Effect of the COVID-19 pandemic on outpatient care and rehabilitation in neuromuscular clinical practice in Japan: a health insurance claims database analysis

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ABSTRACT

Objectives: To evaluate the impact of the COVID-19 pandemic on outpatient care in Japanese patients with neuromuscular diseases (NMDs).

Design: This retrospective cohort study included patients between January 2018 and February 2019; the follow-up period was divided into ‘before COVID-19’ (March 2019–February 2020) and ‘during COVID-19’ (March 2020–February 2021).

Setting: JMDC claims database study.

Participants: Of the 10,655,557 patients identified, we included patients with spinal muscular atrophy (SMA; n=82), neuromyelitis optica (NMO; n=342), myasthenia gravis (MG; n=1,347), Guillain-Barré syndrome (GBS; n=442) or autoimmune encephalitis/encephalopathy (AIE; n=133). Patients were required to have ≥1 month of data available, have a diagnosis of NMD during the enrolment period and be available for follow-up.

Primary and secondary outcome measures: We estimated the proportion of patients with >30% change in outpatient consultation and rehabilitation visits before versus during the COVID-19 pandemic.

Results: Small reductions in the proportion of patients with outpatient consultation/rehabilitation visits were observed before versus during the pandemic. Compared with before the pandemic, 30.4%, 27.8%, 28.7%, 49.4% and 50.0% of patients showed a >30% decrease in outpatient consultation visits and 58.6%, 75.0%, 50.0%, 76.3% and 84.6% showed a >30% decrease in outpatient rehabilitation visits during the pandemic for SMA, NMO, MG, GBS and AIE, respectively. The median change in the number of outpatient consultation visits per year before versus during pandemic was −1.0 day for all NMDs, and that in outpatient rehabilitation visits per year was −6.0, −5.5, −1.5, −6.5 and −9.0 days for SMA, NMO, MG, GBS and AIE, respectively. The reduction in outpatient rehabilitation visits was greater in the absence versus presence of a neurology specialist.

Conclusions: Outpatient consultation and rehabilitation visits during the COVID-19 pandemic were affected in Japanese patients with NMDs. Longer-term evaluations are required to understand if these reductions in outpatient care would affect patient prognosis.

INTRODUCTION

Neuromuscular diseases (NMDs) are a heterogeneous group of disorders that mostly affect the central and peripheral nervous systems, followed by the neuromuscular junctions and muscles.1 2 Examples of well-studied NMDs include spinal muscular atrophy (SMA), myasthenia gravis (MG) and Guillain-Barré syndrome (GBS).1 2 Patients with NMDs experience muscular impairment, together with subsequent loss of ambulation and respiratory muscle weakness.3 In patients with NMD, respiratory failure has been the most common cause of morbidity and mortality.3

NMDs are chronic and require long-term medical care; therefore, physiotherapy and rehabilitation programmes are critical for preventing respiratory problems and maintaining muscle strength, joint flexibility, functional capacity and quality of life in patients with NMDs.4–6

The COVID-19 pandemic has majorly impacted healthcare services worldwide. Particularly, non-essential and treatment services for non-communicable diseases have been widely affected, with 63% of countries having disrupted rehabilitation services. Moreover, in 94% of countries, healthcare...
workers have been reassigned to COVID-19 duties, which has resulted in a decrease in resources for treating or managing other diseases. The safety measures associated with the pandemic, such as imposing lockdowns and stay-at-home orders, have restricted access to healthcare centres and led to the restructuring of care strategies for various diseases. Studies from the USA and Europe have reported major reductions in the utilisation of healthcare services during the COVID-19 pandemic. A systematic review of the extent and nature of changes in the utilisation of healthcare services during the COVID-19 pandemic, covering the USA, Canada, Australia and several countries from Europe, Asia and South America, reported a median 37% reduction in healthcare services overall— a median 42% reduction in hospital visits, a median 28% reduction in admissions, a median 31% reduction in diagnostics and a median 30% reduction in therapeutic administration.

Clinical guidelines for the care of patients with NMDs during the COVID-19 pandemic have been published by European research groups. Briefly, these guidelines include the prominent use of telemedicine approaches and outpatient clinics as valuable alternatives to hospital visits. Although studies have assessed the use of tele-neurology—including telerehabilitation—for patients with chronic neurological conditions, literature on patients with NMDs is scarce. In March 2020, the Japanese Society of Neurology stated that, as the number of patients with COVID-19 increases, more efforts to reduce the medical needs of patients with neurological disorders will be necessary. Additional guidance for respiratory care of patients with both NMDs and COVID-19 have been published.

Limited access to healthcare services has raised concerns regarding the effect of the COVID-19 pandemic on outpatient care and rehabilitation for patients with NMDs, who often require long-term treatment. A definitive reduction in healthcare services in several countries was observed during the first period of the pandemic. In this study, we performed a health insurance claims database analysis to evaluate the impact of the COVID-19 pandemic on outpatient care and rehabilitation for patients with NMDs in Japan. Particularly, we focused on changes in outpatient care between the pre-pandemic and pandemic periods for patients requiring long-term treatment. The evaluation period was categorised into before March 2020 and after March 2020 when the impact of the COVID-19 pandemic became apparent and the government task force was established in Japan.

METHODS
Study design and data source
This was a retrospective cohort study conducted using the JMDC (formerly Japan Medical Data Center) health insurance claims database. The JMDC database includes pooled claims of healthcare data (medical care and drug prescriptions) from several health insurance associations, health examination data and health insurance association membership data since 2005. The cumulative population size is approximately 14 million individuals (as of February 2022) and the data are available as anonymously processed information. In this study, JMDC patient data were accessed between January 2018 and February 2021 (total data period). The patient inclusion period was between January 2018 and February 2019, and the follow-up period was between March 2019 and February 2021. The follow-up period was further divided into ‘before the COVID-19 pandemic’ (March 2019–February 2020) and ‘during the COVID-19 pandemic’ (March 2020–February 2021) to confirm the impact of the COVID-19 pandemic.

Patient and public involvement
Patients or the public were not involved in the design, conduct, reporting or dissemination plans of our research.

Patient inclusion criteria
Considering the diversity of NMDs, the data of patients with diseases involving subacute, acute, slowly progressive and repeated remission/exacerbation manifestations, as well as those that tended to become chronic, were included. Additionally, because the JMDC database comprises the data of individuals aged <75 years, only diseases with a relatively low age of incidence were considered. Therefore, the five NMDs of interest were SMA, neuromyelitis optica (NMO), MG, GBS and autoimmune encephalitis/encephalopathy (AIE). Patients were included if they had a diagnosis of one or more of the five diseases of interest during the inclusion period, available data and a membership with health insurance associations within the total data period for ≥1 month and continuous data during the 2-year follow-up period. Enrolment in the study was confirmed during the follow-up period. There were no exclusion criteria.

Outcomes
All outcomes were calculated for the 1-year period before the COVID-19 pandemic (March 2019–February 2020) and the 1-year period during the COVID-19 pandemic (March 2020–February 2021). Outcomes included the proportion of patients who attended outpatient consultations and outpatient rehabilitation visits among the study participants; the number of outpatient consultation visits and outpatient rehabilitation visits; the proportion of patients with a clinically meaningful >30% decrease, >30% increase and between 30% decrease and 30% increase (‘no change’) in outpatient consultations and outpatient rehabilitation visits during the COVID-19 pandemic versus before the COVID-19 pandemic among those with ≥1 outpatient visits during the 2-year follow-up period and median changes in outpatient consultations and outpatient rehabilitation visits during the COVID-19 pandemic versus before the COVID-19 pandemic.
Statistical analysis

This study was an exploratory analysis, and we did not assume null or alternative hypothesis formally; therefore, statistical hypothesis testing was not performed, and only descriptive statistics are calculated/presented retrospectively. Stratification factors for the outcomes included secondary healthcare areas (metropolitan, rural-urban and depopulated) and the presence or absence of the Japanese Society of Neurology-certified specialists during outpatient consultation and rehabilitation visits. Metropolitan areas were defined as secondary healthcare areas with a population of at least 1 million or a population density of at least 2000/km². Rural-urban areas were defined as secondary healthcare areas with a population of at least 200000 or at least 100000 and a population density of at least 200/km², other than the conditions of metropolitan areas. Depopulated areas were defined as secondary healthcare areas other than the conditions of metropolitan and rural-urban areas. SAS V.9.4 was used for analyses of data and tabulation of results.

RESULTS

Analysis population

A total of 10 655 557 patients with ≥1 month of data available for the total data period (January 2018–February 2021) were identified in the JMEDC database, 3160 of whom had a diagnosis of an NMD of interest. Of these, 2284 patients had confirmation of enrolment during the follow-up period and were included in the study (online supplemental figure 1). Considering the number of patients by NMD, 82 had SMA, 342 had NMO, 1347 had SLE, 442 had GBS and 133 had AIE. Across the five NMDs assessed, 61 patients had a diagnosis of more than one NMD.

Patient demographics and characteristics

Patient characteristics at the start of the follow-up period are shown in table 1. The proportion of females across all five NMDs ranged from 38.5% to 71.6%, and the mean age across all five NMDs ranged from 30.3 years to 47.5 years. The proportion of patients with personal and family insurance ranged from 34.1% to 64.5% and 35.5% to 65.9%, respectively (table 1).

Outpatient consultation and rehabilitation visits before versus during the COVID-19 pandemic

Regarding neurological clinical practice, no major changes were noted in the treatment content of NMDs during versus before the COVID-19 pandemic. In general, cerebrovascular rehabilitation decreased for all NMDs during the pandemic. In patients with GBS and AIE, exercise rehabilitation also tended to decrease (online supplemental table 1). Compared with the 1-year period before the COVID-19 pandemic, small reductions in both outpatient consultation and outpatient rehabilitation visits were observed during the 1-year pandemic period across all five NMDs (online supplemental table 1). Across all five NMDs, small reductions in the proportion of patients with outpatient consultations and outpatient rehabilitation visits were observed before versus during the COVID-19 pandemic (figure 1; online supplemental figure 2). Among patients with ≥1 outpatient visit during the 2-year follow-up, 30.4%, 27.8%, 28.7%, 49.4% and 50.0% had a >30% decrease in the number of outpatient consultation visits (figure 2A), whereas 58.6%, 75.0%, 50.0%, 76.3% and 84.6% had a >30% decrease in the number of outpatient rehabilitation visits (figure 2B) for SMA, NMO, MG, GBS and AIE, respectively. The median change in the number of outpatient consultation visits per year was −1.0 day for all NMDs; however, the median change in the number of outpatient rehabilitation visits per year was −6.0, −5.5, −1.5, −6.5 and −9.0 days for SMA, NMO, MG, GBS and AIE, respectively (online supplemental figure 3).

Outpatient consultation and rehabilitation visits before versus during the COVID-19 pandemic according to the presence versus absence of neurology specialists

Compared with the 1-year period before the COVID-19 pandemic, for outpatient consultation visits, 36.5%, 27.6%, 29.6%, 49.7% and 51.9% of patients with SMA, NMO, MG, GBS and AIE, respectively, showed a >30% reduction in the number of visits during the pandemic in the presence of Japanese Society of Neurology-certified specialists, whereas 28.9%, 40.4%, 38.2%, 57.5% and 50.0% of patients with SMA, NMO, MG, GBS and AIE, respectively, showed a >30% reduction in the absence of Japanese Society of Neurology-certified specialists.

Table 1 Patient characteristics at the start of the follow-up period

<table>
<thead>
<tr>
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<th>Spinal muscular atrophy (N=82)</th>
<th>Neuromyelitis optica (N=342)</th>
<th>Myasthenia gravis (N=1347)</th>
<th>Guillian-Barré syndrome (N=442)</th>
<th>Autoimmune encephalitis/encephalopathy (N=133)</th>
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<tr>
<td>Female, n (%)</td>
<td>39 (47.6)</td>
<td>245 (71.6)</td>
<td>731 (54.3)</td>
<td>170 (38.5)</td>
<td>71 (53.4)</td>
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<td>Age, years, mean (SD)</td>
<td>30.3 (19.9)</td>
<td>45.0 (12.4)</td>
<td>47.5 (14.3)</td>
<td>41.4 (16.0)</td>
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<td>Insurance category, n (%)</td>
<td>Person</td>
<td>28 (34.1)</td>
<td>167 (48.8)</td>
<td>816 (60.6)</td>
<td>285 (64.5)</td>
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<td></td>
<td>Family</td>
<td>54 (65.9)</td>
<td>175 (51.2)</td>
<td>531 (39.4)</td>
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the certified specialists (figure 3A,B; online supplemental figure 4A). For outpatient rehabilitation visits, 30.0%, 70.0%, 40.0%, 66.7% and 87.5% of patients with SMA, NMO, MG, GBS and AIE, respectively, showed a >30% reduction in the number of visits in the presence of the specialists, whereas 56.3%, 100.0%, 55.6%, 84.2% and 100.0% of patients with SMA, NMO, MG, GBS and AIE, respectively, showed a >30% reduction in the absence of the specialists. However, it must be acknowledged that the number of patients with NMO and AIE in this analysis was small (NMO: n=2, AIE: n=4; figure 3C,D; online supplemental figure 4B). The median change in the number of outpatient consultation days per year in the presence of neurology specialists was −1.0 day for all NMDs and that in the absence of specialists ranged from −1.0 day to −1.5 days. Additionally, the median change in the number of outpatient rehabilitation visits per year in the presence of specialists ranged from −2.0 (MG) days to −6.0 (AIE) days and that in the absence of specialists ranged from −1.0 day (MG) to −12.0 days (AIE). Specifically, the median decrease in outpatient rehabilitation visits in the absence of specialists was approximately twice that in the presence of specialists for GBS (−10.0 days versus −5.5 days) and AIE (−12.0 days versus −6.0 days; online supplemental figure 5).

Figure 1 Proportion of patients who had (A) outpatient consultation visits and (B) outpatient rehabilitation visits before and during the COVID-19 pandemic. AIE, autoimmune encephalitis/encephalopathy; GBS, Guillain-Barré syndrome; MG, myasthenia gravis; NMO, neuromyelitis optica; SMA, spinal muscular atrophy.

Figure 2 Proportion of patients with a decrease and an increase in (A) outpatient consultation visits and (B) rehabilitation visits before versus during the COVID-19 pandemic. ‘No change’ indicates between 30% decrease and 30% increase. AIE, autoimmune encephalitis/encephalopathy; GBS, Guillain-Barré syndrome; MG, myasthenia gravis; NMO, neuromyelitis optica; SMA, spinal muscular atrophy.

Figure 3 Proportion of patients with a decrease and an increase in (A, B) outpatient consultation visits and (C, D) outpatient rehabilitation visits before versus during the COVID-19 pandemic in the presence/absence of neurology specialists. *Japanese Society of Neurology-certified specialist. ‘No change’ indicates between 30% decrease and 30% increase. AIE, autoimmune encephalitis/encephalopathy; GBS, Guillain-Barré syndrome; MG, myasthenia gravis; NMO, neuromyelitis optica; SMA, spinal muscular atrophy.

respectively, showed a >30% reduction in the absence of the specialists. However, it must be acknowledged that the number of patients with NMO and AIE in this analysis was small (NMO: n=2, AIE: n=4; figure 3C,D; online supplemental figure 4B). The median change in the number of outpatient consultation days per year in the presence of neurology specialists was −1.0 day for all NMDs and that in the absence of specialists ranged from −1.0 day to −1.5 days. Additionally, the median change in the number of outpatient rehabilitation visits per year in the presence of specialists ranged from −2.0 (MG) days to −6.0 (AIE) days and that in the absence of specialists ranged from −1.0 day (MG) to −12.0 days (AIE). Specifically, the median decrease in outpatient rehabilitation visits in the absence of specialists was approximately twice that in the presence of specialists for GBS (−10.0 days versus −5.5 days) and AIE (−12.0 days versus −6.0 days; online supplemental figure 5).
of outpatient consultation days per year in metropolitan areas was −1.0 day for all NMDs, and the change ranged from −1.0 day to −2.0 days in rural-urban and 0.5 day to −3.5 days in depopulated areas; the median change in the number of outpatient rehabilitation visits ranged from −1.0 day (MG) to −11.5 days (AIE) in metropolitan areas and from −2.0 (MG) days to −7.0 (SMA) days in rural-urban areas and was not calculable in depopulated areas (online supplemental figure 7).

**DISCUSSION**

In this healthcare insurance claims database analysis, we evaluated the impact of the COVID-19 pandemic on outpatient consultations and rehabilitation visits for long-term treatment of NMDs in Japan. Across all NMDs assessed, compared with the period before the pandemic, small decreases in outpatient consultation visits and outpatient rehabilitation visits were observed during the 1-year pandemic period. Among patients with ≥1 outpatient visit during the 2-year follow-up period, 30.4%, 27.8%, 28.7%, 49.4% and 50.0% had a >30% decrease in outpatient consultation visits, whereas 58.6%, 75.0%, 50.0%, 76.3% and 84.6% had a >30% decrease in outpatient rehabilitation visits for SMA, NMO, MG, GBS and AIE, respectively, during the COVID-19 pandemic. Accordingly, median reductions in the number of outpatient consultation visits per year during the pandemic were lower (−1 day) than those for outpatient rehabilitation visits (−1.5 days to −9.0 days). The decrease in the number of outpatient consultation visits was comparable between the presence and absence of specialists. A greater reduction in the number of outpatient rehabilitation visits was observed in the absence of a specialist when compared with that in the presence of a specialist. The outcomes categorised by secondary healthcare areas were comparable, indicating no major differences in the quality of treatment across secondary healthcare areas (metropolitan, rural-urban and depopulated). However, the number of patients in these analyses was small, and the results should be interpreted with caution.

The COVID-19 pandemic has substantially impacted the healthcare systems of several countries worldwide. Specifically, owing to the pandemic, the USA has observed an increased loss of insurance coverage among individuals, financial loss experienced by healthcare providers, racial and ethnic disparities in treatments and general inadequacy of the public health system. Compared with the other nations, the impact of COVID-19 on the healthcare system in Japan remains relatively limited. Nevertheless, the proportion of patients demonstrating a decrease in outpatient consultation visits was smaller than that for outpatient rehabilitation visits, suggesting that the pandemic had a greater impact on outpatient rehabilitation than on outpatient consultation. Patients who undergo rehabilitation basically have decreased motor function regardless of the disease, but there is a possibility that they refrained from outpatient rehabilitation due to...
a variety of factors (circumstances at the medical institutions and rehabilitation facilities or the patient’s request (such as due to fear of infection)).

The marginal decrease in outpatient consultation and rehabilitation visits observed in this study is broadly consistent with the results presented by Sato et al., who used data from the DeSC administrative claims database to measure changes in the frequency of outpatient visits for long-term ambulatory care of epilepsy, migraine, Parkinson’s disease (PD) and Alzheimer’s disease (AD) in Japan. Specifically, monthly outpatient visits were found to have reduced marginally for epilepsy, PD and AD (relative risk (RR): approximately 0.90), whereas those associated with migraine had increased (RR=1.15). The authors concluded that the impact of the COVID-19 pandemic on ambulatory care for epilepsy, PD and AD was relatively limited in Japan. However, a second Japanese study by Matsumura et al. suggested that, overall, COVID-19 had a serious impact on the activities and quality of life of patients with muscular dystrophy. In this study, a web-based survey was conducted to assess the impact of the pandemic on the care for patients with COVID-19 and muscular dystrophy. The results demonstrated that approximately 30% of the patients surveyed deferred consultations and 25% of patients receiving ≥1 consultation per month engaged in telephone consultations. Additionally, a reduced frequency of rehabilitation and outings had resulted in a decrease in exercise levels and an increase in caregiver burden.

Similar trends in the usage of healthcare services have been observed in Europe. An electronic survey conducted by Mauri et al. evaluated the impact of the COVID-19 pandemic on healthcare services provided by neuromuscular centres affiliated with the Italian Association of Myology. Forty percent of the centres surveyed reported a decrease in outpatient visits, and 22% of the centres deferred in-hospital therapies for NMDs. Moreover, rehabilitation services and outpatient visits were the most affected, having been suspended in 95% of the centres. In another study by Gagliardi et al. evaluating the effects of the COVID-19 pandemic on the quality of life of 205 patients with NMD in the Lombardy area of Italy, the authors reported that outpatient visits were postponed in 57.1% of cases and that 50.7% of patients experienced a cancellation of scheduled diagnostic tests, suggesting that the pandemic substantially disrupted clinical and support services for patients with NMDs and reduced quality of life. Taken together, the results from these two European studies suggest that the impact of the COVID-19 pandemic on patients with NMDs was higher in Europe than in Japan; however, the reasons for the higher impact are currently unclear and must be investigated in future studies. One of the factors may be that Japan did not implement a mandatory lockdown similar to that in Europe. In China, the degree of strict lockdown has been reported to have had an impact on healthcare reductions. Our study did not show any major changes in the treatment approach of NMDs during versus before the COVID-19 pandemic. Furthermore, online medical practice did not increase during this study. This suggested that patients requiring continuous medication or treatment did not interrupt outpatient consultations. In particular, patients with chronic NMDs may have preferred face-to-face treatment with specialists to control their symptoms, even during the pandemic. In addition, immediately after the pandemic, we did not have an environment where we could provide online medical care. On the other hand, the American Academy of Neurology recommends postponing hospital visits and introducing telemedicine. Maccarone and Masiero also describe a reorganisation of rehabilitation during the pandemic. In future, various forms of treatment may be considered while reducing patient risk.

The COVID-19 pandemic has affected not only patients with NMDs but also those with other chronic diseases in Japan. A retrospective cohort study of nationwide claims data (2018–2020) reported that the number of physician visits declined at the onset of the COVID-19 outbreak but returned to baseline 1 month later. Additionally, there was no evidence that patients were skipping medications for major chronic diseases (hypertension, diabetes and dyslipidaemia) during the pandemic. Another retrospective cohort study using a hospital-based claims database reported that the number of hospitalisations for respiratory diseases decreased by 55% and that for asthma decreased by 80%, while the number of outpatient rehabilitations decreased by 39% during the first wave of the COVID-19 pandemic in May 2020. By contrast, outpatient chemotherapy and maintenance hemodialysis treatments decreased by <10%. Moreover, a study conducted using the social insurance medical fee data reported that the decline in the number of physician visits was greater following the first state of emergency declaration than following the spread of COVID-19 in the same period. However, this decline was reduced by nearly 50% following the second state of emergency declaration. These results indicate that the impact of COVID-19 on healthcare resource utilisation in Japan was not limited to NMDs alone but extended to other illnesses that were not associated with COVID-19.

Our study had several limitations. First, the JMDC database is not suitable for obtaining data from elderly patients aged ≥75 years. However, most of the NMDs included in the current study occur at a young or middle age, and therefore, the representativeness of patients in the JMDC database is expected to be high. Second, the study population had a slightly increased prevalence of AIE when compared with that commonly reported in the Japanese population, and the proportion of men with NMO was relatively high. Therefore, the results pertaining to these groups must be interpreted with caution. Third, the factors that contribute to the results presented here may be associated with the natural history of primary NMD in addition to the effects of the COVID-19 pandemic; however, these factors could not be identified in the current study. Lastly, the long-term impact of the
COVID-19 pandemic on patients with NMDs could not be evaluated in this study.

CONCLUSIONS
The results of this study indicate that the COVID-19 pandemic had an impact on outpatient consultation and rehabilitation visits for patients with NMDs in Japan. However, further studies are warranted to evaluate the long-term impact of the pandemic on patients with NMDs.

REFERENCES


