However, there was no statistically significant difference observed between the study groups concerning this overall impression score.

Conversely, we identified a significant disparity between the study groups in terms of face validity and content validity. This discrepancy underscores that the groups displayed contrasting views regarding the relevance and accuracy of the evaluation.

In summary, although no substantial divergence was noted between the study groups in relation to the mean overall impression score, we did observe significant variations concerning face validity and content validity. These findings provide valuable insights into how the study groups perceived the evaluation and its alignment with its intended purpose. The model received favorable assessments from a group of young and enthusiastic pediatric surgeons, as well as medical students with an interest in enhancing their minimally invasive surgical skills. High ratings were assigned to its usefulness as both a simulator and an educational tool for training purposes. The hands-on session was deemed very beneficial, with strong recommendations from the participants to other pediatric surgeons. The results from face and content validity assessments suggest that our 3D inanimate model of EA/ TEF is likely to be well-received by both experts and trainees as an effective training tool. It is recommended that future studies delve into the model's training capabilities. Given its cost-effectiveness, reusability, and easy replacement for each participant, our model appears to hold significant potential as a thoracoscopic training device for surgeons. Simulation-based assessment is a crucial aspect of surgery, aiding in the acquisition of new skills and the achievement of improved surgical outcomes. The OSATS score facilitates a detailed analysis of surgical success (25, 26). The tool in question has achieved widespread usage and seeks to minimize the biases associated with subjective performance assessment. In this study, the tool was adapted to focus on specific aspects including dexterity, task flow, spatial orientation, needle handling, instrument movement, sliding knots performing, MIS skills and overall thoracoscopic skills. The results obtained from various participants exhibited similarities, but the most experienced surgeon, who had accumulated more hours of minimally invasive surgical training and conducted more monthly laparoscopic procedures, achieved the highest outcome.

"While a trend was observed for experts to achieve higher scores than novices and intermediates in each task, this difference did not attain statistical significance. This lack of significance is likely attributed to the restricted training on simulators within the

experienced group." There was a statistically significant difference in OSATS scores for the "flow of task" and scores for the "MIS skills" task distinguishing between novice and intermediates and experts. Nair et al. developed a fully synthetic 3D printed neonatal thoracoscopic simulator and aimed to establish its construct validity. By employing the OSATS score, they assessed three thoracoscopic tasks: ring transfer, needle pass, and incision of a blind upper esophageal pouch (EA cut). Within the group of 23 participants, these tasks were particularly effective in distinguishing between novices and experts, as well as novices and intermediates. However, the differentiation between intermediates and experts was not as pronounced. It's plausible that 'intermediate' surgeons already possess a strong spatial awareness and a substantial level of skill in needle manipulation. It could be reasonable to assume that more complex tasks, such as intracorporeal suturing, might be better suited for distinguishing between intermediate and expert surgeons (18). Several models for training in minimally invasive surgery (MIS) esophageal atresia (EA) repair have been documented in the literature (13). The majority of these models are inanimate, but there is an exception with the model described by Barsness et al., which involved an inanimate casing combined with fetal bovine tissue to replicate the organs involved in EA with tracheoesophageal fistula (TEF) repair (27, 28). Maricic et al. developed an inanimate and affordable model for training in minimally invasive surgery (MIS) for repairing esophageal atresia with tracheoesophageal fistula (EA/TEF). They found a correlation between the surgeon's prior experience and their performance in the model. This correlation was observed in terms of factors such as operating time, quality of anastomosis, and peripheral tissue damage (29). Barness et al. developed computer-aided drawings (CAD) to design a synthetic and appropriately sized model for esophageal atresia with tracheoesophageal fistula (EA/TEF). Their study concluded that this simulator can effectively train pediatric surgeons, particularly those in the early stages of their learning curve, in performing thoracoscopic EA/TEF repairs.

Nevertheless, our study does possess certain limitations. Primarily, the model was evaluated by a relatively small number of participants, a common concern observed in trials involving pediatric simulators. This issue is emphasized in the systematic review of simulation in pediatric surgery (30). Another limitation to acknowledge is that the study was conducted exclusively at a single center. Although this was sufficient to establish statistical significance between the groups, it should be noted that the experienced surgeon group was more accustomed to open EA/TEF procedures. the experienced surgeon group was more accustomed to open EA/TEF procedures. In the intermediate group, none of the participants had prior experience with thoracoscopic TEF repair in the past. In the expert group, 2 experts performed thoracoscopic TEF repair in the past, and all had experience with advanced minimally invasive surgery (MIS) in children and open EA/TEF procedure.

In conclusion, through a process of continuous improvement, the study successfully showcased the value of a high-fidelity simulation model for thoracoscopic esophageal atresia/ tracheoesophageal fistula repair. Participants from various expertise levels, including medical students, trainees, and experts, found the simulator to be realistic and relevant for pediatric surgical training. The study also identified areas where the model could be enhanced, and these areas will be the focal points for future refinements of the synthetic EA/TEF repair simulator.

## 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 the National Institute of Children's Diseases Bratislava Ethics Committee, Slovak Republic. The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

## Author contributions

PZ: Data curation, Formal Analysis, Resources, Writing – original draft, Writing – review & editing. JB: Formal Analysis, Methodology, Supervision, Validation, Writing – review & editing. RP: Methodology, Resources, Writing – review & editing. MS: Methodology, Supervision, Writing – review & editing. PV: Methodology, Visualization, Writing – original draft. ML: Methodology, Visualization, Writing – review & editing. TB: Methodology, Visualization, Writing – original draft. BN: Methodology, Visualization, Writing – original draft. Writing – review & editing.

## Funding

The author(s) declare financial support was received for the research, authorship, and/or publication of this article.

This work was supported by the Ministry of Education, Science, Research and Sport of the Slovak Republic (grant No. 1/0564/23).

#### Acknowledgments

The authors would like to thank the RADIX MEDICAL Ltd. team (exclusive distributor of KARL STORZ in Slovak republic) and Zbazy, s.r.o., supporting education in healthcare and medicine (exclusive distributor of LAPARO Sp. z o.o. Poland).

## 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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated

### References

1. Son J, Jang Y, Kim W, Lee S, Jeong JS, Lee S-K, et al. Thoracoscopic repair of esophageal atresia with distal tracheoesophageal fistula: is it a safe procedure in infants weighing less than 2000 g? *Surg Endosc*. (2021) 35:1597–601. doi: 10.1007/s00464-020-07538-z

2. Lepore B, Autorino G, Coppola V, Borgogni R, Carulli R, Cerulo M, et al. Effectiveness of a laparoscopic training model for pediatric surgeons during COVID-19 pandemic: a single-center experience. J Laparoendosc Adv Surg Tech A. (2022) 32:907–12. doi: 10.1089/lap.2022.0158

3. Cook DA, Hatala R, Brydges R, Zendejas B, Szostek JH, Wang AT, et al. Technology-enhanced simulation for health professions education: a systematic review and meta-analysis. *JAMA*. (2011) 306:978–88. doi: 10.1001/jama.2011.1234

4. Torres A, Inzunza M, Jarry C, Serrano F, Varas J, Zavala A. Development and validation of a new laparoscopic endotrainer for neonatal surgery and reduced spaces. *Arq Bras Cir Dig.* (2021) 33:e1559. doi: 10.1590/0102-672020200004e1559

5. Dawe SR, Pena GN, Windsor JA, Broeders JAJL, Cregan PC, Hewett PJ, et al. Systematic review of skills transfer after surgical simulation-based training. *Br J Surg.* (2014) 101:1063-76. doi: 10.1002/bjs.9482

6. Macdonald AL, Haddad M, Clarke SA. Learning curves in pediatric minimally invasive surgery: a systematic review of the literature and a framework for reporting. *J Laparoendosc Adv Surg Tech A*. (2016) 26:652–9. doi: 10.1089/lap.2016.0193

7. Varas J, Mejía R, Riquelme A, Maluenda F, Buckel E, Salinas J, et al. Significant transfer of surgical skills obtained with an advanced laparoscopic training program to a laparoscopic jejunojejunostomy in a live porcine model: feasibility of learning advanced laparoscopy in a general surgery residency. *Surg Endosc.* (2012) 26:3486–94. doi: 10.1007/s00464-012-2391-4

8. Griswold S, Ponnuru S, Nishisaki A, Szyld D, Davenport M, Deutsch ES, et al. The emerging role of simulation education to achieve patient safety: translating deliberate practice and debriefing to save lives. *Pediatr Clin North Am.* (2012) 59:1329–40. doi: 10.1016/j.pcl.2012.09.004

9. Gause CD, Hsiung G, Schwab B, Clifton M, Harmon CM, Barsness KA. Advances in pediatric surgical education: a critical appraisal of two consecutive minimally invasive pediatric surgery training courses. *J Laparoendosc Adv Surg Tech A*. (2016) 26:663–70. doi: 10.1089/lap.2016.0249

10. Badash I, Burtt K, Solorzano CA, Carey JN. Innovations in surgery simulation: a review of past, current and future techniques. *Ann Transl Med.* (2016) 4:453. doi: 10. 21037/atm.2016.12.24

11. Azzie G, Gerstle JT, Nasr A, Lasko D, Green J, Henao O, et al. Development and validation of a pediatric laparoscopic surgery simulator. *J Pediatr Surg.* (2011) 46:897–903. doi: 10.1016/j.jpedsurg.2011.02.026

12. Nair D, Wells JM, Cook N, Moorhead A, Beasley SW. Critical design and validation considerations for the development of neonatal minimally invasive surgery simulators. *J Pediatr Surg.* (2019) 54:2448–52. doi: 10.1016/j.jpedsurg.2019.05.022

13. Joosten M, de Blaauw I, Botden SM. Validated simulation models in pediatric surgery: a review. J Pediatr Surg. (2022) 57:876–86. doi: 10.1016/j.jpedsurg.2022.06.015

14. Massoth C, Röder H, Ohlenburg H, Hessler M, Zarbock A, Pöpping DM, et al. High-fidelity is not superior to low-fidelity simulation but leads to overconfidence in medical students. *BMC Med Educ.* (2019) 19:29. doi: 10.1186/s12909-019-1464-7

15. Bethlehem MS, Kramp KH, van Det MJ, ten Cate Hoedemaker HO, Veeger NJGM, Pierie JPEN. Development of a standardized training course for laparoscopic procedures using Delphi methodology. *J Surg Educ.* (2014) 71:810–6. doi: 10.1016/j.jsurg.2014.04.009

organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

16. Keeley FX, Eden CG, Tolley DA, Joyce AD. The British Association of Urological Surgeons: guidelines for training in laparoscopy. *BJU Int.* (2007) 100:379–81. doi: 10. 1111/j.1464-410X.2007.06902.x

17. Esposito C, Escolino M, Saxena A, Montupet P, Chiarenza F, De Agustin J, et al. European Society of Pediatric Endoscopic Surgeons (ESPES) guidelines for training program in pediatric minimally invasive surgery. *Pediatr Surg Int.* (2015) 31:367–73. doi: 10.1007/s00383-015-3672-5

 Nair D, Wells JM, Cook N, Yi M, Scott V, Beasley SW. Construct validation of a 3D printed neonatal thoracoscopic simulator: can it measure expertise? *J Pediatr Surg.* (2021) 56:1962–5. doi: 10.1016/j.jpedsurg.2021.03.054

19. Deie K, Ishimaru T, Takazawa S, Harada K, Sugita N, Mitsuishi M, et al. Preliminary study of video-based pediatric endoscopic surgical skill assessment using a neonatal esophageal atresia/tracheoesophageal fistula model. *J Laparoendosc Adv Surg Tech A*. (2017) 27:76–81. doi: 10.1089/lap.2016.0214

20. Oh CJ, Tripathi PB, Gu JT, Borden P, Wong BJ-F. Development and evaluation of rhinoplasty spreader graft suture simulator for novice surgeons. *Laryngoscope*. (2019) 129:344–50. doi: 10.1002/lary.27326

21. Yoshida K, Kinoshita H, Kuroda Y, Oshiro O, Matsuda T. Analysis of laparoscopic dissection skill by instrument tip force measurement. *Surg Endosc.* (2013) 27:2193–200. doi: 10.1007/s00464-012-2739-9

22. Mori T, Ikeda K, Takeshita N, Teramura K, Ito M. Validation of a novel virtual reality simulation system with the focus on training for surgical dissection during laparoscopic sigmoid colectomy. *BMC Surg.* (2022) 22:12. doi: 10.1186/s12893-021-01441-7

23. Schout BMA, Hendrikx AJM, Scheele F, Bemelmans BLH, Scherpbier AJJA. Validation and implementation of surgical simulators: a critical review of present, past, and future. *Surg Endosc.* (2010) 24:536-46. doi: 10.1007/s00464-009-0634-9

24. Wells JM, Nair D, Cook N, Yi M, Moorhead A, Maoate K, et al. End-user input into the design and validation of a synthetic thoracoscopic esophageal atresia/tracheoesophageal Fistula simulator. *J Laparoendosc Adv Surg Tech A*. (2020) 30:685–91. doi: 10.1089/lap.2019.0600

25. Cabarcas Maciá I, Marmolejo Franco F, Siu Uribe A, Palomares Garzón C, Rojo Díez R. Pilot study for low-cost model validation in laparoscopic pediatric pyeloplasty simulation. *Cir Pediatr.* (2022) 35:141–5. doi: 10.54847/cp.2022.03.18

26. Martin JA, Regehr G, Reznick R, MacRae H, Murnaghan J, Hutchison C, et al. Objective structured assessment of technical skill (OSATS) for surgical residents. *Br J Surg.* (1997) 84:273–8. doi: 10.1046/j.1365-2168.1997.02502.x

27. Barsness KA, Rooney DM, Davis LM. Collaboration in simulation: the development and initial validation of a novel thoracoscopic neonatal simulator. *J Pediatr Surg.* (2013) 48:1232–8. doi: 10.1016/j.jpedsurg.2013.03.015

28. Barsness KA, Rooney DM, Davis LM, Chin AC. Validation of measures from a thoracoscopic esophageal atresia/tracheoesophageal fistula repair simulator. *J Pediatr Surg.* (2014) 49:29–32; discussion 32–3. doi: 10.1016/j.jpedsurg.2013.09.069

29. Maricic MA, Bailez MM, Rodriguez SP. Validation of an inanimate low cost model for training minimal invasive surgery (MIS) of esophageal atresia with tracheoesophageal fistula (AE/TEF) repair. *J Pediatr Surg.* (2016) 51:1429–35. doi: 10.1016/j.jpedsurg.2016.04.018

30. Patel EA, Aydın A, Desai A, Dasgupta P, Ahmed K. Current status of simulation-based training in pediatric surgery: a systematic review. *J Pediatr Surg.* (2019) 54:1884–93. doi: 10.1016/j.jpedsurg.2018.11.019

Check for updates

#### **OPEN ACCESS**

EDITED BY Giovanni Cobellis, Marche Polytechnic University, Italy

REVIEWED BY Valerio Gentilino, Filippo Del Ponte Hospital, Italy Yener Aydin, Atatürk University, Türkiye

\*CORRESPONDENCE Chang Xu Muaxixuchang@163.com

RECEIVED 27 July 2023 ACCEPTED 06 November 2023 PUBLISHED 23 November 2023

#### CITATION

He T, Sun X, Luo D, Dai S, Yuan M, Yang G, Cheng K and Xu C (2023) Percutaneous transthoracic catheter drainage prior to surgery in treating neonates with congenital macrocystic lung malformation presenting with respiratory distress. Front. Pediatr. 11:1268028. doi: 10.3389/fped.2023.1268028

#### COPYRIGHT

© 2023 He, Sun, Luo, Dai, Yuan, Yang, Cheng and Xu. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

# Percutaneous transthoracic catheter drainage prior to surgery in treating neonates with congenital macrocystic lung malformation presenting with respiratory distress

Taozhen He<sup>1</sup>, Xiaoyan Sun<sup>2</sup>, Dengke Luo<sup>1</sup>, Shiyi Dai<sup>1</sup>, Miao Yuan<sup>1</sup>, Gang Yang<sup>1</sup>, Kaisheng Cheng<sup>1</sup> and Chang Xu<sup>1\*</sup>

<sup>1</sup>Department of Pediatric Surgery, West China Hospital of Sichuan University, Chengdu, China, <sup>2</sup>Health Management Centre, West China Hospital of Sichuan University, Chengdu, China

**Backgound:** It is rarely seen that neonates with congenital macrocystic lung malformation (CMLM) presenting with respiratory distress require emergency intervention. No consensus has been achieved concerning the best policy facing such condition. This study aims to evaluate the efficacy and safety of our strategies in treating neonates with CMLM presenting with respiratory distress.

**Methods:** We retrospectively reviewed the data of six neonates with CMLM presenting with respiratory distress from April 2020 to October 2022 for whom drainage-prior-to-surgery strategy were adopted and favorable outcomes were obtained. The relevant data was reviewed and analyzed.

**Results:** All the patients were prenatally diagnosed with congenital lung malformation and postnatally as congenital macrocystic lung malformation via CT scan. Each neonate accepted percutaneous thoracic catheter drainage prior to surgery. The first and fifth neonates with macrocystic lung mass experienced prompt open lobectomy and delayed thoracoscopic surgery due to failure of air drainage, respectively. The other four patients obtained good drainage of the large air-filled cyst, thus gaining the opportunity for elective thoracoscopic surgery within median 45 days.

**Conclusions:** For neonates with macrocystic lung malformation presenting with respiratory distress due to mediastinal compression, percutaneous thoracic catheter drainage is worth a shot for elective thoracoscopic surgery due to its feasibility and safety.

#### KEYWORDS

thoracic drainage, respiratory distress syndrome, congenital macrocystic lung malformation, cystic adenomatoid lung malformation, neonates

Abbreviations

CMLM, congenital macrocystic lung malformation; PTCD, percutaneous transthoracic catheter drainage; CPAM, congenital pulmonary airway malformation; EXIT, ex-utero intrapartum treatment; CVR, CPAM volume ratio; CLM, congenital lung malformations.

# 1. Introduction

Congenital lung malformations (CLM) are a group of anomalies that are characteristic of pulmonary abnormalities, including congenital pulmonary airway malformation (CPAM) pulmonary sequestration, bronchogenic cyst and lobar emphysema, in which CPAM accounts for the largest proportion. The majority of children diagnosed with CPAM are asymptomatic at birth, and elective pulmonary resection beyond the neonatal period is recommended and associated with favorable outcomes (1-5). In addition, there is a small group of patients with macrocystic CPAM who have early symptoms, manifesting as significant respiratory distress within the neonatal period secondary to mass effects on the heart and lungs (6-8). These neonates are supposed to undergo prompt interventions after birth. Emergency surgery was the mainstay for rescuing them despite a higher anesthetic risk (9-11). Besides, od-transthoracic puncture and drainage of the large cyst followed by elective surgery was once reported (12-14), although rare, in the literature and resulted in excellent outcomes. The latter seems to be more appropriate in stabilizing the neonates using a simple maneuver in emergency. To verify this, we presented our experiences on macrocystic CPAM neonates with respiratory distress and evaluated their safety and efficacy and attempted to delineate an algorithm based on our experiences and the literature.

#### 2. Materials and methods

#### 2.1. Study design and participants

A retrospective chart review was performed to evaluate the congenital macrocystic lung malformation (CMLM) neonates presenting with respiratory distress in the Department of Pediatric Surgery, West China Hospital, Sichuan University, from April 2020 to October 2022. The study was approved by the Research Ethics Board of Sichuan University West China Hospital. All subjects participating in the study gave written informed consent. We included prenatally diagnosed CMLM neonates presenting with progressive respiratory distress after birth due to mediastinal shift and lung compression caused by large space-occupying lesions. We excluded those with concomitant diseases that can also cause respiratory distress, such as pneumonia, congenital cardiac anomalies, and upper respiratory tract obstruction. Medical and operative records were retrieved for data analysis. Patient data including sex, weight, Apgar score, lesion size, CPAM volume ratio(CVR), timing of respiratory distress onset, age at PTCD, age at surgery, surgery type, time to discharge after surgery and complications were extracted and analyzed. Postoperative defined complications were as pneumothorax, tube dislodgement, failure to drain, broncho-pleuric fistula, persistent air leak ( $\geq$ 7 days), major bleeding, and subcutaneous emphysema.

#### 2.2. Techniques

#### 2.2.1. Seldinger maneuver

After lidocaine infiltration a pigtail tube was used through Seldinger manoeuvre, based on preoperative CT scan images. Success of the PTCD maneuver was defined as continuous drainage of trapping air from the lung cyst via the tube, and complete relief of respiratory distress was obtained (SpO2 increase to above 95% without pure O2 uptake). Failure to drain included (1) tube was placed in pleural cavity rather than in large cyst and (2) Tube was placed in the cyst not connecting to the bronchi. At this time, another attempt was reasonable. Thoracoscopic lobectomy through the hilar approach was applied in elective surgery, and thoracotomy in emergency surgery. The details of lobectomy have been described in our previous study (15)

#### 3. Results

A total of six patients were included. All of them were prenatally diagnosed with CLM and classified as MCLM after birth. They all undergo PTCD before surgery. All of the neonates presented respiratory distress (ranging mild to severe) immediately after birth. Neonates (Case 1 and 5) seemed to respond well with respiratory support in the first one to two weeks and underwent first PTCD at d 19 and d 8, respectively. The remaining patients (case 2, 3, 4, 6) presented tachypnea (60 times per min above) and the SPO2 could not be maintained above 95% by oxygen mask ventilation. Therefore, they promptly underwent PTCD on the first day after birth and obtained good drainage of the large air-trapping cyst (Figure 1), thus gaining the opportunity for elective thoracoscopic surgery in median 45 days (range from 35 to 60 days), during which lobectomy were smoothly performed. In Case 1, the neonate experienced prompt open lobectomy due to failure to place chest tube in the airtrapping cyst after two PTCD attempts (Figure 2). This patient obtained a rapid recovery with extubation on the 2nd day after surgery and was discharged home on the 9th postoperative day. In the 5th case, PTCD was performed twice with interim relief of respiratory distress (Figures 3B-D). Pneumothorax occurred after the second Seldinger maneuver, which indicated air from a ruptured cyst entered the pleural cavity (Figure 3E), thetube was removed and another one was inserted to drain the free air and partial lung expansion was obtained (Figure 3F). However, the mass effect of the macrocystic lung lesions did not seem to be alleviated. So emergency thoracoscopic lobectomy was performed on the 36th day after birth and favourable outcome was obtained.

Catheter dislodgement occurred 3 weeks after PTCD maneuver in case 2 and 3 days after PTCD in case 6, respectively. Replacement of chest tube were successfully performed and maintained for a few weeks. They all recovered well, and the relevant data are shown in **Table 1**. All patients were diagnosed with macrocystic CPAM according to the pathological findings. The follow-up (median 18 months) was uneventful. Literature concerning macrocystic CPAM neonates who underwent



#### FIGURE 1

Radiological imaging of PTCD maneuvre in case 2. (A) Preoperative CT scan showed a large cystic lung lesion in the left lower lobe; (A–C) X-ray image showed a large cystic lesion (blue arrows) in the right thoracic cavity; CT scan indicated that the cystic lesion was so large that mediastinal displacement occurred; (D) Chest film demonstrated that a pigtail catheter (yellow arrow) was successfully placed in the lesional cyst; (E) Chest X-ray showed that the lesional cyst (yellow arrow) was restored to its original size when the catheter (green arrow) dislodged out of it; (F) CT scan indicated suction catheter (orange arrow) was successfully placed into the large cyst along the existing fistula in the thoracic wall.

emergency interventions because of mass effect was reviewed and listed in Table 2.

## 4. Discussions

In fact, emergency thoracotomy worked as the most frequent intervention (4, 6, 10, 11, 13, 16–20) with a relatively higher

anesthetic risk, which, to some extent, may lead to an unfavorable outcome. Lecomte B. et al. (10) reported 10 cases of CPAM; two neonates with respiratory distress due to mediastinal shift and polyhydramnios underwent emergency surgeries and died of respiratory failure caused by infection on day 20 and by air leak on day 15, respectively. Manuela and colleagues (19) reported one neonate with disseminated bronchial crackles and productive cough who received two emergency surgeries and



FIGURE 2



died of sepsis. Elective resection in asymptomatic infants is advised to reduce the risk of postoperative complications (2). Zheng et al. (8) retrospectively reviewed 24 medical records of neonates who underwent surgery for symptomatic CPAM from 2010 to 2020 and yielded excellent clinical outcomes after symptoms of respiratory distress had adequately subsided. Moreover, elective surgery allows thoracoscopic resection, which can provide a shorter hospital stay, decreased postoperative pain and better cosmesis (1, 3). In contrast, fifteen of eighteen neonates (83.3%) from the obtained literature (including the current study) underwent emergency thoracotomy rather than the thoracoscopic approach, mainly due to the narrow thoracic space. The thoracic cavity of the neonates would enlarge significantly within couple of weeks, thus facilitating thoracoscopic surgery. In the literature we reviewed, 17 of 18 (94.4%) newborns underwent lobectomy instead of parenchyma-preserving surgery despite the great compensatory potential of the residual lung. Unfortunately, there was only one case (4) in which segmentectomy was performed, thus complicating cystic remnant. Normally, it is a great challenge for any surgeon to perform parenchyma-sparing surgery on a neonate in an emergency setting. Although no lungsparing surgery was performed in this study, PTCD was regarded as our preference for these neonates due to lower anesthetic risk

and the latent benefits of elective surgery (thoracoscopic approach and lung-preserving possibility). As a result, PTCD and surgery was considered to be a better alternative to emergency thoracotomy (2), which was supported by this case series.

In the literature, PTCD was performed on a clinical basis with CT image navigation (13, 14) or under fluoroscopic guidance (20). We considered it adequate for a skilled surgeon to perform this maneuver based on CT images We looked back to case 1, in which the PTCD maneuver should be deemed as failure, for the catheter seemed to be placed outside of the cyst (probably inside the pleural cavity, see Figures 2B,C). Therefore, to raise the success rate of PTCD, radiological confirmation of tube placement is mandatory.

Indeed, PTCD of a lung cyst is different from thoracocentesis to some degree. Most likely, the puncture needle can easily penetrate the cyst wall. However, the pigtail tube could fail to advance along the guidewire because the cystic wall is, unlike the thoracic wall, elastic and unfixed while it advances. Retrospectively, the failure of PTCD in case 1 could be attributed to this explanation. The puncture site, angle and adequate depth of advancement are of great importance. To avoid complications pertaining to lung injury, the puncture point should be a proper surface site to which the target cyst wall is mostly adjacent with the least lung parenchyma



#### FIGURE 3

Radiological imaging of PTCD maneuvre in case 5. (A) Preoperative CT scan showed a large cystic lung lesion in the left lower lobe, during which an liquid-air level was noted; (B) and (C) The first and second post-PTCD radiological image both displayed little relief of mass effect, respectively; (D) CT scan indicated the lung lesion was multicystic and could not be completely decompressed by the pigtail catheter (yellow arrow); (E) CT scan shown pneumothorax occurred secondary to the 2nd PTCD; F: Chest X-ray indicated the air in the thoracic cavity was drained via catheter adjustment.

interposition between the inner thoracic wall and the cyst. The needle should be perpendicular to the cyst surface. Oh SH et al. (20) reported complications of PTCD in seven cases, including pneumothorax (n = 2), catheter displacement (n = 1) and failure to drain (n = 1). They reported pneumothorax occurred due to use of a non-pigtail catheter (chest tube or angiocatheter). Similar to the complications of PTCD reported above, Pneumothorax (n = 1), catheter displacement (n = 2), failure to drain (n = 2) and broncho-pleuric fistula (n = 0) occurred in PTCD using pigtail catheter in the current study. We noted that failing to drain both occurred in the two cases (case 1 and case 5), the former case had faster

postoperative recovery than the latter one (time to discharge home after surgery, 9 days vs. 17 days) even though open surgery was performed in case 1. Maybe the duration of respiratory distress prior to surgery (23 days vs. 36 days) could explain. We assume that the shorter the duration of respiratory distress was, the less impairment to the remaining lung was, the faster the neonate recovered, which needs to be verified in further study.

Maintaining the chest tube after discharge needs careful parental care to avoid a significant number of severe complications. In this study, 2 babies experienced tube dislodgement, thus causing recurrence of respiratory distress.

Death	No	No	No	No	No		No
(mo)	16	18	20	25	9		33
Complication	Failure to drain	Tube dislodgement	None	None	Failure to drain,	Pneumothorax	Tube dislodgement, Pneumothorax
TDAS (d)	6	8	2	8	20		4
Surgery type	OL	TL	TL	TL	TL		TL
Age at surgery	23	60	35	48	36		42
CVR TRDO Age at PTCD (d)	19/22	1	1	1	8/18		ŝ
TRDO	IAB	IAB	IAB	IAB	IAB		IAB
	1.7	4.5	3.7	2.5	2.1		2.4
Lesion size (cm)	4.8	6.5	5.5	6.0	5.3		5.4
Apgar score (1 min, 5 min)	9, 10	9, 10	8.9	9.10	10.10		10.10
GA (wk)	38	36	38	38	39		39
Year Birth weight (g)	3,550	3,160	3,325	3,150	4,130		3,250
Year	2020	2020	2021	2021	2022		2022
Case No		2		4	5		6

time to discharge after surgery; PTCD, percutaneous transthoracic catheter drainage; CVR, CPAM volume ratio; TRDO, timing of respiratory distress onset; IAB, immediately after birth TL, thoracoscopic lobectomy; LUL, left lower lobe; RLL, right lower lobe; RUL, right upper lobe; LLL, left lower lobe; FU, follow . congenital lung mass; GA, gestational age; TDAS, open lobectomy; CLM, оĽ

Fortunately, the respiratory symptom relieved again by reinserting another tube along the external opening of the fistula at the thoracic wall, which lasted couple of weeks prior to surgery. To a certain degree, we overestimated the parents' nursing abilities in securing the chest tube at home for a long time (4–8 weeks).

Although Oh SH, et al. (20) reported that the mean time between PCTD and surgery was 4 days only, they all underwent open lobectomies, which meant greater trauma and potential long-term complication such as chest wall deformity and scoliosis. Narrow thoracic space is an obstacle of thoracoscopic surgery, therefore, we expect the babies to gain more weight, greater thoracic cavity and more mature lung (better tolerating the anesthetic risk), thus facilitating the thoracoscopic surgery. Just as one coin has two sides, although the babies staying at home with chest tube in the body take more risk (hemothorax or dislodgement of chest tube) to a certain extent, the time to discharge home in the current study is shorter (median 8 days vs. 13 days). Beside, the parents strongly requested for thoracoscopic surgery for their babies after full consideration. As such, to decrease the take-tubehome risk, several conditions should be met. Firstly, the parents' compliance should be good enough to master some nursing techniques. Secondly, the patients' home are not far away from the clinics or hospitals where professional medical care are available in emergency. Thirdly, the time of staying home with tube-in-the-body should not be within 1 month depending on the parents' compliance.

It is worth mentioning that Cass DL and colleagues (21) reported that nine prenatally diagnosed CLM newborns comprised of five cystic masses (all macrocystic CPAM) and four solid masses with persistent mediastinal compression treated with ex-utero intrapartum treatment (EXIT) were well discharged at a median of 10 d postoperatively. Similar to the strategy we adopted, EXIT is, to some degree, equivalent to an elective (prophylactic) surgery with full preparation for neonatal delivery and establishment of the respiratory tract prior to dissection of the umbilical cord, thus preventing neonatal respiratory distress. EXIT-to-resection However. conducting an requires multidisciplinary cooperation, which is generally integrated into a tertiary medical center with advanced equipment and specialists, which is not suitable for primary-level medical institutions. In other words, the PTCD procedure could be deemed a first-aid intervention in basic medical units with a higher benefit-risk ratio. Based on our experiences and the literature, an algorithm of neonatal CMLM with respiratory distress was delineated (Figure 4), which could be referenced by others.

Several caveats regarding this study should be acknowledged. Firstly, this was a retrospective, single institution series with a relatively short follow-up period (median 18 months). Secondly, the study population was rather small due to the low morbidity rate. Thirdly, a lack of control will no doubt degrade the evidence level of this study. Last but not least, since the majority of neonatal units' standard policy is to remove the chest tube before discharge, the clinical algorithm mentioned in this study can not be referenced in many low and middle income countries. Despite these limitations, this was the first paper that exclusively

TABLE 1. Characteristics of CMLM neonates undergoing emergency PTCD and/or surgery in current study.

Death	No	No	Yes <sup>f</sup>	Yes <sup>g</sup>	No	No	No	No		No	No	No
Complication	U	n	Air leak $(n = 1)$ , Infection $(n = 1)$	Sepsis	None	n	pneumothorax	Cystic remnant $(n = 1)$		None	None	See Table 1
TDAS (d)	7	12	None	n	21	13 (1-26.5)	12	15.5	(14.75 - 16)	7	16	7.5 (5.5–8.0)
Surgery type	TO	IO	TO	OPLR	TO	TO	TO	OSe		TO	ΤΓ	OL $(n = 1)$ , TL $(n = 5)$
Lesion size (cm) Age at Surgery (d)	6	1	1/14	7	24	4 (1-12)	19	5.5	(3.25 - 7.25)	21	4	39.0 (35.25–46.5)
Lesion size (cm)	4.0	n	n	n	U	5.6 (4.2-6.9)	U	U		U	5.7	5.45 (5.325,5.875)
Apgar score (1 min, 5 min)	6, 8	9,9	~ <	n	7, 9	6 (4.5–8), 8 (7–9)	n	n		U	n	9 (9,9.75), 10 (10,10)
Birth weight (g)	3,525	4,040	C	3,900/2,600	1,590	2,710 (2,304-3,000)	U	2,850	(2,500-3,225)	3,100	2,499	3,287.5 (3,182.5 -3,493.75)
Year Author Gestational age (wk) Birth weight (g)	37	37	Mean 40	37/35.5	31	37.4 (34.6-39.1)	n	n		U	39	38 (38–38.75)
Author	David	Allegaert	Lecomte <sup>a</sup>	Manuela	Chong	Seong <sup>b</sup>	Makhijani	Ito <sup>c</sup>		Disu	Huang	He <sup>d</sup>
Year	1994	2002	2009	2014	2016	2017	2018	2019		2019	2020	2022
Case no	1	2	3	4	5	6	7	8		6	10	11

TABLE 2 Characteristics of CMLM neonates undergoing emergency intervention due to mass effect in the literature.

Frontiers in Pediatrics

lower lobe; RUL, right upper lobe; LLL, left lower lobe; R, right; L, left; OPLR, open partial lung resection; U, unmentioned. <sup>a</sup>Two cases included.

<sup>b</sup>Seven case series. <sup>c</sup>Four case included.

<sup>d</sup>The current study.

<sup>e</sup>Open surgery including lobectomy, segmentectomy, wedge resection and fractionated lung resection. <sup>f</sup>Died of respiratory failure caused by infection on day 20 and by air leak on day 15, respectively. <sup>9</sup>Died of sepsis.



delineated an algorithm of prenatally diagnosed CMLM that may cause respiratory distress after birth. Case control study with larger sample size is anticipated in the future.

# 5. Conclusions

For neonates with macrocystic lung malformation presenting with respiratory distress due to mediastinal compression, percutaneous thoracic catheter drainage is worth a shot for elective thoracoscopic surgery in selected cases, which is deemed as a safer alternative to direct emergency thoracotomy.

## 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 Research Ethics Board of Sichuan University West China Hospital. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation in this study was provided by the participants' legal guardians/next of kin. Written informed consent was obtained from the individual(s), and minor(s)' legal guardian/next of kin, for the publication of any potentially identifiable images or data included in this article.

## Author contributions

TH: Writing – original draft. XS: Conceptualization, Data curation, Writing – review & editing. DL: Data curation, Investigation, Writing – review & editing. SD:

Conceptualization, Formal Analysis, Writing – original draft. MY: Data curation, Software, Writing – review & editing. GY: Validation, Writing – review & editing. KC: Methodology, Software, Writing – original draft. CX: Supervision, Writing – review & editing.

## Funding

The author(s) declare financial support was received for the research, authorship, and/or publication of this article.

CX was granted by the National Natural Science Foundation of China (NO.31201095) and Natural Science Foundation of Sichuan Province (NO.2022NSFSC0354)

## References

1. Rothenberg SS. First decade's experience with thoracoscopic lobectomy in infants and children. J Pediatr Surg. (2008) 43:40–4. doi: 10.1016/j.jpedsurg.2007.09.015

2. Stanton M, Njere I, Ade-Ajayi N, Patel S, Davenport M. Systematic review and meta-analysis of the postnatal management of congenital cystic lung lesions. *J Pediatr Surg.* (2009) 44:1027–33. doi: 10.1016/j.jpedsurg.2008.10.118

3. Singh R, Davenport M. The argument for operative approach to asymptomatic lung lesions. *Semin Pediatr Surg.* (2015) 24:187–95. doi: 10.1053/j.sempedsurg.2015.02.003

4. Ito A, Takao M, Shimamoto A, Kaneda S, Matsushita K, Inoue M, et al. Introduction of thoracoscopic surgery for congenital pulmonary airway malformation in infants: review of 13 consecutive surgical cases. *J Thorac Dis.* (2019) 11(12):5079–86. doi: https://doi: 10.21037/jtd.2019.12.14

5. Annunziata F, Bush A, Borgia F, Raimondi F, Montella S, Poeta M, et al. Congenital lung malformations: unresolved issues and unanswered questions. *Front Pediatr.* (2019) 7:239. doi: 10.3389/fped.2019.00239

6. Sherer DM, Kopelman JN, Rawlings JS, Degnan JE, Schaller RT. Congenital cystic adenomatoid malformation of the lung with prolonged marked lung compression and neonatal survival. *J Clin Ultrasound*. (1994) 22:515–8. doi: 10.1002/jcu.1870220811

7. Kevin NJ, Rodrigo AM, Samir KG, Shaun MK. Short-term respiratory outcomes of neonates with symptomatic congenital lung malformations. *J Pediatr Surg.* (2019) 54:1766–70. doi: 10.1016/j.jpedsurg.2019.01.056

8. Zheng J, Tang H, Xu H, Li J, Mao X, Liu G. Thoracoscopic versus open resection for symptomatic congenital pulmonary airway malformations in neonates: a decade-long retrospective study. *BMC Pulm Med.* (2021) 21:82. doi: 10.1186/s12890-021-01445-2

9. Seo T, Ando H, Watanabe Y, Harada T, Ito F, Kaneko K, et al. Acute respiratory failure associated with intrathoracic masses in neonates. *J Pediatr Surg.* (1999) 34:1633–7. doi: 10.1016/S0022-3468(99)90632-2

10. Lecomte B, Hadden H, Coste K, Gallot D, Laurichesse H, Lemery D. Hyperechoic congenital lung lesions in a non-selected population: from prenatal detection till perinatal management. *Prenat Diagn.* (2009) 29:1222–30. doi: 10.1002/pd.2407

11. Makhijani AV, Wong FY. Conservative postnatal management of antenatally diagnosed congenital pulmonary airway malformations. *J Paediatr Child Health.* (2018) 54:267–71. doi: 10.1111/jpc.13727

## 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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

12. Wallis C. Clinical outcomes of congenital lung abnormalities. *Paediatr Respir Rev.* (2000) 1:328-35. doi: 10.1053/prrv.2000.0072

13. Allegaert K, Proesmans M, Naulaers G, Moerman P, Lerut T, Devlieger H. Neonatal transthoracic puncture in a case of congenital cystic adenomatoid malformation of the lung. *J Pediatr Surg.* (2002) 37:1495–7. doi: 10.1053/jpsu.2002.35430

14. Margau R, Amaral JG, Chait PG, Cohen J. Percutaneous thoracic drainage in neonates: catheter drainage versus treatment with aspiration alone. *Radiology*. (2006) 241:223-7. doi: 10.1148/radiol.2411050966

15. Zhang Y, Xin W, Xu C, Yuan M, Yang G, Cheng K. Thoracoscopic lobectomy through the pulmonary hilum approach for the treatment of congenital lung malformation. *Surg Endosc.* (2022) 36:711–7. doi: 10.1007/s00464-021-08339-8

16. Manuela C, Marta S, Raluca M, Luminita Z. Chest 2014;145 (3\_MeetingAbstracts): 451A (1-2). doi: https://doi: 10.1378/chest.1821588

17. Chong Y, Rhee Y, Han S, Cho H, Kang S, Kang M. Life-Threatening congenital cystic adenomatoid malformation in the premature neonate. *Korean J Thorac Cardiovasc Surg.* (2016) 49:210–3. doi: 10.5090/kjtcs.2016.49.3.210

18. Disu EA, Kehinde OA, Anga AL, Ubuane PO, Itiola A, Akinola IJ, et al. Congenital pulmonary airway malformation: a case report of a rare cause of neonatal respiratory distress and review of the literature. *Niger J Clin Pract.* (2019) 22(11):1621–5. doi: https://doi: 10.4103/njcp.njcp\_20\_19

19. Huang J, Zhou C, Chen Q, Wu D, Hong J, Hong S. Thoracoscopic lobectomy for A 4-day-old neonate with A large congenital pulmonary airway malformation: a case report. *J Cardiothoracic Surg.* (2020) 15:159. doi: 10.1186/s13019-020-01174-3

20. Oh SH, Kim CY, Lee BS, Kim DK, Kim EA, Kim K-S. Transthoracic catheter drainage for large symptomatic congenital pulmonary airway malformation. *Pediatr Pulmonol.* (2017) 52(12):1572–7. doi: https://doi: 10.1002/ppul.23835

21. Cass DL, Olutoye OO, Cassady CI, Zamora IJ, Ivey RT, Ayres NA, et al. EXIT-to-resection for fetuses with large lung masses and persistent mediastinal compression near birth. *J Pediatr Surg.* (2013) 48:138–44. doi: 10.1016/j. jpedsurg.2012.10.067

Check for updates

#### **OPEN ACCESS**

EDITED BY Alessandro Inserra, Bambino Gesù Children's Hospital (IRCCS), Italy REVIEWED BY Carlos Delgado-Miguel,

Hospital Infantil La Paz, Spain Simone Frediani, Bambino Gesù Children's Hospital (IRCCS), Italy

\*CORRESPONDENCE Edoardo Bindi ⊠ edo.bindi88@hotmail.it

RECEIVED 15 October 2023 ACCEPTED 14 November 2023 PUBLISHED 08 December 2023

#### CITATION

Franconi I, Faragalli A, Palego G, Canonici S, Gatti L, Simonini A, Bindi E, Cobellis G and Carle F (2023) Preoperative anxiety management in children. Benefits of humanoid robots: an experimental study. Front. Surg. 10:1322085. doi: 10.3389/fsurg.2023.1322085

#### COPYRIGHT

© 2023 Franconi, Faragalli, Palego, Canonici, Gatti, Simonini, Bindi, Cobellis and Carle. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

# Preoperative anxiety management in children. Benefits of humanoid robots: an experimental study

Ilaria Franconi<sup>1</sup>, Andrea Faragalli<sup>2</sup>, Giulia Palego<sup>3</sup>, Samuele Canonici<sup>3</sup>, Ludovica Gatti<sup>1</sup>, Alessandro Simonini<sup>4</sup>, Edoardo Bindi<sup>5,6</sup>\*, Giovanni Cobellis<sup>5,6</sup> and Flavia Carle<sup>2</sup>

<sup>1</sup>Operating Room, Salesi Children's Hospital, AOU Ospedali Riuniti Ancona, Ancona, Italy, <sup>2</sup>Department of Biomedical Sciences and Public Health, Center of Epidemiology, Biostatistics and Medical Information Technology, Marche Polytechnic University, Ancona, Italy, <sup>3</sup>AOU delle Marche, Salesi Hospital Foundation Onlus, Ancona, Italy, <sup>4</sup>Pediatric Intensive Care Unit, Salesi Children's Hospital, AOU Ospedali Riuniti Ancona, Ancona, Italy, <sup>5</sup>Pediatric Surgery Unit, Salesi Children's Hospital, Ancona, Italy, <sup>6</sup>Department of Clinical Specialty Sciences and Odontostomatology, Università Politecnica of Marche, Ancona, Italy

**Objective:** The purpose of this study was to determine whether the use of a humanoid robot (Estrabot) could reduce preoperative anxiety levels in children. **Methods:** An experimental study was conducted at Azienda Ospedaliero Universitaria delle Marche Hospital, involving the Pediatric Surgery ward and the Operating Room (OR). Patients aged between 2 and 14 years who underwent minor surgery were included. The Instruments used were the *Children's Emotional Manifestation Scale* to evaluate anxiety levels, and *Estrabot*, a humanoid robot that interacts with people. Medical records between April and May 2023 were analyzed and the data was anonymous. The level of anxiety is extrapolated in Pediatric Surgery during the administration of oral pre-medication, and in the Operating Room, during the induction of anesthesia. Patients were divided into an intervention group treated with Estrabot, and a control group without a robot.

**Results:** The population consists of 60 patients (86.7% male) with a median (IQR) age of 6 (4–8) years. The median (IQR) anxiety score during premedication was 7 (5–11), while the median (IQR) anxiety score during anesthesia was 6 (5–10). A significantly lower level of anxiety was reported in the Estrabot group. Patients in the Estrabot group had significantly lower anxiety levels in different age groups.

**Conclusion:** A humanoid robot can reduce preoperative anxiety levels in children during premedication and the induction of anesthesia.

KEYWORDS

anxiety, children, humanoid robot, preoperative, pediatric surgery

## 1. Introduction

Surgery is a stressful experience for a child and his family, both from a physical and psychological point of view. It can become a traumatic event that leads the child to experience high levels of anxiety, mainly caused by the fear of being separated from their parents, the unfamiliar environment, and inadequate preoperative preparation. For these reasons, children appear tense, nervous, fearful, and agitated. The literature shows how preoperative anxiety can influence the intensity of postoperative pain and increase the induction time of anesthesia, which is known as the most stressful time for the child (1).

The management of these high levels of stress, to which the child and the family are subjected during the pre and post-operative period, represents a goal that arises in several different health professions. The nurse is certainly the most involved and suitable figure to assess and treat anxiety and to improve the preoperative experience of the patients, ensuring a quiet and reassuring hospital environment, and providing the child with the most suitable emotional support according to his degree of development (2, 3).

Non-pharmacological techniques and co-therapies have long been recognized as useful and valid tools for the control of fear and anxiety in a hospital setting, as well as for the management of procedural pain.

The American Academy of Pediatrics recommends a combination of pharmacological and non-pharmacological techniques to manage pediatric pain (4). To reduce the child's preoperative stress level, a good preoperative preparation of the young patient and their parents by the nursing staff is necessary. Furthermore, the presence of parents during the induction of anesthesia is essential. This does not make the anxious state disappear but it greatly reduces its impact on the duration of the induction itself (5). Other non-pharmacological methods have been studied with positive results, such as the use of natural sounds (6) and relaxation-guided imagery where the results obtained demonstrate the reduction of preoperative anxiety and post-operative pain (7).

The purpose of this study was to determine whether the use of a humanoid robot (Estrabot) could reduce preoperative anxiety levels in children.

# 2. Methods

#### 2.1. Study design and setting

An experimental study was conducted at *Azienda Ospedaliero Universitaria delle Marche* Hospital, involving the Pediatric Surgery ward and the Operating Room (OR).

# 2.2. Sampling

• Inclusion criteria:

Patients aged between 2 and 14 years underwent minor surgery, such as surgery for inguinal hernia, circumcision, and orchidopexy.

• Exclusion criteria:

Patients under 2 years of age or above 14 years of age, who have undergone major surgery (appendicitis, peritonitis, intestinal obstruction, invagination, abdominal trauma, chest trauma, hemoperitoneum, hemothorax, liver fracture, kidney fracture, spleen fracture, ovarian torsion, testicular torsion, neuroblastomas, ovary neoplasms, congenital megacolon, anorectal malformations) or who have undergone urgency/ emergency procedures.

## 2.3. Instruments

- Children's Emotional Manifestation Scale (CEMS), which considers five variables: facial expression, vocalization, activity, interaction, and cooperation. For each of the five variables it is possible to assign a score from 1 to 5, therefore the overall score varies from 5 to 25. A lower score corresponds to a lower level of anxiety, compared to a higher score that indicates greater anxiety.
- Estrabot: a humanoid robot that speaks in a child's voice. The model is NAO, developed by the French company Aldebaran Robotics in 2008 (acquired in 2015 by Japanese company Softbank), among the most widely used social robots in human-robot interaction, due to its affordability and broad functionality. NAO is 58 cm in height and weighs 5.6 kg; it has four directional microphones and speakers and two cameras which allow it to perform special features such as text-to-speech, speech recognition for 20 languages, object recognition, face detection and recognition, tracking, and more. Thanks to a complex system of joints, the entire body of the robot can move completely with 25 degrees of freedom: it can grab objects, move around, dance, and interact with people. A gyroscope, sensors, and a five-axis control unit provide balance during movements and exploration. It is fully programmable thanks to the included NAO Software Suite. Estrabot is the name chosen for the robot used for the activity. It has been purchased by the Salesi Foundation to implement projects in favor of children hospitalized at the Salesi Hospital in Ancona, Italy (Figure 1).

## 2.4. Data collection and analysis

The medical records were collected between April and May 2023. The level of anxiety is assessed in the Paediatric Surgery



FIGURE 1

Estrabot: an humanoid robot that speaks in a child's voice. The model is NAO, developed by the French company Aldebaran Robotics in 2008 (acquired in 2015 by Japanese company Softbank), among the most widely used social robots in human-robot interaction, due to its affordability and broad functionality.

ward during the administration of oral premedication (sublingual midazolam), and in the Operating Room, during the induction of anesthesia. Patients were randomized into an intervention group treated with Estrabot, and a control group treated without the robot, according to the day of the week.

The experimental group is composed of children operated on Friday, who met the operator of Estrabot before the standard protocol (the same day or the day before), to allow them to know the project and tell something about their preferences, hobbies, and their favorite song. Using this information, the operator can make slight changes to the program, so that it can be easier for the robot to make every child feel safe and relaxed.

On the day of surgery the robot, accompanied by its operator, enters the child's room with the nurse, explains to him in simple words the oral premedication procedure, and creates a first emotional contact by joking with him or, when possible, dancing or playing with the child and their parents.

It is then brought out of the room, before coming back when the child is accompanied to the lift which brings them to the surgery wing; here it reassures both the child and the parents and greets them. During this short journey, Estrabot plays the song chosen by the child, and when they reach the operating room it explains that before sleeping they will play inflating a balloon using a special mask; during the induction of anesthesia the robot pretends to blow in a similar mask, playing again the chosen song. As soon as the child falls asleep, both the robot and the operator quit the OR.

The control group is composed of children operated on Monday following the standard protocol without the company nor the explanations of the robot. This group of children is assisted by the ward nurse during the oral premedication, by auxiliary personnel during the journey to surgery, and by OR nurses during the induction of anesthesia. In both groups, the nurse who administered the premedication and one of those who assisted at the moment of anesthesia filled out the CEMS questionnaires during the procedures.

A non-parametric approach was followed. The characteristics of the investigated sample were summarized by absolute and percentage frequencies medians and interquartile range [IQR] for qualitative and quantitative variables respectively. To investigate the differences between groups, the non-parametric ANCOVA with smoothed regression and Young and Bowman test were applied. Two dependent variables were considered: the CEMS score evaluated during the administration of oral premedication and the CEMS score evaluated in the Operating Room during the induction of anesthesia; age was included as a covariate. One model for each dependent variable was applied. As a sensitivity analysis, the differences in CEMS scores between the two groups were stratified by 3 age classes (< 5 years, between 5 and 7 years, and >7 years) using the Mann–Whitney *U*-test. All statistical analyses were performed using R version 4.1.3.

# 3. Ethics

The Board of the teaching hospital evaluated the study. The research did not require Ethical Committee approbation because it does not involve direct medical, surgical, or pharmacological intervention on the patient. As suggested by the protocols of our hospital center, studies of this kind do not need approval by the Ethical Committee.

Study participation presents no known risks to the children and will not subject them to any additional pain or suffering. Children will receive standard medical management for their admission diagnosis.

Written informed consent to participate was obtained from all participants' parents/guardians and oral one was obtained from all children.

# 4. Results

During the study period, a total of 60 patients were included in the group, 52 (86.7%) of them were males. The median (IQR) age was 6 (4–8) years. In the whole study population, the median (IQR) anxiety score during premedication was 7 (5–11), while the median (IQR) anxiety score during anesthesia was 6 (5–10). No difference in anxiety level was found between male and female patients, both during premedication: 7 (5–9) vs. 10 (8–16), p = 0.08, and during anesthesia: 6 (5–9) vs. 9 (5–14), p = 0.26, respectively.

**Tables 1, 2** shows the comparison of anxiety levels during premedication and anesthesia between groups (Estrabot vs. Control) adjusted by age. A significantly lower level of anxiety was reported in the Estrabot group.

## 5. Discussion

To our knowledge, this is the first experimental study on the use of a humanoid robot to reduce preoperative anxiety levels in children.

TABLE 1 Anxiety levels comparison between Estrabot and Control group: results of ANCOVA models.

		Estrabot ( <i>n</i> = 30)	Control ( <i>n</i> = 30)			
	Dependent variables	Adj. median <sup>a</sup> (Cl 95%)	Adj. median <sup>a</sup> (Cl 95%) <sup>a</sup>	RSE	Coefficient	p
CESM score:						
Model 1	Premedication	6 (5-9)	12 (10-13)	4.43	16.03	0.03
Model 2	Anesthesia	6 (5-8)	10 (8-11)	3.69	9.77	0.03

Each model was adjusted for age.

<sup>a</sup>The adjusted median by smoothed regression model; CI 95%: 95% Confidence Interval; RSE: Residual Standard Error of the model.

		Age <5 years			Age 5–7 years		Age >7 years			
	Estrabot ( <i>n</i> = 11)	Control ( <i>n</i> = 12)	p	Estrabot (n = 8)	Control ( <i>n</i> = 12)	p	Estrabot ( <i>n</i> = 11)	Control ( <i>n</i> = 6)	р	
Dependent Variables	Median (Cl 95%)	Median (Cl 95%)		Median (Cl 95%)	Median (Cl 95%)		Median (Cl 95%)	Median (Cl 95%)		
CESM score:										
Premedication	8 (5; 16)	13 (7; 20)	0,052	6 (5; 7)	8 (6; 13)	0,012	5 (5; 7)	9 (7; 23)	0,001	
Anesthesia	6 (5; 13)	10 (8; 16)	0,033	5 (5; 10)	7 (5; 9)	0,143	5 (5; 6)	9 (5; 25)	0,008	

TABLE 2 Anxiety levels comparison between Estrabot and Control group according to three age groups: results of Mann-Whitney U-test.

Various distraction strategies are useful in reducing preoperative anxiety. Playing comes naturally to children and is often their favorite activity. Providing an environment conducive to play activities, and toys, or using existing handheld game technology to make the environment less threatening has been shown to reduce anxiety (8), and enhance the cooperation of children with medical procedures (9) and anesthesia induction. Ensuring the presence of parents during anesthetic induction reduces anxiety before surgery in children (10). Other studies have demonstrated that nonpharmacological techniques, such as listening to music, may benefit preoperative anxiety (11). The use of a technologically enhanced device may effectively distract children and reduce their perceived anxiety. One recent study on 57 children has demonstrated the effectiveness of child-robot interaction for reducing pain and distress during vaccination (12). A second study has shown a reduction in distress for 40 pediatric oncology patients requiring central venous access (13). Logan et al. (14) proposed to use social robots as engaging tools to address the emotional needs of hospitalized children. The children exposed to an interactive teddy bear robot intervention showed more positive affect and they expressed greater levels of joyfulness and agreeableness than the other conditions. Our results show that a non-pharmacological intervention like a humanoid robot reduces anxiety in children during the preoperative time and it might be an attractive solution to optimize perioperative care in children. As Feigal also stated in a study about non-pharmacological intervention for managing dental anxiety in children (15), behavior management needs to be flexible and individualized for each child; knowing in advance the child's preferences, the operator can adapt the Estrabot program to the patient's tastes, improving the humanoid robot's positive effect of the on the child's feelings.

A limitation of this study is the use of CEMS in adolescents, where more specific assessment scales (16, 17) should be used; CEMS was the tool routinely used in the study hospital.

Furthermore, we did not measure the anxiety level in children undergoing major surgery and in the parents. Future studies will be necessary to confirm our experimental results in children and adolescents using appropriate anxiety assessment tools and considering also major surgery, Parents' anxiety status should be also analyzed. Therefore, the effectiveness of the humanoid robot and its determinants will have to be evaluated by performing observational studies in real clinical practice.

# 6. Conclusion

A humanoid robot can reduce preoperative anxiety levels in children during premedication and the induction of anesthesia.

# Data availability statement

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

## Author contributions

IF: Writing – original draft. AF: Writing – review & editing. GP: Writing – review & editing. SC: Writing – review & editing. LG: Writing – review & editing. AS: Writing – review & editing. EB: Visualization, Writing – review & editing. GC: Writing – review & editing. FC: Writing – review & editing.

## Funding

The author(s) declare that no financial support was received for the research, authorship, and/or publication of this article.

# Acknowledgment

A grant from the Salesi Hospital Foundation supported the present work. The authors are grateful to Laura Mazzanti, Salesi Hospital Foundation Director, for helpful suggestions and Monica Glebocki for language revision.

# **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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated

## References

1. Draskovic B, Simin JM, Kvrgic IM. Psychological aspects of pediatric anesthesia. Med Pregl. (2015) 68(1-2):29-34. doi: 10.2298/MPNS1502029D

2. Dreger VA, Tremback TF. Management of preoperative anxiety in children. AORN J. (2006) 84(5):778-80. doi: 10.1016/s0001-2092(06)63965-3

3. Clair C, Engström Å, Strömbäck U. Strategies to relieve patients' preoperative anxiety before anesthesia: experiences of nurse anesthetists. *J Perianesth Nurs*. (2020) 35(3):314–20. doi: 10.1016/j.jopan.2019.10.008

4. Coté CJ, Wilson S, American academy of pediatrics, American academy of pediatric dentistry. Guidelines for monitoring and management of pediatric patients before, during, and after sedation for diagnostic and therapeutic procedures: update 2016. *Pediatric*. (2016) 138(1):e1-e31. doi: 10.1542/peds.2016-1212

5. Hussain A, Khan FA. Effect of two techniques of parental interaction on children's anxiety at induction of general anaesthesia-a randomized trial. *Turk J Anaesthesiol Reanim.* (2018) 46(4):305–10. doi: 10.5152/tjar.2018.66750

6. Amiri MJ, Sadeghi T, Negahban Bonabi T. The effect of natural sounds on the anxiety of patients undergoing coronary artery bypass graft surgery. *Perioper Med* (*Lond*). (2017) 6:17. doi: 10.1186/s13741-017-0074-3

 Vagnoli L, Caprilli S, Messeri A. Parental presence, clowns or sedative premedication to treat preoperative anxiety in children: what could be the most promising option? *Paediatr Anaesth.* (2010) 20(10):937–43. doi: 10.1111/j.1460-9592.2010.03403.x

8. Patel A, Scheible T, Davidson M, Tran MC, Schoenberg C, Delphin E, et al. Distraction with a hand-held video game reduces pediatric preoperative anxiety. *Paed Anesth.* (2006) 16:1019–27. doi: 10.1111/j.1460-9592.2006.01914.x

9. Denman WT, Tuason PM, Ahmed MI, Brennan LM, Cepeda S, Carr DB. The PediSedate device, a novel approach to pediatric sedation that provides distraction

organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

and inhaled nitrous oxide: clinical evaluation in a large case series. Paed Anesth. (2007) 17(2):162-6. doi: 10.1111/j.1460-9592.2006.02091.x

10. Velayos M, Estefanía K, Álvarez M, Sarmiento MC, Moratilla L, Sanabria P, et al. Healthcare staff as promoters of parental presence at anesthetic induction: net promoter score survey. *World J Clin Pediatr.* (2021) 10(6):159–67. doi: 10.5409/wjcp.v10.i6.159

11. Bradt J, Dileo C, Shim M. Music interventions for preoperative anxiety. *Cochrane Database Syst Rev.* (2013) 6(6):CD006908. doi: 10.1002/14651858.CD006908.pub2

12. Beran TN, Ramirez-Serrano A, Vanderkooi OG, Kuhn S. Reducing children's pain and distress towards flu vaccinations: a novel and effective application of humanoid robotics. *Vaccine*. (2011) 31(25):2772–7. doi: 10.1016/j.vaccine.2013.03.056

13. Jibb LA, Birnie KA, Nathan PC, Beran TN, Hum V, Victor JC, et al. Using the MEDiPORT humanoid robot to reduce procedural pain and distress in children with cancer: a pilot randomized controlled trial. *Pediatr Blood Cancer*. (2018) 65(9):e27242. doi: 10.1002/pbc.27242

14. Logan DE, Breazeal C, Goodwin MS, Jeong S, O'Connell B, Smith-Freedman D, et al. Social robots for hospitalized children. *Pediatrics.* (2019) 144(1):e20181511. doi: 10.1542/peds.2018-1511

15. Feigal RJ. Guiding and managing the child dental patient: a fresh look at old pedagogy. *J Dent Educ*. (2001) 65:1369–77. doi: 10.1002/j.0022-0337.2001.65.12. tb03496.x

16. Behrens B, Swetlitz C, Pine DS, Pagliaccio D. The screen for child anxiety related emotional disorders (SCARED): informant discrepancy, measurement invariance, and test-retest reliability. *Child Psychiatry Hum Dev.* (2019) 50(3):473–82. doi: 10.1007/s10578-018-0854-0

17. Spence SH. A measure of anxiety symptoms among children. *Behav Res Ther.* (1998) 36(5):545–66. doi: 10.1016/s0005-7967(98)00034-5

Check for updates

#### **OPEN ACCESS**

EDITED BY Giuseppe Lauriti, G. d'Annunzio University of Chieti and Pescara, Italy

REVIEWED BY Jun Fujishiro, The University of Tokyo, Japan Heba Taher, Cairo University, Egypt

\*CORRESPONDENCE

Li Zhang ⊠ chengyedi@163.com

RECEIVED 22 September 2023 ACCEPTED 21 December 2023 PUBLISHED 08 January 2024

#### CITATION

Zhang L, Zhang R, Zhang J, Hu H, Chen Z, Fu Y and Li S (2024) To use a simple hernia needle for single-port laparoscopic percutaneous inguinal hernia repair in children: a 5-year experience study. Front. Pediatr. 11:1298643. doi: 10.3389/fped.2023.1298643

#### COPYRIGHT

© 2024 Zhang, Zhang, Zhang, Hu, Chen, Fu and Li. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

# To use a simple hernia needle for single-port laparoscopic percutaneous inguinal hernia repair in children: a 5-year experience study

Li Zhang<sup>1\*</sup>, Rui Zhang<sup>2</sup>, Jianfa Zhang<sup>1</sup>, Hailong Hu<sup>1</sup>, Zihan Chen<sup>1</sup>, Yanxiang Fu<sup>1</sup> and Saihua Li<sup>1</sup>

<sup>1</sup>Department of General Surgery, Yellow River Sanmenxia Hospital, Sanmenxia, China, <sup>2</sup>Institute of Medical Technology, Peking University Health Science Center, Beijing, China

**Purpose:** The aim of this study is to investigate the technique and practical significance of using a simple hernia needle in single-port laparoscopic herniorrhaphy in pediatric patients.

**Methods:** The study conducted a retrospective analysis of all pediatric patients who underwent treatment for inguinal hernia using single-port laparoscopic herniorrhaphy with a simple hernia needle at Yellow River Sanmenxia Hospital from June 2018 to May 2023. The medical records of all the children were collected, and clinical characteristics, procedural information, and follow-up data were carefully reviewed.

**Results:** A total of 848 patients underwent inguinal hernia repair, with ages ranging from 7 months to 13 years ( $2.99 \pm 2.49$  years), including 756 males and 92 females. A total of 528 cases of unilateral hernia and 310 cases of bilateral hernia were reported, with intra-operative findings revealing contralateral occult hernia in 253 cases. Single-port laparoscopic herniorrhaphy was successfully completed in all patients, with no instances of conversion to open surgery. The mean operation time for unilateral hernia repair was ( $7.50 \pm 4.80$ ) min, while for bilateral hernia repair it was ( $11.55 \pm 7.27$ ) min. Five patients presented with subcutaneous emphysema, while two patients experienced a recurrence of inguinal hernia. No complications, such as scrotal hematoma, trocar umbilical hernia and testicular atrophy, were observed. The duration of the follow-up period ranged from 3 to 24 months.

**Conclusion:** The promotion and utilization of single-port laparoscopy combined with a simple hernia needle in clinical practice are justified. Our initial investigation indicates that this surgical approach is both safe and dependable for the management of pediatric inguinal hernia. The procedure presents numerous benefits, including the utilization of uncomplicated instruments, straightforward operation, a clear curative impact, minimal tissue damage, rapid recovery, and the absence of scarring.

#### KEYWORDS

single-port laparoscopic surgery, inguinal hernia, child, pediatric surgery, minimal invasive
## Introduction

In children, inguinal hernia is a prevalent and frequently occurring condition, with a higher incidence in boys compared to girls (1). Additionally, the incidence of inguinal hernia in premature infants is greater than in full-term infants (3%-5% vs. 15%) (2). The majority of these conditions are attributed to congenital factors. Some children may experience anorexia, nausea, vomiting, abdominal pain, and other symptoms as a result of an incarcerated hernia mass, which can have implications for the health and nutritional status of children. As the disease progresses, it can also lead to intestinal necrosis (3). Traditionally, pediatric and infant inguinal herniorrhaphy has been performed using an open technique through the inguinal skin crease. While there are multiple iterations of this approach, the fundamental concept involves isolating, ligating, and ultimately excising the hernia sac as near as feasible to the inner inguinal ring. This procedure remains the most frequently conducted elective surgery in the field of pediatric general surgery (4).

The increasing popularity of laparoscopic repair for pediatric inguinal hernia can be attributed to the emergence of minimal access techniques (5, 6). Recent meta-analyses have shown that laparoscopic hernia repair has fewer or comparable testicular complications and does not increase ipsilateral hernia recurrence compared with open hernia repair. Laparoscopic extraperitoneal herniorrhaphy has a lower incidence of metachronous contralateral inguinal hernia than open herniorrhaphy (7).

Nevertheless, laparoscopy presents a lengthier learning curve for novice practitioners. To mitigate the complexity of the procedure, a novel uncomplicated hernia needle is introduced for enhanced ease of use in single-port laparoscopic hernia repair. The utilization of a novel uncomplicated hernia puncture needle device simplifies and expedites laparoscopic hernia repair, and reduces the learning curve associated with this sophisticated technique. Additionally, an assessment of the safety and efficacy was conducted.

## Methods

#### Clinical data

This study was retrospective in nature, and therefore informed consent may be waived. This retrospective study comprised 848 pediatric patients who underwent hernia repair utilizing the single-port laparoscopic technique from June 2018 to May 2023. All instances were diagnosed through physical examination and preoperative ultrasound scanning. The study received approval from our institutional review board.

The inclusion criteria are as follows: 1. The patients included in the study had an age range from 6 months to 14 years. 2. The patients were monitored for a period exceeding 3 months following the surgical procedure.

Exclusion criteria: 1. Patients are either younger than 6 months or older than 14 years of age. 2. Patients who were lost to follow-up or observed for less than 3 months were excluded from the analysis. 3. Patients with incarcerated hernias or other conditions necessitating simultaneous surgical intervention.

The collected data encompassed medical history, demographic details, clinical characteristics, surgical treatment outcomes, postoperative complications, and recurrence. All the operations were performed by a senior surgeon.

#### Surgical instrument

The equipment used in the pediatric laparoscopy included a 5 mm laparoscope, an arc-shaped hernia needle with a polished sharp end and a drilled hole for silk thread passage (external diameter 1.3 mm, effective working length 15 cm) (Figure 1A), a 1 ml syringe for puncture positioning, and a spring clip needle with a spring-equipped top and a hook groove at the distal end of the needle core. Depress the end of the needle to elongate the crochet, engage the ligating thread, and subsequently release the handle to retract it, allowing for a secure grasp of the thread (Figure 1B,C), 1-0 surgical silk thread.

#### Surgical method

The patients received intravenous general anesthesia while positioned with their head down or with a soft pad placed under their buttocks. A 5 mm incision was performed at the inferior border of the umbilicus, and  $CO_2$  pneumoperitoneum was created through puncture with a pneumoperitoneum needle. The pressure was sustained at approximately 8 mmHg (1 mm Hg =





0.133 kPa), and a 5 mm Trocar was inserted. Following abdominal exploration, the researchers identified the position of the hernia ring and examined the shape and dimensions of the hernia ring, as well as the extent of relaxation of the hernia sac, and assess the presence of any concomitant contralateral occult hernia (Figure 2A). A 1 ml syringe needle was utilized to create a puncture mark on the local skin at the upper edge of the inner ring mouth on the anterior abdominal wall. Hernia needle (1-0 silk thread through the needle hole, with both ends left outside the abdominal cavity) was inserted through the puncture point to the retroperitoneum at the upper edge of the inner ring mouth, and subsequently passed behind the retroperitoneum along the medial edge of the inner ring mouth (Figure 2B). Following passage through the vas deferens (or ligamentum teres), the needle tip changed direction towards the lateral aspect, traversed the spermatic cord (or ligamentum teres) vessels towards the midpoint below the inner ring (which may be slightly lateral to access the lateral abdominal wall), and then pierced the peritoneum, extending into the abdominal cavity (Figure 2C). At this juncture, the assistant proceeded to pull one end of the wire, causing the other end to be drawn out through the needle hole of the wire feeder and left within the abdominal cavity. The surgeon carefully retracted the wire feeding needle along the puncture path gap without removing the wire (Figure 2D). Starting from the initial puncture site, insert the spring clip needle into the retroperitoneum of the inner ring, maneuvering behind the retroperitoneum from the lateral edge to the exit point of the abdominal cavity. Once inside the abdominal cavity, push the end of the needle handle to extend the hook, and use the hook to catch the end of the line (Figure 2E). It is challenging to remove the needle and thread after ensuring precision, carefully exiting the needle and the line, and having the assistant expel the air from the scrotum or labia majora before tightening the suture knot. The inner ring was inspected to ensure precise closure (Figure 2F), and the procedure on that side was completed. Simultaneous treatment was administered for hernias on the contralateral side. The incision was sealed using medical adhesive rather than being sutured. Complete surgical video is available in the supplementary document (Supplementary Video S1).

#### Result

All patients successfully underwent single-port laparoscopic hernia repair without requiring conversion to open surgery. There were 756 male participants and 92 female participants, with a mean age of  $(2.99 \pm 2.49)$  years. The average operating time for unilateral hernia patients was  $(7.50 \pm 4.80)$  minutes, while for bilateral hernia patients it was  $(11.55 \pm 7.27)$  minutes. Out of 310 patients with bilateral hernia, 253 cases were identified during the operation. During the operation, there was no apparent bleeding, and no damage was observed to the inferior epigastric vessels, spermatic cord vessels, or vas deferens. Subcutaneous emphysema was observed in 5 postoperative patients, while no instances of scrotal hematoma, iatrogenic cryptorchidism, foreign body reaction to suture knots, puncture hernia, or other complications were reported. The children were able to consume food and rise from bed 6 h after regaining consciousness from anesthesia without displaying overt signs of pain. The incision exhibited satisfactory healing with no discernible scarring. The patients were monitored for a period ranging from 3 to 24 months (mean  $14.65 \pm 5.34$  mounth), during which 2 patients were identified as having recurrent inguinal hernia (both had a history of chronic constipation) (Table 1).

#### Discussion

Inguinal hernia is one of the most common congenital disorders in children, occurring in 1%-2% of full-term infants and up to 30% of preterm infants (8, 9). The main pathogenesis



#### FIGURE 2

(A) identified the position of the hernia; (B) hernia needle passed behind the retroperitoneum along the medial edge of the inner ring mouth; (C) hernia needle through the vas deferens (or ligamentum teres); (D) retracted the wire feeding needle; (E) use the hook to catch the end of the line; (F) tightening the suture knot.

#### TABLE 1 Patients' characteristics.

Parameters	Observations
No. of patients	838
Mean age	$2.99 \pm 2.49$
Sex	
Male	756
Female	92
Side of hernia	
Unilateral	528 (63.01%)
Bilateral	310 (36.99%)
Contralateral occult	253 (30.19%)
Operating time (min)	
Unilateral	$7.50 \pm 4.80$
Bilateral	11.55 ± 7.27
Hospital stay	1-3 days (1.73 ± 0.32 days)
Follow-up	3-24 months (14.65 ± 5.34 months)
Complications	
Subcutaneous emphysema	5 (0.60%)
Recurrence	2 (0.24%)

is the failure of the vaginal process to close, which allows the contents of the abdominal cavity to enter the groin or scrotum through the open vaginal process, resulting in clinical symptoms (10). Currently, open hernia repair is considered the preferred method for treating pediatric inguinal hernias. However, the procedure carries certain risks because it requires opening the inguinal canal and removing the hernia sac. This process increases the likelihood of damaging the vas deferens, spermatic cord vessels, and other associated structures. As a result, complications such as hematoma, iatrogenic cryptorchidism, testicular atrophy, and even infertility may arise (11, 12). Perlstein reported that 2.3%-15% of patients experience testicular dysplasia, atrophy, or even iatrogenic cryptorchidism (13). Furthermore, the vas deferens is vulnerable to mechanical damage. According to Harrison et al., the incidence of vas deferens obstruction on one side was found to be 26.7% in children who had previously undergone open inguinal hernia surgery (14). In contrast, laparoscopic surgery has gained popularity because of its aesthetic benefits, reduced postoperative pain, and ability to minimize damage to the vas deferens and spermatic cord vessels (15). Furthermore, laparoscopy can effectively identify contralateral occult hernias without requiring additional incisions, thereby reducing the need for further surgery to some extent (16, 17). In this study, 253 cases of contralateral occult hernia were found during laparoscopic surgery, representing 47.92% of unilateral hernias diagnosed before surgery. Some patients may develop an inguinal hernia and subsequently need further treatment.

Laparoscopic techniques have been described in numerous versions, ranging from a three-trocar intraperitoneal approach to one or two trocar percutaneous approaches. The technique is constantly advancing with a trend towards increasing the application of extraperitoneal repair and decreasing the use of assistant instruments and trocars.

Single-port laparoscopic high ligation of a hernia sac requires special operating instruments, such as a multi-channel puncture device, Kirschner wire, epidural puncture needle, and abdominal wall suture straight needle. It also requires a scalpel to make a 2-3 mm skin incision at the projection of the inner ring, which may result in skin scars after the operation. During the procedure, the curved wire delivery needle and spring clip needle were used to puncture the abdominal wall directly into the abdominal cavity. The silk thread was then brought out around the inner ring through the needle hole using the wire delivery and spring clip technique, and the incision was sealed with medical glue. The scar at the puncture site could completely disappear within 2 weeks after the surgery. The pointed end of the wire delivery needle is polished, which not only widens the gap of the extraperitoneal channel but also prevents penetration of the peritoneum and damage to the surrounding tissues. It can also smoothly guide the ligature wire into the needle hole, ensuring a safe and quick process. Despite the numerous advantages of this approach, it should be used cautiously in patients with certain types of hernias, as the presence of the organ increases the risk of injury and the complexity of surgery. We have also updated some references (18, 19).

It is important to consider the following points during the operation: (1) when establishing pneumoperitoneum, the umbilical Trocar should be used to blindly enter the abdominal cavity, and the operator and assistant should lift the abdominal wall to prevent accidental injury to the abdominal organs. (2) During the positioning process, it is recommended to advance the laparoscopic lens in order to identify the deep inferior epigastric vessels. Additionally, the use of a 1 ml syringe for the surface positioning of the inner ring is advised to facilitate the success of the puncture needle at one time and to prevent accidental injury. (3) The puncture site may be chosen approximately 1-2 cm above the inner ring, and the needle and thread can be passed through the lower edge of the arcuate, thus avoiding significant blood vessels and nerves in the inguinal region, which enhances safety. (4) It is preferable that the needle does not intersect the vas deferens and spermatic cord vessels in a vertical manner. The instrument can be swung in front of it to widen the gap, and then the peritoneum over the above structures can be gently manipulated. In this procedure, we employ the technique of refining the pointed section of the wire feeding needle to prevent secondary harm, ensuring convenient and safe operation. (5) In the arc-shaped wire feeding needle process, a 1-0 silk thread is passed through the needle hole, with both ends extending outside the abdominal cavity. When the needle pierces the peritoneum and enters the abdominal cavity, one end of the wire is gently pulled by the assistant, while the other end is drawn out through the needle hole and left in the abdominal cavity. (6) Upon grasping the end of the spring clip needle, it is advisable not to withdraw the needle immediately. Hold the outer abdominal segment with one hand and use the other hand to guide the needle into the abdominal cavity. When one perceives the ability to endure a specific level of tension, it is possible to retract the needle in order to prevent the silk thread from slipping. (7) When performing hernia ring ligation in male children, it is important to ensure proper traction of the testis and to apply pressure to the inguinal area to expel air from the scrotum, thereby preventing postoperative scrotal and

subcutaneous emphysema. Additionally, care should be taken to avoid ligating the spermatic cord vessels and vas deferens in order to prevent iatrogenic cryptorchidism. (8) After tying and cutting the suture, it is recommended to pull the skin at the puncture point and ensure that the suture knot is left in the preperitoneal space to minimize the sensation of a subcutaneous foreign body. (9) The pneumoperitoneum should be released, with the Trocar being exited first, followed by the lens, in order to prevent the omentum tissue from becoming trapped in the puncture hole with the gas.

## Conclusion

This study presents our findings on the use of single-port laparoscopic hernia repair with a basic hernia needle in 838 pediatric patients with inguinal hernia at our hospital over a 5year timeframe. The procedure offers the benefits of minimal trauma, rapid recovery, short operation duration, aesthetically pleasing incisions, and a clear therapeutic impact. It is also capable of diagnosing and treating contralateral occult hernia simultaneously, thereby preventing the need for a second operation and the associated pain. Consequently, this approach has the potential for extensive application in the management of pediatric inguinal hernia.

#### 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 humans were approved by yellow river sanmenxia hospital. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation in this study was provided by the participants' legal guardians/next of kin.

#### References

1. Chang SJ, Chen JY, Hsu CK, Chuang FC, Yang SS. The incidence of inguinal hernia and associated risk factors of incarceration in pediatric inguinal hernia: a nation-wide longitudinal population-based study. *Hernia.* (2016) 20(4):559–63. doi: 10.1007/s10029-015-1450-x

2. Frawley G. Special considerations in the premature and ex-premature infant. *Anaesth Intensive Care Med.* (2020) 21(2):23–9. doi: 10.1016/j.mpaic. 2022.10.013

3. Jing-you D, Ming-lu L, San-shi L, Teng-fei Y, Xue-lei W, Zhi-feng L, et al. Effects of single-port laparoscopic high ligation of hernia sac on body microenvironment, satisfaction of incision aesthetics and economic benefits in treatment of children with indirect inguinal hernia. *Clin Misdiagn Misther*. (2022) 35(3):93–7. doi: 10. 3969/j.issn.1002-3429.2022.03.021

#### Author contributions

LZ: Conceptualization, Data curation, Investigation, Methodology, Project administration, Resources, Supervision, Writing – original draft, Writing – review & editing. RZ: Conceptualization, Data curation, Methodology, Resources, Supervision, Writing – original draft, Writing – review & editing. JZ: Data curation, Investigation, Methodology, Resources, Writing – review & editing. HH: Data curation, Investigation, Resources, Supervision, Writing – review & editing. ZC: Data curation, Investigation, Methodology, Resources, Writing – review & editing. YF: Data curation, Investigation, Methodology, Writing – review & editing. SL: Data curation, Investigation, Methodology, Writing – review & editing.

## Funding

The author(s) declare that no financial support was received for the research, authorship, and/or publication of this article.

## 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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

#### Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fped.2023. 1298643/full#supplementary-material

4. Lao OB, Fitzgibbons RJ Jr, Cusick RA. Pediatric inguinal hernias, hydroceles, and undescended testicles. *Surg Clin North Am.* (2012) 92(3):487–504, vii. doi: 10.1016/j. suc.2012.03.017

5. Shaughnessy MP, Maassel NL, Yung N, Solomon DG, Cowles RA. Laparoscopy is increasingly used for pediatric inguinal hernia repair. *J Pediatr Surg.* (2021) 56 (11):2016–21. doi: 10.1016/j.jpedsurg.2021.01.032

6. Schmedding A, Alsweed A, Muensterer O, Leonhardt J. The status of laparoscopic inguinal hernia surgery in children: a nationwide assessment. *Children (Basel, Switzerland).* (2022) 9:3. doi: 10.3390/children9030348

7. Cheng PL, Duh YC, Chen JJ, Huang FH. Laparoscopic extraperitoneal hernia repair versus open repair in boys with inguinal hernia: a meta-analysis. *J Pediatr Surg.* (2023) 58(7):1322–31. doi: 10.1016/j.jpedsurg.2023.01.054

8. Skoog SJ, Conlin MJ. Pediatric hernias and hydroceles. The urologist's perspective. Urol Clin North Am. (1995) 22(1):119-30. doi: 10.1016/S0094-0143(21)01022-3

9. Wolak PK, Patkowski D. Laparoscopic inguinal hernia repair in children using the percutaneous internal ring suturing technique—own experience. *Wideochir Inne Tech Maloinwazyjne*. (2014) 9(1):53–8. doi: 10.5114/wiitm.2014.40389

10. Wang C, Wang Y, Zhong M, Li R, Shen Z. Logistic analysis of the recurrence of laparoscopic percutaneous extraperitoneal repair of pediatric inguinal hernia: a report of 486 cases. *Asian J Surg.* (2023). doi: 10.1016/j.asjsur.2023.05.013

11. Cam C, Celik C, Sancak A, Iskender C, Karateke A. Inguinal herniorrhaphy in childhood may result in tubal damage and future infertility. *Arch Gynecol Obstet.* (2009) 279(2):175–6. doi: 10.1007/s00404-008-0654-z

12. Prasad R, Lovvorn HN 3rd, Wadie GM, Lobe TE. Early experience with needleoscopic inguinal herniorrhaphy in children. *J Pediatr Surg.* (2003) 38 (7):1055–8. doi: 10.1016/s0022-3468(03)00191-x

13. Perlstein J, Du Bois JJ. The role of laparoscopy in the management of suspected recurrent pediatric hernias. *J Pediatr Surg.* (2000) 35(8):1205-8. doi: 10.1053/jpsu. 2000.8727

14. Harrison MR, Lee H, Albanese CT, Farmer DL. Subcutaneous endoscopically assisted ligation (SEAL) of the internal ring for repair of inguinal hernias in

children: a novel technique. J Pediatr Surg. (2005) 40(7):1177–80. doi: 10.1016/j. jpedsurg.2005.03.075

15. Takehara H, Yakabe S, Kameoka K. Laparoscopic percutaneous extraperitoneal closure for inguinal hernia in children: clinical outcome of 972 repairs done in 3 pediatric surgical institutions. *J Pediatr Surg.* (2006) 41(12):1999–2003. doi: 10.1016/j.jpedsurg.2006.08.032

16. Shalaby R, Ismail M, Samaha A, Yehya A, Ibrahem R, Gouda S, et al. Laparoscopic inguinal hernia repair; experience with 874 children. *J Pediatr Surg.* (2014) 49(3):460–4. doi: 10.1016/j.jpedsurg.2013.10.019

17. Bertozzi M, Melissa B, Magrini E, Bini V, Appignani A. Laparoscopic herniorrhaphy in the pediatric age group: what about the learning curve? *J Endourol.* (2013) 27(7):840-4. doi: 10.1089/end.2012.0690

18. Taher H, Elboraie A, Fares A, Tawfiq S, Elbarbary M, Abdullateef KS. Laparoscopic inguinal hernia repair in bladder exstrophy, a new modified solution to an old problem: a cohort study. *Int J Surg Case Rep.* (2022) 95:107252. doi: 10. 1016/j.ijscr.2022.107252

19. Taher H, Azzam A, Kamal A, Husseiny R, Farrag M, Nayef A, et al. Hernia uteri inguinale in an 18 months old female infant: a case report. *Int J Surg Case Rep.* (2021) 78:1–3. doi: 10.1016/j.ijscr.2020.11.125

Check for updates

#### **OPEN ACCESS**

EDITED BY Edoardo Bindi, Salesi Hospital Foundation, Italy

REVIEWED BY Gergo Jozsa, University of Pécs, Hungary Filippo Maria Senes, Casa di Cura Villa Serena GVM, Italy

\*correspondence Yang Li ⊠ ice0917@sina.com

RECEIVED 30 July 2023 ACCEPTED 27 December 2023 PUBLISHED 10 January 2024

#### CITATION

Yang W, Zhang X, Sun D, Jin S, Chen J and Li Y (2024) Outcomes of olecranon fractures in adolescents: comparison of tension band wiring and Herbert screw fixations. Front. Pediatr. 11:1269628. doi: 10.3389/fped.2023.1269628

#### COPYRIGHT

© 2024 Yang, Zhang, Sun, Jin, Chen and Li. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

## Outcomes of olecranon fractures in adolescents: comparison of tension band wiring and Herbert screw fixations

Weiwei Yang, Xintao Zhang, Dong Sun, Shaobin Jin, Junfei Chen and Yang Li\*

Department of Pediatric Surgery, Qilu Hospital of Shandong University, Shandong, China

**Purpose:** Olecranon fracture is considered intra-articular when there is obvious displacement or an irregular articular surface. Such fractures should be reduced and fixed via surgery. No clear indications regarding the surgical technique to be adopted exist. Therefore, this study aimed to compare the outcomes of tension band wiring (TBW) and Herbert screw fixations for olecranon fractures.

**Methods:** We retrospectively analyzed the clinical data of 29 children with olecranon fractures. They were divided into the tension band wiring and Herbert screw groups. We assessed early epiphyseal closure and maximum length of the ulna using radiography. Patients were clinically evaluated using the average QuickDASH score.

**Results:** Both groups had good radiological outcomes. Herbert screws demonstrated advantages in terms of bleeding, operative time, intraoperative blood loss, surgery duration, and particularly the QuickDASH score (1.57 vs. 4.18, p < 0.05). Complications, including needle loosening and bursitis, occurred in five cases in the TBW group. Six cases had premature physis plate closure, and no difference was observed in limb length at 6 months after surgery. **Conclusion:** Compared with TBW, Hebert screws demonstrated better clinical outcomes and lesser postoperative complications in the treatment of ulnar olecranon fractures in children. However, long-term follow-up is required to assess the effects of screws on the ulnar physis plate and ulna length.

#### KEYWORDS

children, olecranon fracture, Herbert screw fixation, epiphyseal growth plate, tension band wiring

#### 1 Introduction

Olecranon fracture is considered intra-articular when the fracture has an apparent displacement or unsmooth articular surface and should be reduced and fixed via surgery to restore the normal para-position and to reduce elbow joint dysfunction, such as elbow stiffness, malunion, and traumatic arthritis. Olecranon fractures account for approximately 4%–7% of elbow fractures in children (1). Approximately 20% of cases are accompanied by other injuries to the ipsilateral elbow joint (2).

However, surgical indications remain controversial. It is generally believed that when the fracture block separation distance is <2 mm or 2-4 mm, with an elbow flexion of 90°, the

Abbreviations

TBW, tension-band wiring; TBS, tension-band suturing; ROM, range of motion; OI, osteogenesis imperfecta.

fracture is stable, the joint surface is smooth, and the active antigravity extension of the elbow and the fracture is undisplaced, conservative treatment can be chosen. However, when the articular surface is unsmooth or the displacement is >4 mm, it could be an indication for open reduction and internal fixation (3, 4).

Surgical internal fixation techniques include tension-band wiring (TBW), tension-band suturing (TBS), surgery using screws, and other approaches, and Some scholars tried to use bioabsorbable compression screws or polyethylene tension band for fixation of displaced olecranon fractures (5). However, open reduction and TBW are the "gold standard" for treating olecranon fractures (6). The tension band suture can convert the tension through the fracture site's posterior cortex into pressure on the articular surface to increase the fracture fixation's stability and to prevent displacement. Its main disadvantages include steel needle displacement and steel wire stimulation of the skin. Additionally, a second incision is needed for removing the fixation (7). Screw fixation for olecranon fractures can provide sufficient pressure with minimal trauma. Currently, it is mainly used in children with osteogenic insufficiency (9). Due to relative osteoporosis in children with imperfect osteogenesis, the adhesion between the screw and bone is insufficient, leading to graft loosening. However, in relevant studies, no significant difference was observed between screw and tension-band fixation in adults. Corradin et al. reported good results with TBW and screw fixation for olecranon fractures in healthy children (9). Another concern regarding screw fixation is its effect on the epiphyseal plates. Currently, evidence to compare the efficacy of TBW and compression screw fixation for isolated olecranon fractures in healthy children is insufficient. The effect of screw fixation on the ulnar olecranon epiphyseal plate in children has rarely been investigated.

This study aimed to compare the clinical outcomes of open reduction with TBW fixation and closed reduction with Herbert screw internal fixation for treating olecranon fractures in children and to explore the possible effects of internal fixation device on olecranon epiphysis and ulna growth in children with olecranon fractures.

#### 2 Materials and methods

Children (n = 29) with ulnar olecranon fractures who underwent surgery between January 2017 and June 2022 in our treatment center, Qilu Hospital of Shandong University, were included in the study. There were 18 boys and 11 girls, with 11 cases of fractures on the left side and 18 on the right. The causes of injury included 24 cases of falls and 5 of traffic accidents. The age range was 9–14 years. According to the different internal grafts, the children were divided into the TBW (group A: 12 patients) and Herbert screw (group B: 17 patients) groups. The average age was 11.46 (11.6  $\pm$  1.20 vs. 11.3  $\pm$  1.33) years. Data on the sex, age, and fracture side of both groups were statistically analyzed, and the difference was not significant (p > 0.05, Table 1). All the cases were of recent fractures, and the period from injury to surgery was 1–5 (average period 2.42  $\pm$  1.24 vs. 2.24  $\pm$  0.76) days. The Medical Ethics Committee of Qilu Hospital of Shandong University approved this study (approval number KYLL-2020008-165).

#### 2.1 Inclusion and exclusion criteria

The inclusion criteria were as follows: isolated closed ulnar olecranon fractures or fractures at other sites of the forearm that did not require surgery; fracture displacement of 2-4 mm with an unsmooth joint surface or a displacement of >4 mm; patients who underwent open reduction TBW or closed reduction screw fixation; age 9–14 years; and follow-up of not less than 6 months.

The exclusion criteria were ulnar olecranon fractures combined with fractures of other sites of the forearm, which require surgery; open fracture of the ulnar olecranon, pathological fracture, or comminuted fracture; and age <9 years or >14 years. The olecranon epiphyseal plate on the radiograph was closed.

#### 2.2 Surgical methods

Experienced pediatric orthopedic surgeons performed both procedures, The choice of internal fixation depends was based on the physician's discretion and guardian's preference. After inducing anesthesia, the patient was placed supine, and the affected limb was placed on a C-arm and disinfected.

#### 2.2.1 TBW group

A longitudinal posterior incision of approximately 6 cm was made in the proximal ulna. The fracture was exposed, the towel clamp was temporarily reduced, and the fractured piece was immobilized. Based on the child's age, two smooth Kirschner needles with a diameter of 1.6 mm or 2.0 mm were selected. Two parallel Kirschner wires were longitudinally passed from the tip of the olecranon into the distal part of the ulna and close to the articular surface in front of the ulnar olecranon, and the distance between the Kirschner needles was approximately 0.8 cm. The distal point of the Kirschner needle was inserted into the cortex during the distal coronal process. We drilled holes on both sides of the ulnar crest, approximately 4 cm from the distal end of the fracture line and perpendicular to the longitudinal axis of the ulna. We selected a steel wire with an appropriate diameter to pass through the bone

TABLE 1 Characteristics of the patients.

Number	Age (year)	Side (L:R)	Sex (M:F)	Operation waiting time (day)	Surgery duration (min)	Amount of bleeding	Follow-up	QuickDASH score
12	11.6	5:7	7:5	2.4	82.5	39.1	12.2	4.78
17	11.3	6:11	11:6	2.2	32.1	4.9	13.5	1.57

tunnel, cross-fixed in an "8" figure near the fracture line. We tightened and knotted the wires around the Kirschner needle tail, bent and cut the needle tail, and sutured the incision (Figure 1).

#### 2.2.2 Herbert screw group

The elbows were extended to reduce the traction of the triceps muscles on the fracture. Sometimes temporarily inserting a Kirschner needle proximal to the fracture and using the joystick technique to assist the reduction was required. After fracture reduction, two 0.8mm Kirschner wires were inserted as guide needles from the proximal to distal points to fix the fractures temporarily. Two Herbert screws with a 3.0-mm diameter were implanted in the direction of the guide needle, which were then removed (Figure 2).

#### 2.3 Postoperative treatment and evaluation

Patients in both groups were immobilized using a plaster cast postoperatively. AP and lateral radiographs of the elbow joint were reviewed on the first day, 2 weeks, and 4–6 weeks after surgery. Elbow joint function exercises were gradually performed after removing the cast after 4–6 weeks based on the fracture healing. At the 6-month follow-up, we used QuickDASH, bilateral anteroposterior (AP) and lateral radiographs to assess elbow joint function, elbow flexion and extension range, maximum length of ulna, and epiphysis closure. Patients were asked if they wanted their internal fixation removed. All patients had their internal fixation removed at 6–9 months after surgery.

#### 2.3.1 TBW group

The patients in this group underwent a procedure through the original incision to expose the TBW, which was then removed.

#### 2.3.2 Herbert screw group

The patient was placed in the supine position, and the affected limb was placed on a C-arm x-ray device and sterilized. A 0.8-mm Kirschner wire was inserted into the hollow screw and confirmed on AP and lateral radiographs. A 0.5-cm incision was made in the skin around the Kirschner wire, and the Herbert screw was unscrewed in the direction of the Kirschner wire. Compared to TBW, the use of a hollow screw resulted in a smaller incision and less surgical trauma.



#### FIGURE 1

(A) preoperative images; anteroposterior (AP) and lateral radiographs in a 12-year-old boy with an olecranon fracture. (B) Postoperative images from the 1-day follow-up and AP and lateral radiographs in the same patient treated with closed reduction and Herbert screw fixation. (C) Follow-up images obtained at 6 months after surgery in the same patient. (D) Functional outcomes in the same patient at 6 months after surgery. Before removing the cannulated screw, we recorded the patient's elbow range of motion in flexion, extension, pronation, and supination, forearm length, and QuickDASH score.



(A) preoperative images; AP and lateral radiographs in a 13-year-old boy with an olecranon fracture (B) postoperative images from the 1-day follow-up and AP and lateral radiographs in the same patient treated with closed reduction and Herbert screw fixation. (C) Follow-up images obtained at 6 months after surgery in the same patient. (D) Functional outcomes in the same patient at 6 months after surgery. Before removing the cannulated screw, we recorded the patient's elbow range of motion in flexion, extension, pronation, and supination, forearm length, and QuickDASH score.

## 2.4 Statistical methods

IBM SPSS Statistics for Windows, version 19.0 (IBM Corp., Armonk, NY, USA) was used for the analysis and comparison. The data were represented using means  $\pm$  standard deviations. The data of both groups were compared using the t-test for two independent samples or Mann–Whitney *U* test. The threshold for statistical significance was p < 0.05.

## **3** Results

All 29 children were followed-up for 9–22 months, averaging 12.9 months. There were 12 patients in the TBW group (average follow-up period of 12.2 months) and 17 in the Herbert screw group (average follow-up period of 13.5 months). The average operation time of the Herbert screw group was 32.1 min less than that of the TBW group 82.5 min (P < 0.01) The volume of blood lost in the Herbert screw group was 4.9 ml, which was lower than that in the TBW group 39.0 ml (P < 0.01) (Table 1).

The mean QuickDASH scores in the TBW and Herbert screw groups were 4.78 and 1.57, respectively. In the TBW group, the

steel needles loosened and were therefore withdrawn in three patients; further, two developed bursitis at the proximal ulna, which caused pain. Nine of the 12 patients in the TBW group strongly desired to have their internal fixation removed, especially those with a low body mass index. Many patients had difficulty tolerating foreign bodies and tissue scarring behind the elbow (Tables 2, 3).

No patient in the Herbert screw group had foreign body irritation, implant migration, or osteoarthritis. Two of the 17 patients in this group strongly desired to have their internal fixation removed. The internal fixation was removed in most patients at 6–8 months after surgery because cannulated compression screws might affect the growth of epiphyseal or secondary ossification centers. Before removing the internal fixation, elbow function was assessed in both groups, and the elbow flexion of children in the Herbert screw group was better than that of those in the TBW group. No significant difference was observed in elbow flexion and forearm pronation or supination between both groups. In the sixth month, we assessed the limb length of affected sides in two group patients, and no difference in limb length existed in both groups (Table 4).

Patient	Age at trauma	Sex	Side	Operation waiting time (day)	Surgery duration (min)	Amount of bleeding (ml)	Follow-up	Quick DASH	Aspiration of removing internal fixation
1	11.3	М	L	1	80	40	11	3.33	Y
2	10.2	F	R	2	90	30	13	5	N
3	13.6	М	R	4	70	30	9	5	Y
4	10.9	М	R	3	110	40	9	4.14	Y
5	13.7	F	R	5	80	20	15	6.67	Y
6	12.4	М	L	2	90	50	13	2.5	N
7	10.0	М	L	3	80	40	12	8.33	Y
8	10.8	F	R	1	100	40	11	4.14	Y
9	10.8	М	L	1	70	50	14	3.33	Y
10	11.7	F	R	3	80	40	10	4.14	Y
11	12.2	М	L	2	70	50	14	5	Y
12	11.7	F	R	2	70	40	15	5.83	N

#### TABLE 2 Group of patients treated by open reduction and TBW fixation.

TABLE 3 Group of patients treated by closed reduction and Herbert screw fixation.

Patient	Age at trauma	Sex	Side	Operation waiting time	Surgery duration (min)	Amount of bleeding (ml)	Follow-up	Quick DASH	Aspiration of removing internal fixation
1	10.0	М	R	2	40	5	13	1.67	N
2	13.2	М	L	2	25	5	15	0	N
3	12.1	F	R	2	30	3	13	3.33	Y
4	9.8	М	R	2	30	5	12	2.5	N
5	11.2	F	L	3	25	8	14	1.67	N
6	10.6	М	R	4	40	5	22	0	N
7	12.5	М	R	3	30	5	15	0	N
8	12.3	F	R	3	30	2	14	0.83	Y
9	9.8	М	L	2	40	5	15	3.33	N
10	9.6	М	L	2	30	5	10	1.67	N
11	13.0	F	R	1	30	3	9	1.67	N
12	10.8	F	R	3	25	4	13	0.83	N
13	11.7	М	R	2	30	5	14	2.5	N
14	10.5	М	L	2	25	5	14	3.33	N
15	10.6	М	L	2	50	5	15	1.67	N
16	13.9	М	R	1	30	8	9	1.67	N
17	11.3	F	R	2	35	5	12	0	N

TABLE 4 Comparison of ROM and ulna length outcomes between the TBW and Herbert screw fixation.

	TBW	Herbert screw fixation	Р
Elbow flexion (°)	135 (135, 140)	140 (140, 145)	0.024
Elbow extension (°)	0 (0, 5)	0 (0, 5)	0.586
Forearm pronation (°)	85 (80, 85)	85 (85, 85)	0.245
Forearm supination (°)	85 (81.25, 90)	85 (85, 90)	0.499
Maximum length of the ulna (cm)	$22.72 \pm 1.22$	$22.46 \pm 1.37$	0.614

#### 4 Discussion

Ulnar olecranon fractures account for 4%–7% of all elbow fractures in children. Because of the effective fixation methods and excellent results (3), TBW has been regarded as the gold standard for olecranon fractures. However, hardware irritation, persistent joint pain, scar hyperplasia and other common complications trouble the patients. Although compression screw fixation for ulna olecranon fractures has been well described in adults (10), reports in the pediatric population are very few. This study investigated the effects of different fixation modalities on ulnar olecranon fractures.

Ulnar olecranon fracture treatment using TBW technology is a relatively mature surgical method, TBW is considered the gold standard for the surgical treatment of ulnar olecranon fractures in children (11). However, it requires a relatively long incision and adequate exposure of the proximal ulnar olecranon, often causing postoperative incision pain in children. Longer surgical incisions can also cause cosmetic challenges (11). In addition, owing to the low subcutaneous fat in the olecranon area and high incision tension, the needle tip can easily penetrate the skin, infecting the incision (12). Furthermore, this technology can loosen or displace the Kirschner needle, break the steel wire, and cause needle tail irritation of the skin and soft tissues, causing pain (13). In our study, grafts loosened in three cases, causing the tips of the Kirschner needle to touch the skin surface without fracture displacement or local infection during subsequent follow-up. In addition, two children developed bursitis at the proximal ulna, which caused pain and decreased the range of motion (ROM) of the elbow joint. Studies have revealed that the hardware removal

rate is significantly higher in children (63%) than in adults (6%–25%), possibly due to elbow dysfunction caused by Kirschner needle stimulation or the parents' willingness. In the TBW group, removal was performed under general anesthesia using an open incision almost identical to the original incision.

Good reduction and stable fixation are critical for recovery from olecranon fractures. This differs from that observed among the children in the TBW group, as no child in the Herbert screw group experienced graft loosening or local soft tissue irritation. When the Herbert screw was used to fix the ulna olecranon fracture, the ends of the screws were embedded in the ulna's periosteum, and foreign body rejection rarely occurred. In addition, soft tissue stimulation by the screw during elbow movement was negligible. Previous studies have suggested that screw fixation might result in graft loosening in patients with ulnar olecranon fractures with OI, possibly due to insufficient internal fixation and bone adhesion in children with osteoporosis (8). In this group, no patient experienced internal fixation loosening. However, the hollow screw did not reach the contralateral cortex in some patients. We believe that the hollow screw can provide sufficient pressure to the olecranon. During fixation, the screws are pressurized by the head and tail thread difference and number of screw-in threads to reduce the fracture gap and to achieve a better reduction. Hollow compression screws to fix ulnar olecranon fractures can provide sufficient pressure early to reset the fracture. Within 12 h of fixation, 39%-55% of compression disappears; however, good initial compression remains important (14). In addition, the fixation angle of the patients was maintained at approximately 45° of elbow flexion, which helped alleviate the triceps tension, indicating that the Herbert screw could provide sufficient holding force to resist the triceps pull.

All cases in the compression screw group achieved closed reduction, and Kirschner and guide needles were used to fix the fracture fragment temporarily. Compression screws were inserted along the guide needles, and the fracture gap gradually closed. In this procedure, a mini incision of approximately 0.5 cm is required. The compression screw group had apparent advantages in operation time, volume of blood loss, and postoperative pain compared with the TBW group. This is mainly due to the minimally invasive nature of the operation and sufficient stability of the screws. Additionally, when removing the compression screw, inserting the Kirschner needle along the original surgical incision and removing the screw along the guide needle to achieve a minimally invasive operation are necessary.

Hollow screw fixation is often controversial in children with ulnar olecranon fractures because of potential damage to the growth plate caused by a large screw. Compression screw fixation for ulnar olecranon fractures is mostly used in adults and children with osteogenesis imperfecta (15, 16), and the main concern is an injury to the ulnar olecranon epiphyseal plate in children, leading to forearm deformity and growth arrest (17). Ulnar olecranon epiphyseal ossification centers appear at 9–11 years of age and begin to heal with the ulnar shaft around the age of 17 years, whereas 15% of the ulna growth depends on the proximal growth plate, and the growth rate drops to 5% by the age of 8 years (18). In recent years, some researchers have applied screws to ulnar olecranon fractures in healthy children (9, 19) and achieved a clinical efficacy similar to that of tension band wires. Bilateral AP and lateral radiographs were examined after 6 months to study the effect of screws on premature epiphyseal closure and extremity length. Despite the premature closure of epiphyseal plate in six cases, no angle and ulnar length deformity was observed and no significant difference was observed in the elbow ROM score. Considering the smaller growth potential of the remaining olecranon in older children and stimulation of the epiphysis by the screw's small diameter and minimally invasive approach to preserve the olecranon blood supply, we suggest that the use of hollow screws results in negligible ulnar growth in older children. However, long-term follow-up is required to confirm whether the ulna is affected after the epiphyseal plate is completely closed.

We used QuickDASH and elbow ROM as evaluation index at 6 months after surgery. This scoring system can balance children's subjective feelings with the doctor's objective evaluation (20). In the TBW group, more than one child had a low score owing to a restricted ROM and pain. However, in the Herbert Screw group, children had higher acceptance, lower QuickDASH scores, and lesser endoplant-induced pain and activity restriction than those in the TBW group. We investigated the subjective feelings of the children and their parents when they had a strong desire to remove the internal fixation. Compared with those in the hollow screw group, patients in the TBW group strongly desired to have their internal fixation removed. Subjective factors are involved in this process; however, the compression screw group can be better accepted in evaluating the result.

This study had some limitations. First, this was a single-center study, and the sample size was small. Second, this study was retrospective, and the choice of surgical modality depended on the preference of the surgeon rather than randomization. Third, the follow-up duration was short. Finally, no data were provided on the ulna evaluation when the ulnar epiphysis was closed.

In conclusion, according to our study, TBW and screws achieved similar clinical results in treating ulnar olecranon fractures in children. Screw fixation is not contraindicated; rather, it offers unique advantages in terms of bleeding, operative time, and postoperative complications. Long-term follow-up is needed to observe ulnar growth after screw fixation.

#### Data availability statement

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

#### Ethics statement

The studies involving humans were approved by ethics committee of Qilu Hospital. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation in this study was provided by the participants' legal guardians/next of kin. Written informed consent was obtained from the individual (s), and minor(s)' legal guardian/next of kin, for the publication of any potentially identifiable images or data included in this article.

#### Author contributions

WY: Data curation, Writing – original draft. XZ: Writing – original draft, Conceptualization, Investigation. DS: Data curation, Formal Analysis, Methodology, Software, Writing – review & editing. SJ: Data curation, Formal Analysis, Writing – review & editing. JC: Data curation, Formal Analysis, Conceptualization, Writing – review & editing. YL: Supervision, Writing – review & editing.

#### Funding

The author(s) declare financial support was received for the research, authorship, and/or publication of this article.

This research was supported by the Natural Science Foundation of Shandong Province (grant No. ZR2020QH056).

#### References

1. De Maio F, Gorgolini G, Caterini A, Luciano C, Covino D, Farsetti P. Treatment of olecranon fractures in childhood: a systematic review. *Front Pediatr.* (2022) 10:1046243. doi: 10.3389/fped.2022.1046243

2. Caterini R, Farsetti P, D'Arrigo C, Ippolito E. Fractures of the olecranon in children. Long-term follow-up of 39 cases. J Pediatr Orthop B. (2002) 11:320-8. doi: 10.1097/00009957-200210000-00010

3. Chalidis BE, Sachinis NC, Samoladas EP, Dimitriou CG, Pournaras JD. Is tension band wiring technique the "gold standard" for the treatment of olecranon fractures? A long term functional outcome study. *J Orthop Surg Res.* (2008) 3:9. doi: 10.1186/1749-799X-3-9

4. Evans MC, Graham HK. Olecranon fractures in children: part 1: a clinical review; part 2: a new classification and management algorithm. *J Pediatr Orthop.* (1999) 19:559–69. doi: 10.1097/00004694-199909000-00001

5. Crozier-Shaw G, Mahon J, Bayer TC. The use of bioabsorbable compression screws & polyethylene tension band for fixation of displaced olecranon fractures. *J Orthop.* (2020) 22:525–9. doi: 10.1016/j.jor.2020.08.029

6. Gortzak Y, Mercado E, Atar D, Weisel Y. Pediatric olecranon fractures: open reduction and internal fixation with removable Kirschner wires and absorbable sutures. *J Pediatr Orthop*. (2006) 26:39–42. doi: 10.1097/01.bpo.0000187988.86892.a2

7. Romero JM, Miran A, Jensen CH. Complications and re-operation rate after tension-band wiring of olecranon fractures. *J Orthop Sci.* (2000) 5:318–20. doi: 10. 1007/s007760070036

8. Persiani P, Ranaldi FM, Graci J, De Cristo C, Zambrano A, D'Eufemia P, et al. Isolated olecranon fractures in children affected by osteogenesis imperfecta type I treated with single screw or tension band wiring system: outcomes and pitfalls in relation to bone mineral density. *Medicine (Baltimore)*. (2017) 96:e6766. doi: 10. 1097/MD.00000000006766

 Corradin M, Marengo L, Andreacchio A, Paonessa M, Giacometti V, Samba A, et al. Outcome of isolated olecranon fractures in skeletally immature patients: comparison of open reduction and tension band wiring fixation versus closed reduction and percutaneous screw fixation. *Eur J Orthop Surg Traumatol.* (2016) 26:469–76. doi: 10.1007/s00590-016-1774-y

10. Oitment C, Koziarz A, Bozzo A, Johal H. Intramedullary cancellous screw fixation of simple olecranon fractures. *Strategies Trauma Limb Reconstr.* (2019) 14 (3):163–7. doi: 10.5005/jp-journals-10080-1435

The funders had no role in the study design, data collection and analysis, decision to publish, or preparation of the manuscript.

#### Acknowledgments

The authors acknowledge the staff involved in this study for their valuable collaboration.

## 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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

11. Kim W, Choi S, Yoon JO, Park HY, Kim SH, Kim JS. Double tension band wiring for treatment of olecranon fractures. *J Hand Surg Am.* (2014) 39:2438–43. doi: 10.1016/j.jhsa.2014.09.020

 Çağlar C, Akçaalan S, Özaslan Hİ, Yağar H, Uğurlu M. Comparison of tension band wiring and plate fixation in mayo type 2A olecranon fractures. *Jt Dis Relat Surg.* (2021) 32:85–92. doi: 10.5606/ehc.2021.75812

13. Rao C, Hu H, Tan R. Comparative analysis of clinical effects of the cable-pin system and kirschner wire tension band (TBW) internal fixation in the treatment of the olecranon fracture. *Contrast Media Mol Imaging*. (2022) 2022:3073121. doi: 10. 1155/2022/3073121

14. Gruszka DS, Burkhart KJ, Nowak TE, Achenbach T, Rommens PM, Müller LP. The durability of the intrascaphoid compression of headless compression screws: in vitro study. *J Hand Surg Am*. (2012) 37:1142–50. doi: 10.1016/j.jhsa. 2012.03.018

15. Sinha S, Maharjan R, Khanal GP, Pokharel B, Drolia N, Gupta S, et al. Comparison of functional and radiological outcomes of olecranon fractures treated with tension band wiring with kirschner wires to transcortical screw fixation-A randomised controlled study. *Strategies Trauma Limb Reconstr.* (2020) 15:131–7. doi: 10.5005/jp-journals-10080-1510

16. Raju SM, Gaddagi RA. Cancellous screw with tension band wiring for fractures of the olecranon. *J Clin Diagn Res.* (2013) 7:339–41. doi: 10.7860/JCDR/2013/4450. 2762

17. Abzug JM, Herman MJ, Kozin S. Pediatric Elbow Fractures: A Clinical Guide to Management. Berlin: Springer (2018).

18. Rabinovich A, Adili A, Mah J. Outcomes of intramedullary nail fixation through the olecranon apophysis in skeletally immature forearm fractures. *J Pediatr Orthop.* (2005) 25:565–9. doi: 10.1097/01.bpo.0000173249. 04561.10

19. Mun F, Suresh KV, Hayashi B, Margalit A, Sponseller PD, Lee RJ. Compression screw fixation for pediatric olecranon fractures. *J Pediatr Orthop.* (2023) 43:e151–6. doi: 10.1097/BPO.00000000002284

20. Lopes RC, Carvalho M, Ling TP, Balacó I, Cardoso P, Tarquini O, et al. Operative pediatric hand trauma. *Glob Pediatr Health*. (2022) 9:2333794X221132462. doi: 10.1177/2333794X221132462 Check for updates

#### **OPEN ACCESS**

EDITED BY Gabriele Lisi, University of Studies G. d'Annunzio Chieti and Pescara, Italy

REVIEWED BY Giacomo Deiro, University of Turin, Italy Heba Taher, Cairo University, Egypt

\*CORRESPONDENCE Jixiao Zeng ⊠ zengjixiao@163.com

RECEIVED 28 August 2023 ACCEPTED 02 February 2024 PUBLISHED 19 February 2024

#### CITATION

Liang Z, Lan M, Xu X, Liu F, Tao B, Wang X and Zeng J (2024) Case Report: Robotic pylorus-preserving pancreatoduodenectomy for periampullary rhabdomyosarcoma in a 3-year-old patient. Front. Surg. 11:1284257. doi: 10.3389/fsurg.2024.1284257

#### COPYRIGHT

© 2024 Liang, Lan, Xu, Liu, Tao, Wang and Zeng. This is an open-access article distributed under the terms of the Creative Commons Attribution License (CC BY). The use, distribution or reproduction in other forums is permitted, provided the original author(s) and the copyright owner(s) are credited and that the original publication in this journal is cited, in accordance with accepted academic practice. No use, distribution or reproduction is permitted which does not comply with these terms.

# Case Report: Robotic pylorus-preserving pancreatoduodenectomy for periampullary rhabdomyosarcoma in a 3-year-old patient

Zijian Liang, Menglong Lan, Xiaogang Xu, Fei Liu, Boyuan Tao, Xinxing Wang and Jixiao Zeng<sup>\*</sup>

Department of Pediatric Surgery, Guangzhou Women and Children's Medical Center, Guangzhou Medical University, Guangzhou, Guangdong, China

Periampullary neoplasm is rare in pediatric patients and has constituted a strict indication for pancreatoduodenectomy (PD), which is a procedure sporadically reported in the literature among children. Robotic PD has been routinely performed for periampullary neoplasm in periampullary neoplasm, but only a few cases in pediatric patients have been reported. Here, we report the case of a 3-year-old patient with periampullary rhabdomyosarcoma treated with robotic pylorus-preserving PD and share our experience with this procedure in pediatric patients. A 3-year-old patient presented with obstructive jaundice and a mass in the pancreatic head revealed by imaging. A laparoscopic biopsy was performed. Jaundice progressed with abdominal pain and elevated alphaamylase leading to urgent robotic exploration in which a periampullary neoplasm was revealed and pathologically diagnosed as rhabdomyosarcoma by frozen section examination. After pylorus-preserving PD, we performed a conventional jejunal loop following a child reconstruction, including an endto-end pancreaticojejunostomy, followed by end-to-side hepaticojejunostomy and duodenojejunostomy. Delayed gastric emptying (DGE) presented with increasing drain from the nasogastric tube (NGT) a week after the surgery and improved spontaneously within 10 days. In a 13-month follow-up until the present, our case patient recovered well without potentially fatal complications, such as pancreatic fistula. Robotic PD in pediatric patients was safe and effective without intra- or postoperative complications.

#### KEYWORDS

robotic pancreatoduodenectomy, periampullary neoplasm, rhabdomyosarcoma, child, case report

## Introduction

Pancreatoduodenectomy (PD) was first introduced in a case report by Whipple et al. in 1935 (1). The pylorus-preserving modification was described by Traverso and Longmire in 1978 (2). Currently, PD is routinely performed for pancreatic head neoplasms. Parallel to the development of surgical instruments and the introduction of minimally invasive surgery, laparoscopic and robotic PD has become a common procedure and has been

reported to have better clinical outcomes compared with open surgery (3–6). However, laparoscopic and robotic PD for pediatric patients has only been sporadically reported in the literature (7–9). Here, we report the case of a 3-year-old patient with periampullary rhabdomyosarcoma treated with robotic pyloruspreserving pancreatoduodenectomy (PPPD) and share our experience with this procedure in pediatric patients. We present the following case in accordance with the SCARE criteria (10).

## **Case description**

A 3-year-old Chinese patient was referred to our hospital because of white, clay-like stools and a 34 mm  $\times$  29 mm  $\times$  34 mm mass in the lower part of the biliary tract and pancreatic uncinate process revealed by ultrasonic examination. The patient had intermittent abdominal pain and did not have any signs of fever, abdominal distention, emesis, or skin itching in the previous 20 days. No eventful history was found after a detailed consultation. Physical examination revealed slight conjunctival jaundice without any abdominal abnormal signs. Laboratory tests revealed elevated liver enzymes [alanine aminotransferase (ALT) 455 U/L, aspartate aminotransferase (AST) 366 U/L, gamma-glutamyl transferase (GGT) 1,637 U/L] and obstructive jaundice with total bilirubin of 74.2  $\mu$ mol/L and direct bilirubin of 52.6  $\mu$ mol/L. Carbohydrate antigen-199 (Ca-199) was 3,752.91 IU/ml and neuron-specific enolase (NSE) was 56.21 ng/ml.

Computed tomography (CT) scan of the abdomen and pelvis with an intravenous contrast agent revealed an ill-defined 2.9 cm  $\times$  2.8 cm mass with delay enhancement in the uncinate process and common bile duct (CBD) and dilation in the biliary system (Figures 1A,C,E). Magnetic resonance (MR) imaging showed a 2.6 cm  $\times$  3.6 cm  $\times$  2.1 cm mass assumed to originate in the CBD, presenting an equal signal on T1WI and an equal-high signal on T2WI, which caused a thorough cutoff of the CBD (Figures 1G,H). Proximal biliary tract diameter was 2.2 cm while the MPD measured 0.1 cm. Positron emission tomography (PET) did not show any extrapancreatic disease.

As malignant pancreatic tumors are rare in children, the diagnosis with the pathological result would be the key for further management. A laparoscopic biopsy with cholecystostomy drainage was performed. The cholecystostomy for biliary drainage was successful as the level of bilirubin decreased; however, the biopsy was negative. While waiting 1 week for the pathological result, we performed another CT scan, which showed that the mass had enlarged to 4.4 cm  $\times$ 4.3 cm (Figures 1B,D,F) while alpha-amylase had risen to 549 U/L. Surgery was decided upon due to the rapid evolution of tumor size and increased signs of abdominal pain. After malignancy was confirmed by frozen section examination, a PPPD with an end-to-end pancreatojejunostomy followed by end-to-side hepaticojejunostomy and duodenojejunostomy was performed. Liver enzymes, bilirubin, alpha-amylase, and Ca-199 gradually decreased to normal levels within one week after the surgery. Amylase levels in the drainage fluid, tested every





CT and MR results before radical surgery. (A,C,E) CT scan of the abdomen and pelvis with intravenous contrast agent showing an ill-defined 2.9 cm  $\times 2.8$  cm mass with delayed enhancement of the uncinate process with proximal dilatation. (G,H) MR imaging showing a 2.6 cm  $\times 3.6$  cm  $\times 2.1$  cm mass originating from the common bile duct (CBD), presenting an equal signal on T1WI and an equal-high signal on T2WI. The common bile duct (CBD) was plugged up completely by the mass, resulting in the dilatation of the proximal biliary system. The dilatation of the common bile duct (CBD) reached 2.2 cm in diameter, and the pancreatic duct also presented a slight dilation of 0.1 cm. (B,D,F) CT scan for reevaluation showing the enlarged mass.

3 days, were negative. The drains were removed when the daily drains were <20 ml and lasted for more than 3 days. The nasogastric tube (NGT) was removed 17 days postoperatively as the drain had dropped to under 20 ml in the last 3 days, and the patient was able to maintain unlimited oral intake in 21 days postoperatively. Delayed gastric emptying (DGE) was defined as Grade B according to the International Study Group of Pancreatic Surgery (11). Pathological examination with desmin, myogenin, and myoD1 staining confirmed the diagnosis of embryonal RMS originating from the ampulla (Figure 2). Because of negative surgical margins and the absence of lymph node involvement, the patient was classed as low risk and was recommended to undergo chemotherapy after fully recovering from surgery. At 13 months of follow-up, the patient has tolerated chemotherapy well and has shown no signs of recurrence.



#### FIGURE 2

Pathological results confirmed the diagnosis of rhabdomyosarcoma. In hematoxylin–eosin staining, the lesion was located under the epithelium and formed a neoplastic layer parallel to the epithelium with small round atypic cells with obvious heteromorphism (A), and positive markers such as desmin (B), myoD1 (C), and myogenin (D) staining confirmed the diagnosis of embryonal RMS originated in the ampulla.

#### Surgical procedure

The patient was placed in the supine position. A five-port approach is shown in Figure 3A with the da Vinci Xi Surgical System. After creating a pneumoperitoneum (8 mmHg), we placed a 8 mm trocar at the umbilicus. Next, the remaining 8 mm robotic arm ports were inserted under safe vision. The first robotic arm (R1) was placed along the left midclavicular line crossing with the transverse umbilical line. The second robotic arm (R2) was placed along the right midclavicular line crossing 2 cm below the transverse umbilical line. The third robotic arm (R3) was placed under the costal margin crossing with the right anterior axillary line. Another 5 mm trocar was placed for the assistant surgeon to control suction and pass sutures.

A brief view of the abdominal cavity showed no metastasis lesions, and the CBD was dilated to a maximum diameter of 2 cm. The gastrocolic ligament was divided using the harmonic scalpel, and the stomach was lifted upward with two transabdominal stay sutures so that the enlarged pancreatic head was exposed. Next, we performed a choledochotomy where a neoplasm that resembled a cluster of grapes was revealed (Figure 3B). Biopsy was taken from both the neoplasm in the CBD and pancreatic head for fast-frozen section examination. The neoplasm showed small round atypic cells with obvious heteromorphism within the lesion; therefore, a malignant pancreatic head tumor was suspected, and the PD procedure was indicated.

After performing a 5-0 polydioxanone suture (PDS) in the biopsy site for hemostasis and closure of the tumor, we identified the anatomy in the hepatic hilum. Next, we identified the common hepatic artery (CHA), gastroduodenal artery (GDA), portal vein (PV), and biliary system, ligated the GDA at its origin, and identified and dissected the superior margin of the pancreas. During the dissection, the superior mesenteric vein (SMV) and the branches, including the superior right colic vein, the right gastroepiploic vein, and the gastrocolic trunk, were carefully divided. Then, we moved the horizontal portion of the duodenum and the dorsal portion of the pancreatic head using the Kocher maneuver and partially divided the Treitz ligament and duodenojejunal flexure for better transection of the first jejunal loop. Following the identification and ligation of the cystic artery, the gallbladder and the CBD were dissected and removed. The duodenum was then transected distal to the pylorus (Figure 3C). Simultaneously, we performed lymph node dissection of the coeliac trunk, hepatoduodenal ligament, SMV, the right side of the superior mesenteric artery, and the



#### FIGURE 3

Robotic dissection in pylorus-preserving pancreatoduodenectomy (PPPD). (A) Port set up during surgery. (B) A neoplasm resembled a cluster of grapes inside the common bile duct (CBD). (C) The duodenum was transected at 2 cm distal to the pylorus. (D) The skeletonized SMV/SMA and dissected retroperitoneum with peripancreatic soft tissue and the nerve plexuses removed. (E) After the specimen was taken out, a stump of the common hepatic duct, pancreas, and duodenum. (F) A 3F ureteral stent tube was inserted into the MPD as an internal stent and fixed to the MPD with 4-0 Prolene.

lymphatic tissue behind the pancreatic head; the specimens were sent for pathological examination (Figure 3D).

Next, we began to divide the pancreas and SMV from the pancreatic head to create the pancreatic tunnel using a harmonic scalpel. Through the tunnel, the transection of the pancreatic parenchyma was made until the main pancreatic duct (MPD) was identified and cut by scissors. We found a 1.5 mm MPD in the posteriolateral portion of the pancreas. An end-to-end pancreatojejunostomy was preferred to avoid anastomotic stenosis. We dissociated the stump from the tissues around the pancreas for 2 cm, which were used later for anastomosis.

After the dissection was completed, the specimens were put into an Endo Bag to avoid tumor dissemination. The margin of the duodenum, pancreas, and dissected peripyloric lymph nodes were sent for fast-frozen section examination; a negative result supported our choice for a PPPD.

A child reconstruction was performed sequentially: an endto-end pancreaticojejunostomy, followed by an end-to-side



FIGURE 4

Robotic child reconstruction and specimens. (A) End-to-end pancreatojejunostomy. (B) End-to-side single layer barbed suture hepaticojejunostomy. (C) The distal and free portion of omentum was fixed with 5-0 Prolene suture around the pancreatojejunostomy anastomosis as a mattress. (D) End-to-side single-layer barbed suture duodenojejunostomy. (E) Specimens removed with the Endo Bag.

hepaticojejunostomy and duodenojejunostomy (Figure 3E). A 3F ureteral stent tube was inserted into the MPD as an internal stent and fixed with 4-0 Prolene (Figure 3F). Then a single layer of continuous suturing was performed in anterior and posterior anastomosis between the pancreatic stump and dissected jejunum, respectively. To avoid pancreatic leaks, we passed every suture through the entire layer of the jejunum and vertically into the pancreas. Finally, both ends of the sutures in the anterior and posterior walls were tightened and tied for complete coverage by the jejunal end (Figure 4A). Before the hepaticojejunostomy and duodenojejunostomy anastomosis was performed, the jejunal loop was rotated behind the mesenteric root, and hepaticojejunostomy was created where the loop was placed at the hepatic hilum without any tension. The hepaticojejunostomy anastomosis was performed in a single layer with a barbed suture, posterior and anterior, respectively (Figure 4B). The distal and free portion of the omentum was fixed with a 5-0 Prolene suture around the pancreatojejunostomy anastomosis as a mattress (Figure 4C). Then the jejunal loop was placed in an antecolic position, and end-toside duodenojejunostomy was created. The duodenojejunostomy

was performed in a single layer with a barbed suture, posterior and anterior, separately (Figure 4D). Three drains were placed close to the anastomoses. After being taken out with the Endo Bag, the specimens were dissected; these revealed a neoplasm that originated in the ampulla (Figure 4E).

#### Discussion

RMS is a rare malignant tumor morphologically akin to the skeletal muscle (12). Its incidence rate is 4.5 cases per million people per year, occurring more often in children than in adults (13). In children, the most common sites include the head and neck, genitourinary tract, and extremities; the retroperitoneum or biliary tract has only been sporadically reported in the literature (14, 15). The disease presents differently according to the involved site. Diagnosis is usually made by direct biopsy (12, 14–16). Multidisciplinary treatments such as chemotherapy, radiation, and surgery have helped improve the survival rate to 70% over the past 30 years (17).

Our intraoperative decision to perform PPPD was fully justified as our patient had no direct invasion into the surrounding organs or any peripyloric lymph node metastases, as confirmed by a frozen section examination (4, 18). The rationale behind PPPD was to reduce complications following gastric resection, such as diarrhea, dumping, ulceration, and bile reflux gastritis (18, 19). Other reported advantages are shorter surgical time (4, 19), less intraoperative blood loss (4, 19) with similar complications (4, 19), reoperation (4), mortality (4), and cumulative survival rates (4) compared with standard PD. However, the effectiveness of PPPD has been doubted since it was practiced clinically for the common postoperative complication DGE and a compromised resection that may fail to reach R0 resection. In our case, we found DGE as Grade B and performed R0 resection successfully, and lymph nodes were confirmed by pathological results (11). DGE is one of the most common postoperative complications in PPPD and is thought to be caused by damage to the gastroenteric nervous system during surgery (18). DGE is reported to be transient and will be recovered with gastric suction for more than 10 days (18). In our case, the drain from the NGT gradually decreased, and meals became tolerated 17 days after the surgery, which was consistent with the literature. An early clinical report by Roder et al. (5) argued that an incomprehensive resection in PPPD may lead to a failure of R0 resection. Other later studies have rejected this opinion, suggesting that experienced surgeons can successfully perform a standard PPPD with complete removal of lesions and lymph nodes, leading to a successful R0 resection (4, 18). In conclusion, PPPD was a safe and effective surgical procedure for cancer in the pancreatic head in selected cases (4, 18).

Various suture techniques for pancreatojejunostomy have been introduced, but none of these techniques have been accepted as being optimal. The most popular techniques include end-to-side duct-to-mucosa anastomosis and end-to-end dunking or invagination anastomosis (20, 21); other methods were mostly modified from the above techniques (22–24). End-to-side duct-to-mucosa anastomosis is one of the most popular techniques, as it is thought as the most histologically compatible and has been reported to have excellent results in adults (5, 22, 25). Spagnoletti et al. performed PPPD and reported a successful end-to-side duct-to-mucosa anastomosis in a 5-month-old patient; the authors determined the method to be safe for pediatric patients (7). However, the authors also warned that the small MPD should be carefully managed during the suture in case of tearing the fragile tissue (7). Narrow MPD was a relative contraindication for duct-to-mucosa anastomosis (26). This technique is a complex procedure that requires sutures through the fragile tissue of the pancreas and has a high risk of tearing (27), especially in pediatric patients. Therefore, our team chose single-layer continuous sutures in the pancreatojejunostomy anastomosis with a stent to avoid tissue tearing and stenosis of the MPD. Our experience in end-to-end pancreatojejunostomy in pediatric patients may provide a reference for other surgeons who encounter similar cases.

Postoperative pancreatic fistula is the most challenging complication in PD. A pancreatic fistula is defined as any measurable volume of drain fluid on or after postoperative day 3 with an amylase level elevated to more than three times the upper limit of normal amylase (28). Our team sent the fluid to the anastomotic site for amylase examination every 3 days to exclude the pancreatic fistula. In our case, the patient recovered well without leak or fistula formation. Pancreatic leak and fistula formation are mostly related to the decrease in rate in the technique of pancreatic anastomosis and precise suturing of anastomoses by surgeons (21). In our experience, with the assistance of a modern robotic system, the three-dimensional field of vision made the anatomy of the pancreatic duct so clear that the anastomosis could be precisely sutured. However, no significant difference was found in the rate of pancreatic fistula between the robotic and open PD in recent studies (29-31). In addition to the suture technique, there are multiple factors responsible for pancreatic fistula formation after PD. Avoidance of pancreatic fistula has still a long way to go.

The robotic system used in the present study provided a wide three-dimensional field of vision, flexible tools, EndoWrist allowing seven degrees of freedom, and a steady traction without physiological tremor and allowed better control of hemostasis and precise dissection of tissues and hard sutures (32). After being practiced for more than a decade, the robotic approach is thought to be a major improvement over the traditional laparoscopic approach and could be applied to more complex procedures in a minimally invasive way (32). PD is widely accepted as one of the most complicated procedures in general surgery because of the wide dissection and three anastomoses involved. Robotic PD in adults has been reported to have better efficacy and safety (3, 25, 33, 34). Despite the reported advantages of robotic surgery, this approach is rarely used in pediatric surgery with only a few published reports (7-9). Our patient recovered well without complications, which proves robotic PD in pediatric patients to be safe and effective.

In conclusion, we presented here the first and youngest case of robotic PPPD for periampullary RMS in a pediatric patient. With the assistance of a modern robotic system, we performed an R0 resection and a child reconstruction with end-to-end pancreatojejunostomy, end-to-side hepaticojejunostomy, and duodenojejunostomy. Although DGE was found in our case, the patient recovered quickly within 3 weeks without fatal complications such as pancreatic fistula or leak. The case reported here demonstrates robotic PD in pediatric patients to be safe and effective without intra- or postoperative complications. However, further studies with longer follow-ups are required to evaluate clinical results.

#### 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 the Ethics Committee of Guangzhou Women and Children's Medical Center (GWCMC) [Approval No. (2023)061A01]. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation in this study was provided by the participants' legal guardians/next of kin. Written informed consent was obtained from the individual(s), and minor(s)' legal guardian/next of kin, for the publication of any potentially identifiable images or data included in this article.

#### Author contributions

ZL: conceptualization, data curation, writing – original draft, writing – review and editing. ML: data curation, writing – review

#### References

1. Whipple AO, Parsons WB, Mullins CR. Treatment of carcinoma of the ampulla of Vater. Ann Surg. (1935) 102:763–79. doi: 10.1097/00000658-193510000-00023

2. Traverso LW, Longmire WP Jr. Preservation of the pylorus in pancreaticoduodenectomy. Surg Gynecol Obstet. (1978) 146:959–62.

3. Baimas-George M, Watson M, Murphy KJ, Iannitti D, Baker E, Ocuin L, et al. Robotic pancreaticoduodenectomy may offer improved oncologic outcomes over open surgery: a propensity-matched single-institution study. *Updates Surg.* (2020) 34:3644–9. doi: 10.1007/s00464-020-07564-x

4. Seiler CA, Wagner M, Bachmann T, Redaelli CA, Schmied B, Uhl W, et al. Randomized clinical trial of pylorus-preserving duodenopancreatectomy versus classical Whipple resection-long term results. *Br J Surg.* (2005) 92:547–56. doi: 10.1002/bjs.4881

5. Roder JD, Stein HJ, Hüttl W, Siewert JR. Pylorus-preserving versus standard pancreatico-duodenectomy: an analysis of 110 pancreatic and periampullary carcinomas. *Br J Surg.* (1992) 79:152–5. doi: 10.1002/bjs.1800790219

6. Liu R, Zhang T, Zhao ZM, Tan XL, Zhao GD, Zhang X, et al. The surgical outcomes of robot-assisted laparoscopic pancreaticoduodenectomy versus laparoscopic pancreaticoduodenectomy for periampullary neoplasms: a comparative study of a single center. *Surg Endosc.* (2017) 31:2380–6. doi: 10.1007/s00464-016-5238-6

7. Spagnoletti G, Larghi Laureiro Z, Maria Fratti A, Maiorana A, Garganese MC, Pizzoferro M, et al. Case report: pylorus-preserving pancreatoduodenectomy for

and editing. XX: writing – review and editing. FL: writing – review and editing. BT: validation, writing – review and editing. XW: data curation, writing – review and editing. JZ: supervision, writing – review and editing.

#### Funding

The authors declare that no financial support was received for the research, authorship, and/or publication of this article.

This work was supported by grants from the National Natural Science Foundation of China (grant no. 82170528), Natural Science Foundation of Guangdong Province (grant no. 2022A1515012254), Guangzhou Science and Technology Plan Municipal University (Institute) Joint Funding Project (grant no. 202201020612), Guangzhou Science and Technology Projects (grant no. 202201011846), and Guangzhou Clinical Characteristic Technology Project (grant no. 2023C-TS48).

## **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

All claims expressed in this article are solely those of the authors and do not necessarily represent those of their affiliated organizations, or those of the publisher, the editors and the reviewers. Any product that may be evaluated in this article, or claim that may be made by its manufacturer, is not guaranteed or endorsed by the publisher.

focal congenital hyperinsulinism in a 5-month-old baby. Front Surg. (2022) 9:1085238. doi: 10.3389/fsurg.2022.1085238

8. Hagendoorn J, Nota CLMA, Borel Rinkes IHM, Molenaar IQ. Robotic pancreatoduodenectomy for a solid pseudopapillary tumor in a ten-year-old child. *Surg Oncol.* (2018) 27:635–6. doi: 10.1016/j.suronc.2018.07.013

9. Chinnusamy P, Dey S, Chittawadagi B, Gurumurthy S, Sabnis S, Palanisamy S. First robotic pylorus preserving pancreaticoduodenectomy for Frantz's tumour in an adolescent girl. J Minim Access Surg. (2018) 14:349–53. doi: 10.4103/jmas.JMAS\_272\_17

10. Agha RA, Fowler AJ, Saeta A, Barai I, Rajmohan S, Orgill DP, et al. The SCARE statement: consensus-based surgical case report guidelines. *Int J Surg.* (2016) 34:180–6. doi: 10.1016/j.ijsu.2016.08.014

11. Wente MN, Bassi C, Dervenis C, Fingerhut A, Gouma DJ, Izbicki JR, et al. Delayed gastric emptying (DGE) after pancreatic surgery: a suggested definition by the International Study Group of Pancreatic Surgery (ISGPS). *Surgery*. (2007) 142:761-8. doi: 10.1016/j.surg.2007.05.005

12. Gartrell J, Pappo A. Recent advances in understanding and managing pediatric rhabdomyosarcoma. *F1000Res.* (2020) 9:685. doi: 10.12688/f1000research. 22451.1

13. Rogers TN, Dasgupta R. Management of rhabdomyosarcoma in pediatric patients. Surg Oncol Clin N Am. (2021) 30:339–53. doi: 10.1016/j.soc.2020.11.003 14. Hunt GC, Corless CL, Terry AB, Katon RM. Embryonal rhabdomyosarcoma in a 3-year-old child diagnosed at ERCP: case report and review. *Gastrointest Endosc.* (2002) 56:445–7. doi: 10.1016/S0016-5107(02)70060-6

15. Hayes-Jordan A, Andrassy R. Rhabdomyosarcoma in children. Curr Opin Pediatr. (2009) 21:373-8. doi: 10.1097/MOP.0b013e32832b4171

16. Wang C. Childhood rhabdomyosarcoma: recent advances and prospective views. J Dent Res. (2012) 91:341–50. doi: 10.1177/0022034511421490

17. Yohe ME, Heske CM, Stewart E, Adamson PC, Ahmed N, Antonescu CR, et al. Insights into pediatric rhabdomyosarcoma research: challenges and goals. *Pediatr Blood Cancer*. (2019) 66:e27869. doi: 10.1002/pbc.27869

18. Ogata Y, Hishinuma S. The impact of pylorus-preserving pancreatoduodenectomy on surgical treatment for cancer of the pancreatic head. *J Hepatobiliary Pancreat Surg.* (2002) 9:223–32. doi: 10.1007/s005340200023

19. Ohtsuka T, Yamaguchi K, Ohuchida J, Inoue K, Nagai E, Chijiiwa K, et al. Comparison of quality of life after pylorus-preserving pancreatoduodenectomy and Whipple resection. *Hepatogastroenterology.* (2003) 50:846–50.

20. Kennedy EP, Yeo CJ. Dunking pancreaticojejunostomy versus duct-to-mucosa anastomosis. J Hepatobiliary Pancreat Sci. (2011) 18:769–74. doi: 10.1007/s00534-011-0429-v

21. Braasch JW, Gagner M. Pylorus-preserving pancreatoduodenectomytechnical aspects. *Langenbecks Arch Chir.* (1991) 376:50-8. doi: 10.1007/ BF00205128

22. Torer N, Ezer A, Nursal TZ. Mattress sutures for the modification of end-to-end dunking pancreaticojejunostomy. *Hepatobiliary Pancreat Dis Int.* (2013) 12:556–8. doi: 10.1016/S1499-3872(13)60088-0

23. De Pastena M, Bannone E, Andreotti E, Filippini C, Ramera M, Esposito A, et al. Pancreatic anastomosis in robotic-assisted pancreaticoduodenectomy: different surgical techniques. *Dig Surg.* (2023) 40(1–2):1–8. doi: 10.1159/000528646

24. Machado MA, Mattos BV, Lobo Filho MM, Makdissi FF. A new technique of duodenojejunostomy may reduce the rate of delayed gastric emptying after pylorus-preserving pancreatoduodenectomy: the growth factor technique (with video). *Surg Oncol.* (2023) 46:101902. doi: 10.1016/j.suronc. 2023.101902

25. Ng CP, Yang GP, Tang CN. Robot-assisted Whipple's operation for solid pseudopapillary tumour of pancreas. J Robot Surg. (2013) 7:91-3. doi: 10.1007/s11701-012-0362-3

26. Batignani G, Fratini G, Zuckermann M, Bianchini E, Tonelli F. Comparison of Wirsung-jejunal duct-to-mucosa and dunking technique for pancreatojejunostomy after pancreatoduodenectomy. *Hepatobiliary Pancreat Dis Int.* (2005) 4:450–5.

27. Rao AC, Gabriel G, Serrano J, Benedicto R. Inkwell pancreaticojejunal anastomosis after pancreaticoduodenectomy. *Am J Surg.* (2004) 187:410–2. doi: 10. 1016/j.amjsurg.2003.12.023

28. Bassi C, Marchegiani G, Dervenis C, Sarr M, Abu Hilal M, Adham M, et al. The 2016 update of the International Study Group (ISGPS) definition and grading of postoperative pancreatic fistula: 11 years after. *Surgery*. (2017) 161:584–91. doi: 10. 1016/j.surg.2016.11.014

29. Zhao W, Liu C, Li S, Geng D, Feng Y, Sun M. Safety and efficacy for robot-assisted versus open pancreaticoduodenectomy and distal pancreatectomy: a systematic review and meta-analysis. *Surg Oncol.* (2018) 27:468–78. doi: 10.1016/j.suronc.2018.06.001

30. Boggi U, Palladino S, Massimetti G, Vistoli F, Caniglia F, De Lio N, et al. Laparoscopic robot-assisted versus open total pancreatectomy: a case-matched study. *Surg Endosc.* (2015) 29:1425–32. doi: 10.1007/s00464-014-3819-9

31. Shin SH, Kim YJ, Song KB, Kim SR, Hwang DW, Lee JH, et al. Totally laparoscopic or robot-assisted pancreaticoduodenectomy versus open surgery for periampullary neoplasms: separate systematic reviews and meta-analyses. *Surg Endosc.* (2017) 31:3459–74. doi: 10.1007/s00464-016-5395-7

32. Parisi A, Desiderio J, Trastulli S, Cirocchi R, Renzi C, Boselli C, et al. Robotic pylorus-preserving pancreaticoduodenectomy: technical considerations. *Int J Surg.* (2015) 21(Suppl 1):S59–63. doi: 10.1016/j.ijsu.2015.06.061

33. Shyr YM, Wang SE, Chen SC, Shyr BU, Shyr BS. Robotic pancreaticoduodenectomy for pancreatic head cancer and periampullary lesions. *Ann Gastroenterol Surg.* (2021) 5:589–96. doi: 10.1002/ags3.12457

34. Da Dong X, Felsenreich DM, Gogna S, Rojas A, Zhang E, Dong M, et al. Robotic pancreaticoduodenectomy provides better histopathological outcomes as compared to its open counterpart: a meta-analysis. *Sci Rep.* (2021) 11:3774. doi: 10.1038/s41598-021-83391-x

# Frontiers in **Pediatrics**

## Addresses ongoing challenges in child health and patient care

Explores research that meets ongoing challenges in pediatric patient care and child health, from neonatal screening to adolescent development.

# **Discover the latest Research Topics**



#### Frontiers

Avenue du Tribunal-Fédéral 34 1005 Lausanne, Switzerland frontiersin.org

#### Contact us

+41 (0)21 510 17 00 frontiersin.org/about/contact



