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## Original Article

# Hospital-based care utilization of children with medical complexity in Japan

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**Abstract** **Background:** Few studies have investigated the hospital-based care utilization of children with medical complexity (CMC) in Japan. This study examined the frequency and differences in hospital-based care utilization for CMC according to the level of medical complexity (moderate and severe).

**Methods:** Medical records of three pediatric tertiary hospitals in one prefecture were examined in 2014. We examined the number of outpatient visits and of admissions to the hospital for CMC in the 5 years after the introduction of home medical care.

**Results:** Of 92 CMC, 55 had medical complexity that was moderate (CMC-moderate) and 37 had medical complexity that was severe (CMC-severe). The number of CMC who had medical care introduced at home had increased year by year, especially that of CMC <2 years old; the number of older CMC (i.e. 7–17 years old) had also increased in 2010–2014. The median total outpatient visits was 20 (IQR, 13–29 visits) for CMC-moderate and 20 (IQR, 17–26 visits) for CMC-severe in the first year. CMC-severe had significantly longer length of admissions in the 5 years than CMC-moderate. The number of total visits and admissions during the subsequent 4 years (from the second to the fifth year) was slightly decreased compared with the first year, but this was not significantly different.

**Conclusions:** CMC had high utilization of hospital-based care, and consistently utilized hospital-based care in the 5 years after the introduction of home medical care. Further study is needed to examine both hospital-based and home/community-based services use.

**Key words** children with medical complexity, hospital-based care utilization, Japan, severe motor and intellectual disability.

Severe motor and intellectual disabilities (SMID) has been used to define children with severe or multiple disabilities based on two dimensions of physical and intellectual abilities since the 1960s in Japan.<sup>1</sup> This term is similar to profound intellectual and multiple disabilities (PIMD) established by the International Association for the Scientific Study of Intellectual Disability.<sup>2</sup> According to recent developments in neonatal and pediatric intensive care, the increased number of children living with medical care has attracted public attention. In 1991, the term “severe motor and intellectual disabilities–medical care dependent group” (SMID-MCDG) was introduced to describe severity of disability in terms of caregiving situations due to medically complex care.<sup>3</sup> In 2008, the Japan Pediatric Society (JPS) first reported the proportion of children aged 0–19 years with SMID-MCDG as approximately 0.30/1,000 population.<sup>4</sup> The JPS estimated that around 7,350 children with

SMID-MCDG lived in Japan, and approximately 70% of them stayed at home. The caregiving burden for these children mainly falls on family members: for example, only 18% of children with SMID-MCDG received home-visiting nurse services, and 88% of them regularly visited hospitals to receive medical care by physicians.<sup>4</sup>

Understanding health-care utilization is important to reduce the care-related burden on families and maintain the lives of children with high medical care need in the community. Cohen *et al.* proposed a definitional framework of children with medical complexity (CMC) to characterize children with coexisting multiple service needs, chronic conditions, functional limitations, and frequent health-care use.<sup>5</sup> With regard to CMC health-care utilization in other countries, the rate of admissions for CMC increased significantly from 1995 to 2010 in US hospitals.<sup>6</sup> Health-care expenditure for CMC accounted for almost one-third of all children, although the CMC comprised <1% of the child population in Ontario, Canada.<sup>7</sup>

Children with high medical needs, referred to as SMID-MCDG in Japan, share similarities with CMC with regard to

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the high need for multiple services and frequent utilization of health care. Few studies, however, have examined health-care utilization in children with high medical needs in Japan. Therefore, the aim of this study was to examine the characteristics of children with high medical care needs and hospital-based care utilization in one prefecture in Japan, especially with regard to the number of outpatient visits and admissions to hospital in the 5 years after the introduction of medical care at home.

## Methods

### Setting

This was a descriptive study with medical record review in three hospitals in one prefecture in Japan, located approximately 1–2 h by train from Tokyo. This prefecture has a population of 2.97 million and occupies an area of 6,096 km<sup>2</sup>. All three hospitals participating in this study were center hospitals of emergency pediatric medicine in the prefecture. These hospitals are also centers for maternal, fetal, and neonatal health. We selected the subjects from these three hospitals based on the SMID-MCDG scoring system, because children with medical high needs tend to have diseases or disabilities related to neonatal and emergency medicine.

### Defining CMC

The scoring system for children with SMID-MCDG was used to select the subjects (Table 1).<sup>3</sup> This scoring system has been used in health insurance claims since 1996 to calculate additional fees during hospitalization associated with increased complex care needs. It has been widely and practically used to evaluate the severity of medical care in children. Suzuki *et al.* defined a score 10–24 points as semi SMID-MCDG, and  $\geq 25$  points as genuine SMID-MCDG.<sup>3</sup> The current study focused on CMC as defined by Cohen *et al.*<sup>5</sup> to examine the utilization of hospital-based care. Therefore, in the present study we focused on children who scored  $\geq 10$  points, and classified children who scored 10–24 points as having medical complexity that was moderate (CMC-moderate), and children who scored  $\geq 25$  points as having medical complexity that was severe (CMC-severe).

### Subject selection

Selection of CMC-moderate or -severe according to the SMID-MCDG definition (i.e. those whose physical function at best was achievement of the sitting position, and who scored  $\geq 10$  points on the SMID-MCDG scoring system) was carried out in two steps. First, we selected children visiting the outpatient departments of pediatrics and pediatric surgery in three hospitals who incurred medical fees related to the scoring system items from health insurance claims in 2014. Patients usually pay these home medical fees at outpatient regular visits and receive medical equipment. According to health insurance

**Table 1** SMID-MCDG scoring system for CMC

	Physical function at best: Sitting position	Score
1	Ventilator <sup>†</sup>	10
2	Endotracheal intubation or tracheotomy	8
3	Nasal airway	5
4	Oxygen inhalation or SpO <sub>2</sub> < 90%	5
5	Frequent suction of airway secretions: more than once per hour	8
	More than six times per day	3
6	Frequent use of nebulizer: more than six times per day or continuously	3
7	Total parenteral nutrition	10
8	Oral intake with total assistance by caregiver <sup>‡</sup> , or Nasogastric feeding tube or gastrostomy tube feeding <sup>‡</sup>	3
		5
9	Duodenal gastric feeding tube or jejunostomy tube feeding <sup>‡</sup> , or Constant infusion pump	8
		3
10	Hyperhidrosis due to hypertonicity, which is uncontrolled by operation or medication, requires frequent changing of clothes or position more than three times per day	3
11	Continuous dialysis treatment	10
12	Regular urethral catheterization: more than three times per day	5
13	Colostomy	5
14	Frequent of position changes: More than 6 times per day	3

<sup>†</sup>Ventilation includes mechanical pressure ventilation required every day, such as nasal intermittent positive pressure ventilation, continuous positive airway pressure machine, and mechanical insufflation–exsufflation device. <sup>‡</sup>To determine feeding score, one item is selected from items 8–9 (8, oral intake, nasogastric tube, gastrostomy tube; 9, duodenal gastric feeding tube, constant infusion pump). CMC, children with medical complexity; SMID-MCDG, severe motor and intellectual disabilities–medical care dependent group.

claims, we selected children who scored  $\geq 5$  points for any kind of medical fees. The reason for setting the cut-off at  $\geq 5$  was because the medical fee did not cover several items of the scoring system such as frequency of suction, nebulizer, changing position, or oral intake. Therefore, the home caregiving situation, such as frequency of suction, nebulizer, or changing position was confirmed via conversation with the family members of the children who scored  $\geq 5$  points on health insurance claims using any medical devices at home. After confirmation of the home caregiving situation, children who scored <10 points or who could walk or stand were excluded. Finally, 92 CMC were selected for the study.

### Hospital-based care utilization

We examined medical records for at most 5 years after the introduction of any medical devices at home. The timing of introduced medical care was defined as the time at which children started to use any kind of device, such as mechanical ventilator, tracheotomy, or feeding tubes, which required payment of medical fees related to the items of the SMID-MCDG scoring system. This study evaluated the number of regular

outpatient visits, emergency outpatient visits, planned admissions, and emergency admissions as hospital-based care utilization at the corresponding hospital for each child. Emergency hospital-based care utilization is a different burden for families than planned care utilization in advance. Therefore, this study examined planned versus unplanned utilization separately. Regular visits were defined as appointments in advance, and planned admissions included admissions for surgery or examinations scheduled in advance. Emergency visits and admissions were defined as unplanned visits or admission without appointment. Admissions to other hospitals were included in the number of admissions and were obtained from referral forms. The total number of outpatient visits and total number of admissions were defined as the sum of regular and emergency visits or planned and emergency admissions, respectively. We also investigated the child characteristics (age at introduction of medical care and sex), need for medical care at home, and fiscal year of medical care introduction.

We also examined the medical records of home/community-based care utilization. These services included physician home visit (i.e. regular physician visit to the child's home to provide medical care), home-visit nursing care, home help service, home bathing service, short stay services as a means of respite care, and day services. The identification of service utilization was based on medical record review.

### Statistical analysis

First, we compared the differences in child characteristics between the three hospitals. Second, we performed bivariate analysis to compare differences in child characteristics, need for medical care at home, and home/community-based care utilization between CMC-moderate and CMC-severe using chi-squared test, or Fisher's exact test. Third, we described hospital-based care utilization, and compared the total number of visits and admissions between CMC-moderate and CMC-severe on Wilcoxon rank-sum test. Because the follow-up duration differed between children, the number of visits and of admissions was counted by year. The length of hospitalization was compared between CMC-moderate and CMC-severe using Wilcoxon rank-sum test. Last, we compared the number of hospital visits and of admissions between the first year and the next 4 years (from the second to fifth year) for CMC followed for all 5 years, using Wilcoxon signed-rank test. The institutional review boards at all three hospitals approved this study.

### Results

Table 2 lists the characteristics for CMC-moderate and CMC-severe according to hospital. No significant differences were seen in sex, age or year of medical care introduction, or in prevalence of CMC -moderate and -severe according to hospital. The year of medical care introduction differed with participating hospital. The majority of CMC in hospitals A (86.4%) and C (88.0%) had medical care introduced at home since

2005, while 30% of CMC in hospital B had started medical care during 1994–2004. Hospital B also provided home medical care by physician ( $n = 6$ ) and planned admission as a means of respite care ( $n = 8$ ). We examined medical records from the beginning of medical care at home to at most 5 years. More than half of the subjects (62.0%) were followed up to the fifth year. No significant difference was seen in the proportion of CMC-severe with hospital.

Figure 1 shows the year of home medical care introduction according to CMC age. The majority of CMC had medical care introduced at home before the age of 2 years old, but the number of older CMC (i.e. 7–17 years old) with introduction of home medical care also increased in 2010–2014.

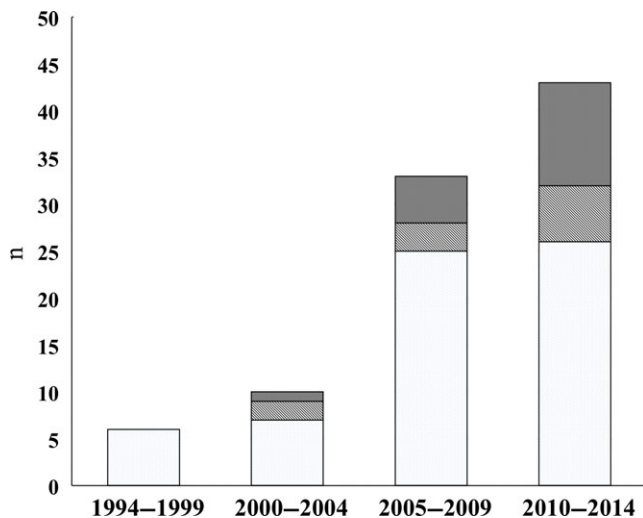
Table 3 lists the differences in CMC characteristics, year of home medical care introduction, use of home medical care from SMID-MCDG score, and home/community-based care utilization according to level of MC. With regard to CMC-severe, most had home medical care introduced before 2 years old (83.8%). In contrast, for CMC-moderate, one-fourth (25.5%) had home medical care introduced after 7 years old. The number of CMC with home medical care introduction increased gradually for both CMC-moderate and CMC-severe by year. The most frequent type of home medical care introduced was nasogastric or gastrostomy tube (moderate MC, 76.4%; severe MC, 81.1%). CMC-severe differed significantly from CMC-moderate with regard to use of ventilator, tracheotomy, use of constant infusion pump, and frequent change of position more than six times per day. The underlying medical conditions consisted of neurological disease (e.g. malformation of the brain, encephalopathy, cerebral palsy etc.,  $n = 40$ ), neonatal disease (e.g. neonatal asphyxia, hypoxic-ischemic encephalopathy etc.,  $n = 18$ ), muscle disorders (muscular dystrophy, spinal muscular atrophy etc.,  $n = 12$ ), chromosomal abnormality ( $n = 8$ ), multiple abnormalities ( $n = 8$ ), and other diseases ( $n = 2$ ). Four children became CMC due to external causes such as drowning ( $n = 3$ ) and suffocation ( $n = 1$ ). The most frequently used service at home or in the community was home-visit nursing care (53.3%). CMC-severe were significantly more likely to have physician home visit (18.9%) and home-visit nursing care (78.4%).

Table 4 lists the annual number of hospital visits and admissions according to MC status for up to 5 years. Hospital B provided home medical care by physician for six CMC-severe and periodically planned admissions as a means of respite care for eight CMC-severe. These CMC received regular home-visits by physicians, instead of outpatient care, and had regular hospital admission for respite care. These CMC were therefore excluded from this analysis because this type of hospital-based care utilization might differ from that of CMC without home-based physician services or respite care. The CMC-moderate and CMC-severe had an annual median of 20 outpatient visits in the first year (IQR: CMC-moderate, 13–29 visits; CMC-severe, 17–26 visits). Of the total 20 visits, the median of emergency visits was four for CMC-moderate and five for CMC-severe. During the successive 4 years after

**Table 2** CMC characteristics vs hospital

		Total ( <i>n</i> = 92)		Hospital A ( <i>n</i> = 44)		Hospital B ( <i>n</i> = 23)		Hospital C ( <i>n</i> = 25)	
		<i>n</i>	(%)	<i>n</i>	(%)	<i>n</i>	(%)	<i>n</i>	(%)
Sex	Male	40	(43.5)	23	(52.3)	6	(26.1)	11	(44.0)
	Female	52	(56.5)	21	(47.7)	17	(73.9)	14	(56.0)
Age at home medical care introduction (years)	0–1	64	(69.6)	34	(77.3)	17	(73.9)	13	(52.0)
	2–6	11	(12.0)	2	(4.5)	4	(17.4)	5	(20.0)
	7–17	17	(18.5)	8	(18.2)	2	(8.7)	7	(28.0)
Year of home medical care introduction	1994–1999	6	(6.5)	3	(6.8)	3	(13.0)	0	(0.0)
	2000–2004	10	(10.9)	3	(6.8)	4	(17.4)	3	(12.0)
	2005–2009	33	(35.9)	20	(45.5)	3	(13.0)	10	(40.0)
	2010–2014	43	(46.7)	18	(40.9)	13	(56.5)	12	(48.0)
Follow-up period (months)	<12	2	(2.2)	1	(2.3)	1	(4.4)	0	(0.0)
	12–23	16	(17.4)	8	(18.2)	3	(13.0)	5	(20.0)
	24–35	12	(13.0)	7	(15.9)	1	(4.4)	4	(16.0)
	36–47	5	(5.4)	0	(0.0)	4	(17.4)	1	(4.0)
	48–60	57	(62.0)	28	(63.6)	14	(60.9)	15	(60.0)
CMC score (points), Median, (IQR)		21	(16–29)	21	(16–28)	21	(18–28)	24	(13–29)
CMC-moderate	10–24	55	(59.8)	27	(61.4)	14	(60.9)	14	(56.0)
CMC-severe	≥25	37	(40.2)	17	(38.6)	9	(39.1)	11	(44.0)

CMC, children with medical complexity; CMC-moderate, children with medical complexity that is moderate; CMC-severe, children with medical complexity that is severe.



**Fig. 1** Introduction of medical care at home in children with medical complexity according to year in those aged (□) 0–1 year; (▒) 2–6 years; and (■) 7–17 years.

the first year of home-based medical care introduction, the median total visits was 15–17 per year for CMC-moderate and 14.5–16 per year for CMC-severe. No significant differences were seen in the 5 years between CMC-moderate and CMC-severe for total outpatient visits per year. Likewise, the number of emergency visits was not significantly different between CMC-moderate and CMC-severe ( $P = 0.776, 0.470, 0.810, 0.695,$  and  $0.516$ , in each year respectively; not shown in Table 4).

In terms of admissions, CMC-severe had three per year, and CMC-moderate had two per year in the first year. During

the next 4 years, the median total admissions was 0–1.5 per year for CMC-moderate, and 1–2 per year for CMC-severe. CMC-severe had significantly more admissions in the fifth year ( $P = 0.003$ ) compared with CMC-moderate. In addition to the number of admissions, the length of admission was also significantly longer for CMC-severe (median, 234 days; IQR, 88–372 days) than for CMC-moderate (median, 128.5; IQR, 61–200 days) in the CMC followed for 5 years ( $P = 0.027$ ).

Next, we compared the numbers of hospital visits and admissions between the first year and in the next 4 years for CMC who could be followed for the 5 years. Although the numbers of total visits and admissions were slightly decreased during the 5 years, there was no significant difference between the first year and the next 4 years in total visits (not shown in Table 4: CMC-moderate,  $P = 0.224$ ; CMC-severe,  $P = 0.375$ ) or in total admissions (CMC-moderate,  $P = 0.443$ ; CMC-severe,  $P = 0.082$ ).

## Discussion

This was the first study to examine hospital-based health-care utilization in children with high medical care needs using SMID-MCDG score. The notable findings in this study were the extremely high utilization of hospital care in CMC: a median of 20 total outpatient visits, which included 4–5 emergency outpatient visits in the first year. In the following 4 years, the number of total visits and admissions was slightly, but not significantly, decreased compared with the first year. CMC-severe had significantly more admissions in the fifth year, and the length of admission was significantly longer compared with CMC-moderate.

**Table 3** CMC characteristics and type of medical care vs level of MC

Characteristics	Total ( <i>n</i> = 92)		CMC-moderate ( <i>n</i> = 55)		CMC-severe ( <i>n</i> = 37)		<i>P</i> -value
	<i>n</i>	(%)	<i>n</i>	(%)	<i>n</i>	(%)	
Sex (male)	40	(43.5)	26	(47.3)	14	(37.8)	0.970 <sup>‡</sup>
Age at introduced medical care (years)							
0–1	63	(68.5)	33	(60.0)	31	(83.8)	<b>0.043</b> <sup>†</sup>
2–6	10	(10.9)	8	(14.5)	3	(8.1)	
7–17	17	(18.5)	14	(25.5)	3	(8.1)	
Year of medical care introduction							
1994–1999	6	(6.5)	5	(9.1)	1	(2.7)	0.320 <sup>†</sup>
2000–2004	9	(9.8)	7	(12.7)	3	(8.1)	
2005–2009	32	(34.8)	16	(29.1)	17	(45.9)	
2010–2014	43	(46.7)	27	(49.1)	16	(43.2)	
SMID-MCDG score							
1. Ventilator	41	(44.6)	12	(21.8)	29	(78.4)	<b>&lt;0.001</b> <sup>‡</sup>
2. Tracheotomy	49	(53.3)	20	(36.4)	29	(78.4)	<b>&lt;0.001</b> <sup>†</sup>
3. Nasal airway	9	(9.8)	7	(12.7)	2	(5.4)	0.305 <sup>†</sup>
4. Oxygen inhalation	32	(34.8)	15	(27.3)	17	(45.9)	0.065 <sup>‡</sup>
5. Suction (>1/h)	33	(35.9)	17	(30.9)	16	(43.2)	0.226 <sup>‡</sup>
Suction (>6/day)	23	(25.0)	10	(18.2)	13	(35.1)	0.066 <sup>‡</sup>
6. Nebulizer	4	(4.3)	2	(3.6)	2	(5.4)	1.000 <sup>†</sup>
7. TPN	2	(2.2)	0	(0.0)	2	(5.4)	0.159 <sup>†</sup>
8. Oral intake with assistance	4	(4.3)	4	(7.3)	0	(0.0)	0.146 <sup>†</sup>
Nasogastric tube or gastrostomy tube	72	(78.3)	42	(76.4)	30	(81.1)	0.591 <sup>‡</sup>
9. Duodenal gastric tube or jejunostomy tube	10	(10.9)	3	(5.5)	7	(18.9)	0.083 <sup>†</sup>
Constant infusion pump	14	(15.2)	4	(7.3)	10	(27.0)	<b>0.016</b> <sup>†</sup>
10. Changing clothes/position due to hypertonicity (>3/day)	11	(12.0)	6	(10.9)	5	(13.5)	0.751 <sup>†</sup>
11. Continuous dialysis	1	(1.1)	0	(0.0)	1	(2.7)	0.402 <sup>†</sup>
12. Regular urethral catheterization (>3/day)	5	(5.4)	3	(5.5)	2	(5.4)	1.000 <sup>†</sup>
13. Stoma	1	(1.1)	1	(1.8)	0	(0.0)	1.000 <sup>†</sup>
14. Changing position (>6/day)	42	(45.7)	18	(32.7)	24	(64.9)	<b>0.002</b> <sup>‡</sup>
Health care utilization							
Physician home visit	7	(7.6)	0	(0.0)	7	(18.9)	<b>0.001</b> <sup>†</sup>
Home-visit nursing care	49	(53.3)	20	(36.4)	29	(78.4)	<b>&lt;0.001</b> <sup>‡</sup>
Home help service	7	(7.6)	2	(3.6)	5	(13.5)	0.113 <sup>†</sup>
Home bathing service	3	(3.3)	1	(1.8)	2	(5.4)	0.562 <sup>†</sup>
Short stay service	12	(13.0)	4	(7.3)	8	(21.6)	0.060 <sup>†</sup>
Day service	4	(4.3)	2	(3.6)	2	(5.4)	1.000 <sup>†</sup>

Bold, *P* < 0.05. <sup>†</sup>Fisher's exact test; <sup>‡</sup> chi-squared test. CMC, children with medical complexity; MC, medical complexity; TPN, total parenteral nutrition.

A similar high outpatient use has been previously reported for children with high medical needs. Cohen *et al.* reported that CMC had a median of 13 physician outpatient visits, and six specialist outpatient visits in the 2 years after the index hospitalization.<sup>7</sup> O'Mahony *et al.* reported that the rate of pediatric admission after emergency department (ED) visits and of repeat ED visits increased with increasing level of MC.<sup>8</sup> For example, the average number of pediatric ED visits in 2 years according to condition was 1.3 for non-chronic conditions, 2.0 for episodic, 2.4 for lifelong chronic, 3.4 for progressive chronic, and 3.3 for malignancy. In the present study the total outpatient visits and emergency visits to each tertiary hospital were higher compared with these studies. This may be because of the higher level of complexity in both CMC-moderate and CMC-severe in the present study (i.e. 89.2% needed any kind of tube feeding and 44.6% depended

on mechanical ventilation at home). This may also be the reason for the lack of significant difference in total and emergency visits according to MC status in the present study, and for the fact that all of the present CMC depended on tertiary hospitals as a primary resource for regular and emergency medical care after the introduction of home medical care.

One advantage of this study is that it examined tertiary hospital care utilization for these children with high MC for up to 5 years. This follow-up length is longer than in previous studies (i.e. 2 years<sup>7,8</sup>). The number of total outpatient visits and emergency visits did not significantly decrease or change in the 5 years for CMC-moderate or CMC-severe. Comparing the first and the successive 4 years, the number of total visits and of admissions were slightly, but not significantly, decreased. This indicated that both CMC-moderate and CMC-severe needed high-level medical care consistently throughout the

**Table 4** Annual hospital visits and admissions vs MC status

Type of visit (n)	Year of home medical care introduction									
	1st year		2nd year		3rd year		4th year		5th year	
	Median	(IQR)	Median	(IQR)	Median	(IQR)	Median	(IQR)	Median	(IQR)
CMC-moderate										
Regular visit	17	(12–22)	15	(9–19)	13	(5.5–18)	13.5	(10–20)	13	(10–16.5)
Emergency visit	4	(1–7)	2.5	(1–5)	3	(0–4)	2.5	(0–4)	2	(1–3)
Total visit	20	(13–29)	17	(12–24)	15	(8–23.5)	15.5	(13–22)	15	(12.5–20.5)
CMC-severe										
Regular visit	15	(11–20)	13	(7–20)	13	(9–17)	13	(10–17)	13	(7–16)
Emergency visit	5	(2–7)	4	(1–6)	2	(1–5)	2	(1–4.5)	2	(1–4)
Total visit	20	(17–26)	16	(11–24)	14.5	(13–19)	16	(12.5–19.5)	15	(7–20)
P-value <sup>†</sup>		0.953		0.938		0.626		0.924		0.918
CMC-moderate										
Planned admission	0	(0–1)	0	(0–1)	0	(0–0)	0	(0–1)	0	(0–1)
Emergency admission	1	(0–3)	1	(0–2)	1	(0–2)	1	(0–1)	0	(0–1)
Total admission	2	(0–4)	1.5	(0–3)	1	(0–2)	1	(0–2.5)	0	(0–1)
CMC-severe										
Planned admission	0	(0–1)	0	(0–1)	0	(0–2)	1	(0–1)	0	(0–1)
Emergency admission	2	(1–4)	1	(0–3)	1	(0–2)	1	(0–2)	1	(0–2)
Total admission	3	(1–4)	1	(0–3)	2	(1–4)	2	(1–3.5)	2	(1–3)
P-value <sup>†</sup>		0.072		0.400		0.086		0.093		<b>0.003</b>

Bold,  $P < 0.05$ . <sup>†</sup>Wilcoxon rank-sum test. To calculate number of visits/admissions to hospital, we did not include CMC receiving care by physician at home ( $n = 6$ ), or children regularly using planned admission as a means of respite care ( $n = 8$ ). The total number of CMC followed up at each year for visits was 86, 85, 70, 58, and 56 from the first to the fifth year, and those for admissions was 84, 83, 58, 56, and 53, respectively. CMC-moderate, children with medical complexity that is moderate; CMC-severe, children with medical complexity that is severe; MC, medical complexity.

5 years. In the fifth year, CMC-severe had a significantly higher number of hospitalizations compared with CMC-moderate. This might be related to the change in medical care needs according to physical development, or to the prognosis of the underlying conditions. Further research is needed to examine the timing and reasons for visits and hospitalizations to prevent emergency visits/admissions and to reduce total hospital-based care utilization.

It is possible that the CMC could have visited or been admitted to other hospitals or clinics, but this was not examined in the present study. Ishida *et al.* examined the health-care utilization of 1,024 children in Japan compared with US children, and found that Japanese children visited a physician in the community clinic 2.5-fold more often, and visited a hospital-based outpatient clinic 11-fold more often than US children.<sup>9</sup> Although health-care utilization varies with country and health-care system, high utilization of health care can become burdensome for families. In a nationwide US survey, 46.3% of CMC families paid >\$US1,000 in out-of-pocket health-care costs, and 54.1% of families stopped working due to the caregiving burden.<sup>10</sup> The CMC, especially technology-dependent CMC, require multiple family members for assistance even for hospital visits. Therefore, both CMC-moderate and CMC-severe have a high caregiver burden with regard to utilization of tertiary hospital-based medical care. Practitioners and policy makers therefore need to keep in mind the fragile physical or psychological health, complex medical needs, support for transportation, financial concern, caregiving burden, and availability of home/community-based services

associated with CMC. Further study is needed to evaluate a wider range of health-care utilization, including multiple hospitals and primary pediatric clinics, and to evaluate caregiving burden with severity of medical care needs.

The other advantage of this study was that it was able to examine a relatively large proportion of CMC in one prefecture, from three tertiary hospitals. Given the prevalence of SMID-MCDG of 0.30/1,000 population aged 0–19 and the proportion of these children living at home of approximately 70%,<sup>4</sup> the number of CMC in the targeted prefecture was estimated at 163; 116 of whom lived at home. The present study involved 92 CMC, that is, 79.3% of the estimated number of CMC at home in this prefecture. This relatively high coverage rate was due to the fact that the three participating hospitals were tertiary pediatric hospitals that provided intensive care and introduced medical care at home. Using this sample of CMC in one prefecture, we investigated the trends in CMC numbers. The number of CMC with introduction of medical care at home increased year by year, especially in CMC <2 years old (Table 2; Fig. 1). In addition, the number of older CMC was also increased in 2010–2014, because some CMC, such as those with muscle disorders, had gradually declining respiratory function that necessitated the introduction of mechanical ventilation at home. The present study may have failed to include some CMC as a result of death, moving house, or stopping of medical care due to improvement in their condition after starting of home medical care in 2014. Even allowing for these possibilities, the number of CMC aged <2 years has been constantly increasing.

A similar tendency of increasing number of children with high medical needs in Japan was also noted in a previous study that examined >7,000 patients with SMID admitted to institutions, in which the number of CMC increased from 1,198 (CMC-severe/moderate, 488/710) in 2001 to 1,457 (CMC-severe/moderate, 676/781) in 2008.<sup>11</sup> CMC admission to institutions has become difficult due to the lack of available beds. It is also problematic that a limited number of children received physician home visit and planned admission as respite care. Therefore, increasing the resources for home/community-based services would be essential to maintain or improve the lives of children with higher medical care needs in the community. The present proportion of CMC receiving home-visit nursing services was higher than in a previous study conducted in 2008,<sup>4</sup> but utilization of other types of home/community-based services was limited. Increasing resources for home/community-based services would be critical to maintain or improve the lives of children with high medical care needs in the community. In addition to increasing services, widening the eligibility criteria for services is also necessary. We excluded some children who could walk or run, even if they needed high-complexity medical care, because a required condition of SMID-MCDG was “physical function at best was the sitting position”. These ambulant children with high medical care needs may require different types of support and consideration in the community. Therefore, further research is needed to include a wider range of children with high medical needs, and to examine the combination of both hospital-based and home-based health-care utilization that can best reduce the care burden related to emergency visits or admissions.

### Limitations and future directions

This study had several limitations. First, the sample consisted of 92 CMC for whom medical claims were obtained from hospital records from 2014 onwards. We could not include children who had home medical care introduction before 2014 that had already ceased; children who moved to another place or were admitted to permanent institutions; or children who died before 2014. The estimated coverage rate of CMC, however, was 79.3%, and the SMID-MCDG score was determined by asking family members about daily care directly, in addition to medical claim data. Therefore, this study was able to assess children with high medical care needs in one prefecture from three hospitals.

Second, information on health-care utilization was limited to medical records from the three hospitals. We could not investigate visits by CMC to other hospitals or clinics in the community. The number of admissions to other hospitals, however, was able to be determined from referral forms, but the number of visits could be determined only for the three target hospitals. Even though the actual number of outpatient visits might be higher, the number of CMC with hospital-based care utilization was still very high. To understand the complete picture of health care utilization, a system that collects data from multiple hospitals is needed.

Third, higher hospital-based care utilization becomes burdensome for children and families, health professionals, and hospitals, and increases the medical care costs. But, regardless of whether the visit or admission was planned or unexpected, parents might have mixed feelings such as relief, stress, or anxiety about the psychological, financial, or social burden. Therefore, future research should examine health-care utilization with regard to caregiving burden and the associated feelings of the families.

In conclusion, CMC in one prefecture had high medical needs and a high utilization of hospital-based services. CMC-moderate and -severe had a consistently high requirement for medical care throughout the 5 years after the introduction of medical care at home. Care coordination with home/community-based and hospital-based services is needed to sustain their lives in the community in light of the recently increasing number of CMC in Japan.

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### Disclosure

The authors declare no conflict of interest.

### Author contributions

Y.Y. and N.T. designed the study; Y.Y., A.W., Y.M. and R.T. collected data; Y.Y. analyzed the data and wrote the manuscript; N.T., A.W., Y.M., R.T., A.M. and R.S. critically reviewed the manuscript and supervised the whole study process. All authors read and approved the final manuscript.

### References

- 1 Asakura T. *Total Care of Children with Severe Motor and Intellectual Disabilities: Seeking the Direction of New Development Supports*. Health Publishing, Tokyo, 2006.
- 2 Nakken H, Vlaskamp C. A need for a taxonomy for profound intellectual and multiple disabilities. *J. Policy Pract. Intellect. Disabil.* 2007; **4**: 83–7.
- 3 Suzuki Y, Takai S, Takechi N *et al.* [New scoring system for patients with severe motor and intellectual disabilities, medical care dependent group.] *J. Jpn. Soc. Severe Mot. Intellect. Disabil.* 2008; **33**:303–9(in Japanese).
- 4 The Ethics Committee of the Japanese Pediatric Association. [Current situation and problems of children with severe motor and intellectual disabilities, the medical care dependent group: Questionnaire survey of 8 prefectures in nationwide.] *J Japan Pediatr. Soc.* 2008; **112**:94–101(in Japanese).



- 5 Cohen E, Kuo DZ, Agrawal R *et al.* Children with medical complexity: An emerging population for clinical and research initiatives. *Pediatrics* 2011; **127**: 529–38.
- 6 Burns KH, Casey PH, Lyle RE, Bird TM, Fussell JJ, Robbins JM. Increasing prevalence of medically complex children in US hospitals. *Pediatrics* 2010; **126**: 638–46.
- 7 Cohen E, Berry JG, Camacho X, Anderson G, Wodchis W, Guttman A. Patterns and costs of health care use of children with medical complexity. *Pediatrics* 2012; **130**: e1463–70.
- 8 O'Mahony L, O'Mahony DS, Simon TD, Neff J, Klein EJ, Quan L. Medical complexity and pediatric emergency department and inpatient utilization. *Pediatrics* 2013; **131**: e559–65.
- 9 Ishida Y, Ohde S, Takahashi O *et al.* Factors affecting health care utilization for children in Japan. *Pediatrics* 2012; **129**: e113–9.
- 10 Kuo DZ, Cohen E, Agrawal R, Berry JG, Casey PH. A national profile of caregiver challenges among more medically complex children with special health care needs. *Arch. Pediatr. Adolesc. Med.* 2011; **165**: 1020–6.
- 11 Sasaki M. Study on the Etiology and Pathology, Treatment and Care, and the Facilities of Patients with Severe Motor and Intellectual Disabilities. Summary report for Ministry of Health, Labour, and Welfare, Research and Development Expenses for Mental and Neurological Diseases. Tokyo, National Center of Neurology and Psychiatry, 2011.