Supplementary information

# SUPPLEMENTARY TABLE 1 Search terms used for literature search of the PubMed database (14 July 2023).

|  |  |  |  |
| --- | --- | --- | --- |
| Thematic group | Search number | Search terms | Number of results |
| Cost | 1 | ("Costs and Cost Analysis"[MeSH Terms] OR "Fees and Charges"[MeSH Terms] OR "Budgets"[MeSH Terms] OR "budget\*"[Title/Abstract] OR "economic\*"[Title/Abstract] OR "cost"[Title/Abstract] OR "costs"[Title/Abstract] OR "costly"[Title/Abstract] OR "costing"[Title/Abstract] OR "price"[Title/Abstract] OR "prices"[Title/Abstract] OR "pricing"[Title/Abstract] OR "pharmacoeconomic\*"[Title/Abstract] OR "pharmaco economic\*"[Title/Abstract] OR "expenditure"[Title/Abstract] OR "expenditures"[Title/Abstract] OR "expense"[Title/Abstract] OR "expenses"[Title/Abstract] OR "financial"[Title/Abstract] OR "finance"[Title/Abstract] OR "finances"[Title/Abstract] OR "financed"[Title/Abstract] OR "value for money"[Title/Abstract] OR "monetary value\*"[Title/Abstract]) | 1,099,224 |
| Economic modelling | 2 | ("models, economic"[MeSH Terms] OR "economic model\*"[Title/Abstract] OR "Decision Theory"[MeSH Terms] OR "decision tree\*"[Title/Abstract] OR "decision analy\*"[Title/Abstract] OR "decision model\*"[Title/Abstract]) OR "Economics"[MeSH Terms:noexp]) | 47,924 |
| Autoimmune conditions | 3 | ("autoimmune diseases of the nervous system"[MeSH Terms] OR ("autoimmune"[All Fields] AND "diseases"[All Fields] AND "nervous"[All Fields] AND "system"[All Fields]) OR "autoimmune diseases of the nervous system"[All Fields] OR ("autoimmune"[All Fields] AND "encephalitis"[All Fields]) OR "autoimmune encephalitis"[All Fields] OR ("oligodendrocyte myelin glycoprotein"[MeSH Terms] OR ("oligodendrocyte myelin"[All Fields] AND "glycoprotein"[All Fields]) OR "oligodendrocyte myelin glycoprotein"[All Fields] OR ("myelin"[All Fields] AND "oligodendrocyte"[All Fields] AND "glycoprotein"[All Fields]) OR "myelin oligodendrocyte glycoprotein"[All Fields] OR "myelin oligodendrocyte glycoprotein"[MeSH Terms] OR ("myelin oligodendrocyte"[All Fields] AND "glycoprotein"[All Fields]) OR "myelin oligodendrocyte glycoprotein"[All Fields] OR ("myelin"[All Fields] AND "oligodendrocyte"[All Fields] AND "glycoprotein"[All Fields]) | 94,685 |
| Exclusions | 4 | ("multiple sclerosis"[MeSH Terms] OR ("multiple"[All Fields] AND "sclerosis"[All Fields]) OR "multiple sclerosis"[All Fields]) | 84,507 |
|  | 5 | (2000/1/1:2023/6/14[pdat]) | 22,368,570 |
| Total | 6 | (((#1 OR #2) AND #3) NOT #4) AND 5 | 512 |

MeSH, medical subject heading.

### SUPPLEMENTARY TABLE 2 Further details of studies included in the review.

| **Disease(s)** | **Citation** | **Study year** | **Cost year, currency** | **Study type** | **Country / countries or region(s)** | **World Bank income level (1)** | **Number of patients with the disease(s) of interest** | **Sex (males), %** | **Age, years, median** | **Acute attack rate** |
| --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- |
| Autoimmune encephalitis | Cai et al., 2022 (2) | 2014–2020 | CNY | Cost-efficiency analysis (retrospective, comparison of IV methylprednisolone monotherapy vs IV methylprednisolone plus IVIg combination therapy) | China | Upper middle | 78 | 61.5 | 40 (IQR, 24–58) | NR |
| Autoimmune encephalitis | Cohen et al., 2019 (3) | 2005–2015 | 2017, USD | Cost analysis (retrospective; includes comparison of antibody-positive vs. antibody-negative patients) | US | High | 63 | 46.0 | Mean, 47.2 (SE, ±2.3) | NR |
| Autoimmune encephalitis | Li et al., 2020 (4) | 2012–2018 | 2018, RMB (selected values also reported in USD) | Cost analysis (retrospective, non-comparative) | Western China | Upper middle | 208 | 49.0 | 29 (IQR, 20–44) | NR |
| Autoimmune encephalitis | Sharp et al., 2021 (5) | 2018–2019 | 2018–2019, USD | Cost analysis (relating to autoimmune encephalopathy testing panels; costs compared before and after implementing an algorithm for ordering panels) | US | High | 58 patients in 2018 and 53 patients during the first 7 months of 2019 | NR | NR | NR |
| CIDP | Allen et al., 2020 (6) | 2004–2005 and 2012–2016 | 2019, USD | Hypothetical cost analysis (reported within a review article; includes comparison of low-dose SCIg, high-dose SCIg and IVIg) | US | High | 289 | NR | NR | NR |
| CIDP | Darbà & Marsà, 2022 (7) | 2004–2018 | 2017, EUR | Cost analysis (retrospective; includes comparison of two different time periods (2004–2015 vs 2015–2016) | Spain | High | 2,805 | 64.7 | 60 (95% CI, 59–61) | NR |
| CIDP | Divino et al., 2018 (8) | 2010–2014 | 2016, USD | Cost analysis (case-control study, retrospective) | US | High | 790 | 53.7 | 52 | NR |
| CIDP | Guptill et al., 2014 (9) | 2011 | USD | Analysis of costs and healthcare resource use (retrospective, non-comparative) | US | High | 73 | 56.0 | 51 | NR |
| CIDP | Le Masson et al., 2018 (10) | 2012–2014 | 2016, EUR | Cost minimisation analysis (before and after analysis with prospective data collection; comparison of home- vs hospital-administered IVIg) | France | High | 24 | 2:1 (M:F) | Mean, 52.3 (SD, 12.2) | NR |
| CIDP | Mahdi-Rogers et al., 2014 (11) | 2008 | 2006–2007, GBP | Cost-of-illness study (patient survey, non-comparative) | UK | High | 43 | 65.1 | Mean, 65.0 (12.6) | NR |
| CIDP | McCrone et al., 2003 (12) | 1998–1999 | 2000–2001, GBR converted to EUR using Oct 2022 exchange rate | Cost–utility analysis (comparison of prednisolone vs IVIg therapy) | Belgium, Czech Republic, Greece, Italy, The Netherlands, Spain, UK | High | 25 | Prednisolone:  69  IVIg:  58 | Prednisolone:  Mean, 53.9 (SD, 17.3)  IVIg:  Mean, 52.0 (SD, 13.6 | NR |
| CIDP | Mengel et al., 2018 (13) | 2013–2014 | 2013, EUR | Cost-of-illness study (patient survey, non-comparative) | Germany | High | 108 | 77.8 | Mean, 63.1 (±12.6) | NR |
| CIDP | Perraudin et al., 2020 (14) | N/A | 2020, CHF | Cost minimalisation analysis (comparison of hospital-based IVIg vs home-based SCIg) | Switzerland | High | N/A (modelling study) | N/A | N/A | NR |
| CIDP | Piscitelli et al., 2021 (15) | N/A | NR, EUR | Cost analysis (retrospective; comparison of SCIg vs IVIg) | Italy | High | 12 | 30.8\* | Mean, 56 (±15)\* | NR |
| CIDP | Rajabally et al., 2019 (16) | 2014–2018 | 2018, GBP and EUR | Retrospective study (review of hospital patient records; comparison of different dosing methods for IVIg) | UK | High | 39 | 74.4 | Mean, 58.4 (SD, 15.1) | NR |
| Guillain–Barré syndrome | Frenzen, 2008 (17) | 2004 | NR, USD | Cost analysis (non-comparative) | US | High | [Pan-US, patients hospitalised] 5,473 | NR | NR | NR |
| Guillain–Barré syndrome | Maheshwari et al., 2018 (18) | 2012–2014 | 2012–2013, INR and USD | Cost minimisation analysis (includes comparison of IVIg vs plasmapheresis therapy) | India | Lower middle | 40 | 67.4 | Mean, 34.0 (SD, ± 17.1) | NR |
| Guillain–Barré syndrome | Oliveira et al., 2022 (19) | 2017–2019 | 2018, USD | Cost-of-illness study (patient survey, non-comparative) | Brazil | Upper middle | 46 | 65.5 | 42 (IQR, 28–52) | NR |
| Guillain–Barré syndrome | Rumalla et al., 2017 (20) | 2002–2011 | 2011, USD | Retrospective study (review of data from the US Nationwide Inpatient Sample, comparison of patients with Guillain–Barré syndrome with vs without hyponatraemia) | US | High | 54,778 | No hyponatremia:  55.9  Hyponatremia:  52.6 | No hyponatremia:  Mean, 52.6 (SD, 17.8)  Hyponatremia:  Mean, 59.5 (SD, 16.3) | NR |
| Guillain–Barré syndrome | Tsai et al., 2007 (21) | 1999–2004 | 2005, NTD | Cost-effectiveness study (retrospective; includes comparison of IVIg vs plasma exchange therapy) | Taiwan | High | 24 | PLEX:  70  IVIg:  86  Control:  86 | PLEX:  Mean, 61.0 ± 17.4  IVIg:  Mean, 45.0 ± 25.0  Control:  Mean, 49.1 ± 23.0 | NR |
| Guillain–Barré syndrome | van Leeuwen et al., 2016 (22) | 2009–2010 | EUR | Analysis of resource use and costs (non-comparative) | The Netherlands | High | 87 | 56.0 | 49 (IQR, 30–64) | NR |
| Guillain–Barré syndrome | Winters et al., 2011 (23) | 2010–2011 | 2011, USD | Cost minimisation analysis (comparison of IVIg vs plasma exchange) | US | High | Not applicable (modelling study) | NR | NR | NR |
| MOGAD and NMOSD† | Hümmert et al., 2022 (24) | 2017–2019 | 2018, EUR | Analysis of costs and HRQoL (patient survey, non-comparative) | Germany | High | 212 | 20 | 50 (range, 19–83) | NR |
| Myasthenia gravis | Fan et al., 2020 (25) | Not reported | CNY | HRQoL study (cross-sectional, non-comparative) | China | Upper middle | 69 | 62.3 | Mean, 54.7 (±13.7) | NR |
| Myasthenia gravis | Guptill et al., 2011 (26) | 2008–2010 | USD | Cost analysis (includes comparison of costs for IVIg vs plasma exchange therapy) | US | High | 1,288 | 41 | Mean, 59.8 | NR |
| Myasthenia gravis | Guptill et al., 2012 (27) | 2009 | USD | Cost analysis (case-control study) | US | High | 113 | 35 | Mean, 53 | NR |
| Myasthenia gravis | Harris et al., 2020 (28) | 2013–2019 | NR | Healthcare resource utilisation study (retrospective, non-comparative) | US | High | 782 | Ever-refractory gMG:  27  Non-refractory gMG:  43 | Every-refractory gMG:  Mean, 51.6 (SD, 14.3)  Non-refractory gMG:  Mean, 59.2 (SD, 13.9) | 6-month probability of an exacerbation decreased over time, from ~60% at baseline to ~20–40% at 4 years |
| Myasthenia gravis | Ignatova et al., 2022 (29) | 2020 | 2020, EUR | Cost-of-illness study (patient survey, non-comparative) | Bulgaria | High | 54 | 19 | Mean, 45 (SD, 13) | NR |
| Myasthenia gravis | Lin et al., 2020 (30) | 2013–2015 | [Costs expressed as percentages of total expenses; actual values not provided] | Cost analysis (retrospective, non-comparative) | China | Upper middle | 3,341 | 41.6 | 50–59, 22.7%  60–69, 20.6%  70–79, 16.9% | NR |
| Myasthenia gravis | Mandawat et al., 2010 (31) | 2000–2005 | Year not reported, USD | Economic outcomes study (retrospective, comparison of patients with myasthenia gravis with vs without crisis) | US | High | 1,606 (908 patients with myasthenia gravis and 698 patients with myasthenia gravis crisis) | Myasthenia Gravis:  PLEX, 34.3  IVIG, 37.4  Myasthenia Gravis crisis:  PLEX, 45.4  IVIG, 29.6 | Myasthenia Gravis:  PLEX, mean, 53.2 (SD, 18.4)  IVIg, mean, 50.7 (SD, 23.7)  Myasthenia Gravis crisis:  PLEX, mean, 58.9 (SD, 18.5)  IVIg, mean, 56.3 (SD, 22.0) | NR |
| Myasthenia gravis | Qi et al., 2022 (32) | 2014–2019 | 2018, USD | IVIg utilisation study (retrospective, comparison of chronic versus intermittent users of IVIg) | US | High | 1,225 | 43.1 | Mean, 58.9 (SD, 14.8) | NR |
| Myasthenia gravis | Schepelmann et al., 2010 (33) | 2005 | 2009, EUR | Patient questionnaires and diaries (non-comparative) | Germany | High | myasthenia gravis (*n* = 41) | 43.9 | 51–70, 29.3%  >70, 36.6% | NR |
| Myasthenia gravis | Sonkar et al., 2017 (34) | 2014–2016 | 2016, INR and USD | Cost analysis (prospective, non-comparative) | India | Lower middle | 66 | 59.0 | 42 (range, 6–75) | NR |
| Myasthenia gravis | Ting et al., 2023 (35) | 2008–2019 | 2019, USD | Analysis of healthcare resource utilisation and costs (retrospective study of insurance claims data, non-comparative) | US | High | 1,498 | 46.9 | 59.0 (IQR, 48.0–71.0) | During the 1-year baseline period, there was a mean of 0.95 exacerbations per patient (SD 3.18). Over the 2-year follow-up period, 49% of the total cohort had ≥1 myasthenia gravis exacerbation and, among these patients, the mean number of exacerbations per patient was 5.10 (10.55). The mean number of claims per patient among the total population was 2.49 (7.80) |
| Neuroimmunological conditions (paediatric) | Nosadini et al., 2016 (36) | 2000–2014 | 2015, USD | Retrospective study (chart review; includes comparison of immunoglobulin costs for patients with different neuroimmunological conditions) | Australia | High | 196 | 49.0 | 5.1 (range, 0.3–15.8) | NR |
| NMOSD | Beekman et al., 2019 (37) | Not reported | Year NR, USD | Cost analysis (performed within a study of patient experience and quality of life, non-comparative) | US | High | 193 | 11.4 | Mean, 49.2 ± 12.8 (range, 19–76) | NR |
| NMOSD | Exuzides et al., 2021 (38) | 2014–2018 | 2019, USD | Cost analysis (case-control study) | US | High | 162 | 26.5 | 45 (Q1–Q3, 33–58) | NR |
| NMO | Holroyd et al., 2019 (39) | 2018 | 2011, USD | Analysis of availability and affordability of neuromyelitis optica testing and treatment (physician survey, non-comparative) | Global | High, upper middle, lower middle and low | 60 physicians | N/A | N/A | NR |
| NMOSD | Hughes et al., 2022 (40) | 2016–2018 | 2016–2017, GBP | Analysis of health utilities and costs (patient survey, non-comparative) | UK | High | 117 patients and 74 informal carers | 22 | Mean, 53 (SD, 15) | Mean of 3 relapses (range 0–10) after their first attack since diagnosis; mean duration since first attack of 12 years (SD 8 years) |
| NMOSD | Knapp et al., 2022 (41) | 2013–2019 | Year NR, EUR | Cost analysis (based on health insurance data, comparison of patients with active disease vs inactive disease vs controls) | Germany | High | 130 | 42 | 46.5 (range, 3–89) | NR |

\*Sex and age data include 13 patients that received IVIG treatment and were selected to switch to SCIG therapy. One patient returned to IVIG therapy after symptoms worsened. †Patients with MOGAD diagnosed according to Jarius et al. 2018 (42).  
The study by Mahdi-Rogers et al. (11) included three different diseases (CIDP, multifocal motor neuropathy and paraproteinaemic demyelinating neuropathy), but here we have extracted data for chronic inflammatory demyelinating polyneuropathy only. The study by Schepelmann et al. (33) included three different diseases (amyotrophic lateral sclerosis, facioscapulohumeral muscular dystrophy and myasthenia gravis), but here we have extracted data for myasthenia gravis only. The publication by Allen et al. (6) is a review, retained because cost data therein appear to be original (study dates have been taken from the PATH and ICE studies, and the number of patients is the sum of participants from these two studies). CHF, Swiss francs; CIDP, chronic inflammatory demyelinating polyneuropathy; CNY, Chinese yuan; EUR, Euros; GBP, British pounds; HRQoL, health-related quality of life; INR, Indian rupees; IV, intravenous; IVIg, intravenous immunoglobulin; MOGAD, myelin oligodendrocyte glycoprotein antibody-associated disease; NMO, neuromyelitis optica; NMOSD, neuromyelitis optica spectrum disorder; NR, not reported; NTD, new Taiwan dollars; PLEX, plasma exchange; RMB, renminbi; SCIg, subcutaneous immunoglobulin; SD, standard deviation; USD, US dollars.

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