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Research Paper

Rehabilitation Outcomes in Children With Acute Flaccid Myelitis From 2014 to 2019: A Multicenter Retrospective Review

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ABSTRACT

Background: Acute flaccid myelitis (AFM) is a childhood illness characterized by sudden-onset weakness impairing function. The primary goal was to compare the motor recovery patterns of patients with AFM who were discharged home or to inpatient rehabilitation. Secondary analyses focused on recovery of respiratory status, nutritional status, and neurogenic bowel and bladder in both cohorts.
Methods: Eleven tertiary care centers in the United States performed a retrospective chart review of children with AFM between January 1, 2014, and October 1, 2019. Data included demographics, treatments, and outcomes on admission, discharge, and follow-up visits.
Results: Medical records of 109 children met inclusion criteria; 67 children required inpatient rehabilitation, whereas 42 children were discharged directly home. The median age was 5 years (range 4 months to 17 years), and the median time observed was 417 days (interquartile range = 645 days). Distal upper extremities recovered better than the proximal upper extremities. At acute presentation, children who needed inpatient rehabilitation had significantly higher rates of respiratory support ($P < 0.001$), nutritional support ($P < 0.001$), and neurogenic bowel ($P = 0.004$) and bladder ($P = 0.002$). At follow-up, those who attended inpatient rehabilitation continued to have higher rates of respiratory support (28% vs 12%, $P = 0.043$); however, the nutritional status and bowel/bladder function were no longer statistically different.

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Conclusions: All children made improvements in strength. Proximal muscles remained weaker than distal muscles in the upper extremities. Children who qualified for inpatient rehabilitation had ongoing respiratory needs at follow-up; however, recovery of nutritional status and bowel/bladder were similar.

Introduction

Acute flaccid myelitis (AFM) is a potentially devastating illness that has been diagnosed in over 600 children in the United States with acute paralysis since 2014.[1] Enteroviruses have been implicated in causing this illness, with more recent autopsy evidence identifying enterovirus D68 in the cerebrospinal fluid and spinal cord of an infected individual.[2]

AFM presents with sudden-onset paralysis of one to four extremities that is associated with a prodromal febrile illness, typically accompanied by respiratory or gastrointestinal symptoms.[3] Incidence of cases has peaked in the fall of 2014, 2016, and 2018 with fewer cases during the human coronavirus disease pandemic (2019 to 2022).[1] In the most severe cases, children with AFM develop asymmetric weakness that rapidly progresses over hours to days resulting in quadriparesis with respiratory and possible bulbar dysfunction, requiring intubation and feeding support. There is limited evidence to support any one specific treatment in acute care; there are suggestions that early and rigorous physical, occupational, and speech therapies can improve function.[4]

Understanding the trajectory of recovery after AFM can improve the prognostication guidance regarding expected recovery. The primary purpose of this study was to determine the motor recovery patterns of patients with AFM who were discharged home or to inpatient rehabilitation and to characterize the recovery of respiratory status, nutritional status, and neurogenic bowel and bladder in patients admitted to inpatient rehabilitation compared with those who were able to be discharged directly to their homes. We hypothesized that patients who attended inpatient rehabilitation would show greater strength recovery than those who were discharged directly to their home. Secondary outcomes included the measure of recovery in respiratory, nutritional, bowel, and bladder status in all children.

Methods

Data collection

Data were collected from 11 different pediatric hospitals in the United States. Providers from each institution performed retrospective chart reviews on patients diagnosed with AFM to capture prespecified data points. Cases were eligible for inclusion if the child was diagnosed between January 1, 2014, and October 1, 2019, by institutional interpretation of the 2015 Centers for Disease Control and Prevention (CDC)-confirmed case definition of AFM.[5] Cases were excluded if they reported pathology that could confound interpretation of recovery, including the following: brachial plexus injury, spinal cord injury, Guillain-Barré syndrome, transverse myelitis, peripheral neuropathy, relapse of previously diagnosed AFM, evidence of neural autoimmune disease, or presence of pre-existing tracheostomy or gastrostomy tube.

Institutional Review Board (IRB) approval was obtained through Children's Mercy – Kansas City. External sites agreed to rely on Children's Mercy's IRB as the primary institution of approval or pursued institutional approval through a local IRB. Once IRB approval and Data Use Agreements were obtained, an electronic data submission site created through Research Electronic Data Capture (REDCap) was distributed. REDCap is a secure, web-based software platform designed to support data capture for research studies, providing an intuitive interface for validated data capture, audit trails for tracking data manipulation and export procedures, automated export procedures for seamless data downloads to common statistical packages, and procedures for data integration and interoperability with external sources.[6,7]

Data elements

For each record, data from the acute presentation for hospital admission and at most recent follow-up were collected. For those admitted to inpatient rehabilitation, additional data were also collected at admission to and discharge from inpatient rehabilitation.

Acute hospitalization data

At the time of initial hospitalization, the disease characteristics (prodromal symptoms, location and extent of weakness upon onset, imaging of the spinal cord, virology, autonomic dysfunction, pain, and respiratory status) were recorded. Interventions, including medications, intubation, ventilation requirements, tracheostomy placement, and use of continuous positive airway pressure, bilevel positive airway pressure, cough assist, suction, oxygen, and a nasogastric tube or gastrostomy tube, were also collected. Patients were considered to have neurogenic bladder if they required indwelling or intermittent catheterizations for bladder management and neurogenic bowel if they required scheduled daily bowel medications by medical record review.

Strength, determined by the weakest manual muscle testing (MMT) examination during their acute care hospitalization, was documented.[8] Extremity strength data were divided into proximal and distal anatomic sections for comparison.[9] *Proximal upper extremity* strength included shoulder abduction, elbow flexion, and elbow extension; *distal upper extremity* strength included wrist extension, finger abduction, and finger flexion; *proximal lower extremity* strength included hip flexion and knee extension; and *distal lower extremity* strength included ankle dorsiflexion and plantar flexion.

Inpatient rehabilitation

If following their acute care hospitalization, patients were discharged to inpatient rehabilitation; their pediatric functional independence measures (WeeFIM) in self-care, mobility, and cognition were collected on admission and discharge along with length of stay. Each functional domain of the WeeFIM has five to eight tasks that were scored on a scale of 1 to 7 depending on the support needed to complete the task.[10] A score of 1 to 5 indicated the child needed assistance to perform a task. A score of 6 or 7 indicated no assistance was required.[11] Raw WeeFIM domain scores and totals were then divided by age-predicted norms and multiplied by 100 to obtain a developmental functional quotient (DQ). DQs are population-based percentiles of predicted WeeFIM scores from ages five to 83 months and remove age-specific bias when there are a significant number of children aged less than seven

years in a population sample (e.g.., for a two-year-old, the average level of function recorded at 30 months or 24 + 6 months was used).[11] Changes in WeeFIM scores from admission to discharge were calculated and then analyzed using DQ.

Analysis

Descriptive statistics were computed for the acute hospitalization data and compared with Fisher exact test. For study variables in which multiple concurrent values could be selected (e.g., location of weakness), each item was analyzed as a binary indication with a 2 × 2 Fisher test. The resulting set of test statistics was then adjusted for multiple comparisons using the Holm-Bonferroni step-down method.[12,13]

Next a series of analyses explored the MMT scores of patients admitted to inpatient rehabilitation and those discharged home. To begin MMT scores across the four extremity regions were compared between cohorts at both the acute and follow-up time points, independently. Given potential variance in the measurement or patient cohorts between centers, a linear mixed model was used with random intercepts for each institution ID, dependent variable of MMT extremity score, and a singular independent variable for cohort. Models were fit with a Nelder-Mead optimizer, using statsmodels package v0.11.1,1[14] and dependent variables underwent a Box-Cox power transformation where needed to aid in obtaining normal residuals assessed by the Jarque-Bera test.

We then compared the change in MMT scores between acute hospitalization and follow-up. Again, a mixed model was used; however, the dependent variable represents the total change (follow-up minus acute score) for each child with independent variable for the cohort. The model was fit without an intercept, to obtain coefficients for each cohort separately but continue to allow

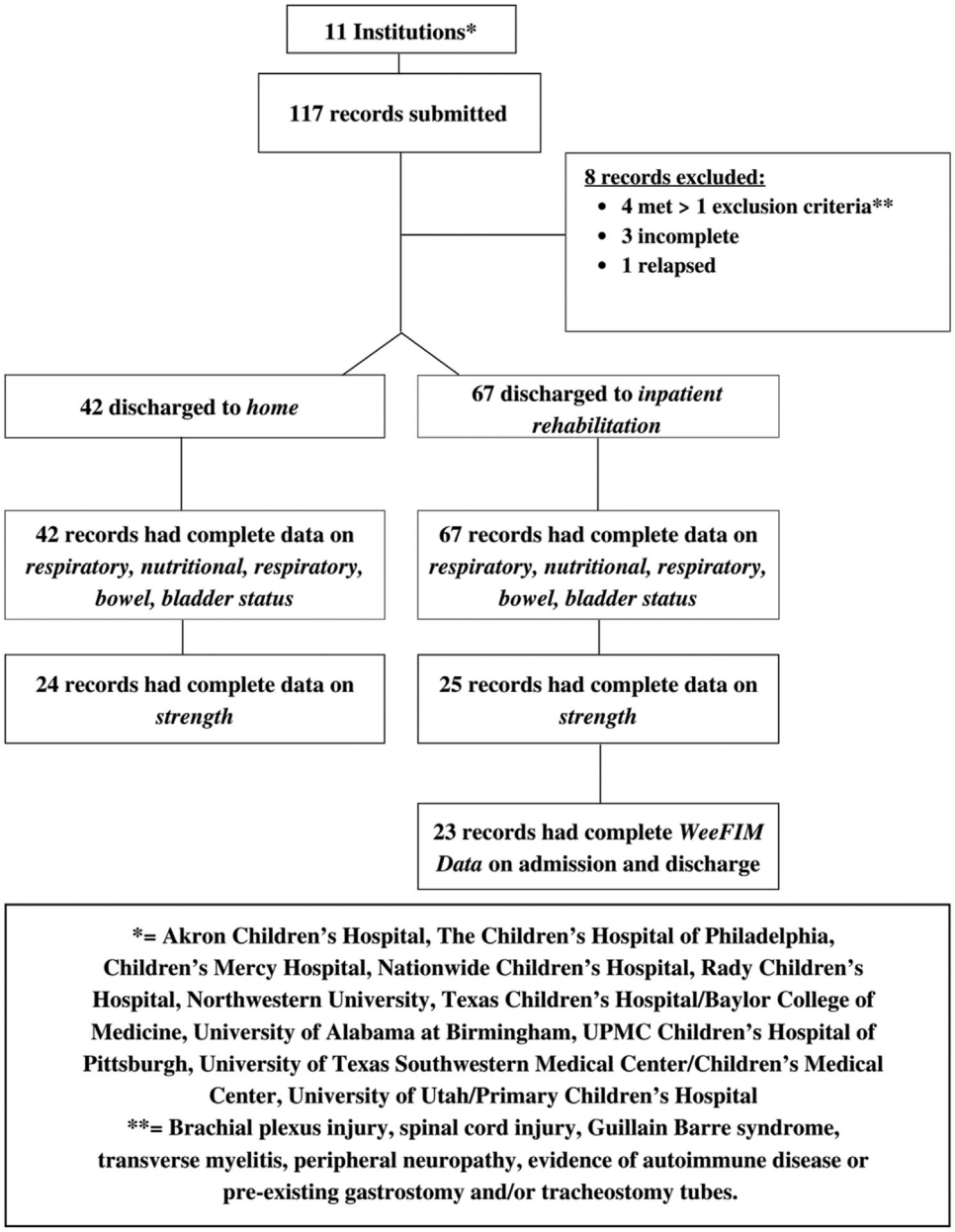

Figure 1. Inclusion and exclusion criteria.

TABLE 1.
Patient Demographics

Characteristics	Discharged to Home (N = 42)	Discharge to Inpatient Rehabilitation (N = 67)	Totals (N = 109)	*P* Value	Adjusted *P* Value*
Age					
0-3 years	12 (29%)	28 (42%)	40 (37%)	0.4	NA
4-5 years	10 (24%)	13 (19%)	23 (21%)		
6 years and up	20 (48%)	26 (39%)	46 (42%)		
Gender					
Male	20 (48%)	36 (54%)	56 (51%)	0.85	NA
Female	21 (50%)	30 (45%)	51 (47%		
Unknown	1 (2%)	1 (1%)	2 (2%)		
Location of weakness					
Bulbar	10 (24%)	16 (24%)	26 (24%)	1	1
Trunk	10 (24%)	25 (37%)	35 (32%)	0.21	1
RUE	26 (62%)	38 (57%)	64 (59%)	0.69	1
LUE	17 (40%)	39 (58%)	56 (51%)	0.07	0.55
RLE	19 (45%)	37 (55%)	56 (51%)	0.33	1
LLE	21 (50%)	38 (57%)	59 (54%)	0.56	1
Other	2 (5%)	10 (15%)	12 (11%)	0.12	0.74
Viral positivity					
EVD68	11 (26%)	17 (25%)	28 (26%)	1	1
EVA71	2 (5%)	0	2 (2%)	0.15	0.73
Other	12 (29%)	25 (37%)	37 (34%)	0.41	1
Unknown	6 (14%)	6 (9%)	12 (11%)	0.53	1
Negative	14 (33%)	26 (39%)	40 (37%)	0.68	1
MRI hyperintensities					
Brainstem	17 (40%)	20 (30%)	37 (34%)	0.3	1
Cervical	30 (71%)	53 (79%)	83 (76%)	0.37	1
Thoracic	25 (60%)	45 (67%)	70 (64%)	0.54	1
Lumbar	11 (26%)	17 (25%)	28 (26%)	1	1
Sacral	9 (21%)	12 (18%)	21 (19%)	0.8	1
Other	6 (14%)	7 (10%)	13 (12%)	0.56	1
No records	1 (2%)	1 (1%)	2 (2%)	1	1
Acute treatment					
IVIG	31 (74%)	54 (81%)	85 (78%)	0.47	1
Steroids	29 (69%)	54 (81%)	83 (76%)	0.17	1
Plasmapheresis	9 (2%)	29 (43%)	38 (35%)	0.02	0.18
Antivirals	7 (2%)	13 (19%)	20 (18%)	0.8	1
Antibiotics	1 (2%)	14 (21%)	15 (14%)	0.008	0.08
Fluoxetine	1 (2%)	14 (21%)	15 (14%)	0.008	0.08
Other	0	2 (3%)	2 (2%)	0.52	1
Unknown	0	1 (1%)	1 (0.01%)	1	1
None	2 (5%)	0	2 (2%)	0.14	1
Pain	26 (62%	47 (70%)	73 (67%)	0.28	NA
Autonomic dysfunction	11 (26%)	25 (37%)	36 (33%)	0.2	NA
Time from acute presentation to last follow-up (days)	627 (S.D. ± 576.2)	591 (S.D. ± 431.5)	NA	0.61	NA

Abbreviations:
IVIG = Intravenous immunoglobulin
LLE = Left lower extremity
LUE= Left upper extremity
MRI = Magnetic resonance imaging
RLE = Right lower extremity
RUE = Right upper extremity
There were no significant differences between groups in regards to age, gender, location of weakness, and viral positivity.
* Compared using the Holm-Bonferroni step-down method.

data to be modeled together to retain power. Finally, to evaluate if the *degree* of change between follow-up and acute strengths scores was significantly different for those who underwent inpatient rehabilitation versus those who did not, the same model was fit; however, an intercept was included in the model to capture the relative difference in scores between the cohorts.

We concluded with an evaluation of the DQ WeeFim scores for those patients who were admitted to inpatient rehabilitation. Changes in WeeFim scores from admission to discharge were assessed across the categories of self-care and mobility and cognition, independently. Again, a series of linear mixed model was used to model change. No independent variables were included beyond the intercept, representing a hypothesis of 0 change between time points for the cohort.

Results

In total 117 records from patients were collected, of which eight were excluded. Four of these patients met at least one exclusion criteria; three records were not completed and the eighth reported an incidence of relapse. As such, 109 medical records met the final inclusion criteria and were carried forward for use in the analyses throughout the remainder of this article (Fig 1).

There were no significant differences between children who required inpatient rehabilitation and those who did not with respect to age, gender, location of weakness, location and presence of magnetic resonance imaging hyperintensities, viral positivity, therapeutic medications, pain, or autonomic dysfunction (Table 1). More than half (n = 73.67%) reported pain and a third (n = 36.33%)

TABLE 2.
Respiratory, Nutritional, and Elimination Status

System	Acute Presentation			*P* Value	Follow-up			*P* Value
	Home (N = 42)	Inpatient Rehabilitation (N = 67)	Total (N = 109)		Home (N = 42)	Inpatient Rehabilitation (N = 67)	Total (N = 109)	
Respiratory support	9 (21%)	43 (64%)	52 (48%)	<0.001	5 (12%)	19 (28%)	24 (22%)	0.043
Intubation	8 (19%)	37 (55%)	45 (41%)	<0.001	0	0	0	†
Tracheostomy/ventilator	0	0	0	†	3 (7%)	10 (15%)	13 (12%)	0.222
Tracheostomy/room air	0	2 (3%)	2 (2%)	†	0	2 (3%)	2 (2%)	†
NIVV*	1 (2%)	4 (6%)	5 (5%)	0.383	2 (5%)	7 (17%)	9 (8%)	0.293
Nutritional support	10 (24%)	37 (55%)	47 (43%)	0.001	4 (10%)	13 (19%)	17 (16%)	0.166
Neurogenic bowel	7 (17%)	29 (43%)	36 (33%)	0.004	3 (7%)	8 (12%)	11 (10%)	0.418
Neurogenic bladder	7 (17%)	30 (45%)	37 (34%)	0.002	1 (2%)	4 (6%)	5 (5%)	0.383

Abbreviations:
BIPAP = Bilevel positive airway pressure
CPAP = Continuous positive airway pressure
NIVV = Non-invasive ventilation
The patients who were discharged to inpatient rehabilitation were more affected on acute presentation. On follow-up, those discharged to inpatient rehabilitation had higher rates of respiratory support but nutritional status and bowel/bladder recovery were similar to those discharged directly home
* Includes CPAP, BIPAP, cough assist, supplemental oxygen, and/or suction.
† *P* values were not calculated for proportions containing 0.

had signs of autonomic dysfunction. Hypertension (n = 24.22%) was the most common sign of autonomic dysfunction, followed by tachycardia (n = 16.15%), bradycardia (n = 7.6%), and fevers of unknown origin (n = 7.6%) (Table 1).

At acute presentation, compared with children who were discharged home, children who were eventually discharged to inpatient rehabilitation had higher rates of intubation (64% vs 21%, $P < 0.001$), external nutritional support (55% vs 24%, $P = 0.001$), and neurogenic bowel (43% vs 17%, $P = 0.004$) and bladder (45% vs 17%, $P = 0.002$) (Table 2). The respiratory system remained impaired for both groups, although the inpatient rehabilitation group still had higher rates of respiratory support at follow-up (28% vs 12%, $P = 0.043$). However, there were no significant differences at follow-up between groups in need for external nutritional supports (19% vs 10%, $P = 0.16$) and rates of neurogenic bowel (12% vs 7%, $P = 0.418$) and neurogenic bladder (6% vs 2%, $P = 0.38$) (Table 2).

The children who required inpatient rehabilitation had lower MMT scores at acute presentation and follow-up than those who were discharged home (Table 3). This was statistically significant in the proximal and distal lower extremities at both acute presentation and follow-up (Supplemental Table 2). At follow-up, both groups had strength recovery, but this was not statistically significant (Table 4). The upper extremities remained weaker in the proximal muscles than the distal muscles for both groups (Table 3).

Functional assessment (WeeFIM) data were available for 23 of the 67 eligible children who received inpatient rehabilitation. Mobility and self-care were the most affected domains upon admission with all domains improving by discharge (Fig 2). Using the mixed model to remove hospital variance, WeeFIM DQ self-care improved on average by 16.0 (95% confidence interval [CI] 7.5 to 24.6, $P < 0.001$), mobility by 21.2 (95% CI 12.5 to 29.9, $P < 0.001$), and cognition by 2.1 (95% CI 1.7 to 2.5, $P < 0.001$) (Fig 2).

At outpatient follow-up, children who attended inpatient rehabilitation had significantly higher rates of orthotic use (46% vs 17%, $P = 0.002$), manual wheelchair use (24% vs 7%, $P = 0.025$), and power wheelchair use (21% vs 5%, $P = 0.021$). This difference was not present for walking aids (1% vs 5%, $P = 0.46$) or medical-grade strollers (6% vs 1%, $P = 0.383$) (Supplemental Table 1).

Discussion

This study evaluated a large cohort of children with AFM who were seen by either a physiatrist or neurologist between 2014 and 2019. At this time, the CDC has recognized 682 confirmed cases of AFM since it began tracking this diagnosis in 2014.[1] Although the authors did not directly confirm all 109 children represented in this study had been reported to the CDC, the inclusion criteria directly matched the CDC definition of AFM. The primary objective of this study was to evaluate the motor recovery of patients with AFM who were discharged home or to inpatient rehabilitation. Overall, children who were admitted to inpatient rehabilitation were weaker at onset than those who were discharged home. The need for

TABLE 3.
Average Strength Scores Using MMT Scale at Acute Presentation and Follow-up for Both Groups

Extremity	Region	Discharged to Home (N = 24)		Discharged to Inpatient Rehabilitation (N = 25)	
		Acute Presentation Mean (S.D.)	Follow-up Mean (S.D.)	Acute Presentation Mean (S.D.)	Follow-up Mean (S.D.)
Upper	Proximal	3.06 (1.42)	3.76 (1.35)	2.00 (2.07)	3.04 (1.81)
	Distal	3.33 (1.45)	4.09 (1.35)	2.31 (2.14)	3.33 (1.82)
Lower	Proximal	3.03 (1.96)	4.23 (1.41)	1.25 (1.68)	2.96 (1.91)
	Distal	3.13 (2.01)	4.28 (1.43)	1.47 (1.91)	2.85 (1.96)

Abbreviation:
MMT = Manual muscle testing
MMT scale 0 to 5: 0 = flaccid, 1 = flicker, 2 = active range of motion with gravity eliminated, 3 = active range of motion against gravity, 4 = examiner can overcome, 5 = full strength.

TABLE 4.
Strength Change From Acute Presentation to Follow-up for Each Group and Between Groups

Extremity	Region	Discharged to Home (*Follow-up-Acute*)	*P* Value	Discharged to Inpatient Rehabilitation (*Follow-up-Acute*)	*P* Value	Strength Return Between Groups (*Inpatient - Home*)	*P* Value
Upper	Proximal	0.71 (0.77)	<0.001	1.04 (1.13)	<0.001	0.33 (0.28)	0.358
	Distal	0.76 (1.01)	<0.001	1.02 (1.46)	<0.001	0.26 (0.36)	0.796
Lower	Proximal	1.2 (1.72)	<0.001	1.71 (1.69)	<0.001	0.51 (0.49)	0.110
	Distal	1.15 (1.79)	<0.001	1.38 (1.98)	<0.001	0.23 (0.54)	0.954

improvement in motor strength was a significant determining factor in discharge disposition. The proximal upper extremities remained weaker than the distal upper extremities for both cohorts. This is similar to a prior study that noted a risk of shoulder subluxation in pediatric patients after AFM.[15] For the lower extremities, strength recovery was more uniform across proximal and distal segments. Long term, there were not significant differences in strength return between groups, even though both groups had improvement in strength.

Previous studies of AFM have looked at functional outcomes, with the majority focusing on strength and the need for assistance in activities of daily living (ADLs). Three-quarters of patients experienced significant challenges in independent ADL completion after four months.[4] In addition, after the acute illness, more than 80% of patients from a nationwide sample remained weak at nine months[16] and 75% of patients in a Colorado cohort remained weak at 12 months.[9] The median follow-up in these studies was 1 year, and the results varied. Owing to the large number of cases in children aged less than seven years, we calculated the DQs in addition to the WeeFIM scores to remove age bias. Having DQ scores available, we were able to compare our inpatient rehabilitation cohort with a similar cohort from Kennedy Krieger.[15] The admission DQ scores were lower in our study than in the Kennedy Krieger cohort, but both groups reported significant gains by discharge with hospital variance removed, supporting the important role of intensive therapy in recovery.[15]

A secondary objective of this study was to examine the manifestations of AFM on the respiratory, gastrointestinal, and urologic systems. As expected, children who were admitted to inpatient rehabilitation had higher respiratory, nutritional, and bowel/bladder needs. These needs may be significant reasons why children are unable to be discharged immediately to a home setting. Few studies have documented the ongoing need for tracheostomies, ventilators, and/or gastrostomy tubes at follow-up,[15] which are significant factors in quality of life[17,18] and contribute to long-term health care expenses.[19,20] A prior AFM inpatient rehabilitation study observed that seven of nine children were eventually cleared for a regular diet after using a gastrostomy tube for nutrition.[15] However, there was a higher incidence of oral aversion in children who required ventilator support, delaying their specific oral-pharyngeal recovery.[15] Owing to the low rate (19% vs 10%, $P = 0.16$) of gastrostomy tube usage at follow-up in this study, it may be reasonable to consider delaying placement of gastrostomy tubes during the critical phase of care unless the child has a tracheostomy. Of note, 47 patients in total (43%) required nutritional support at acute presentation but only 17 (16%) had ongoing nutritional support at most recent follow-up. Ultimately, the consideration of a gastrostomy tube should be at the discretion of the treating physician and family after evaluation of the clinical picture.

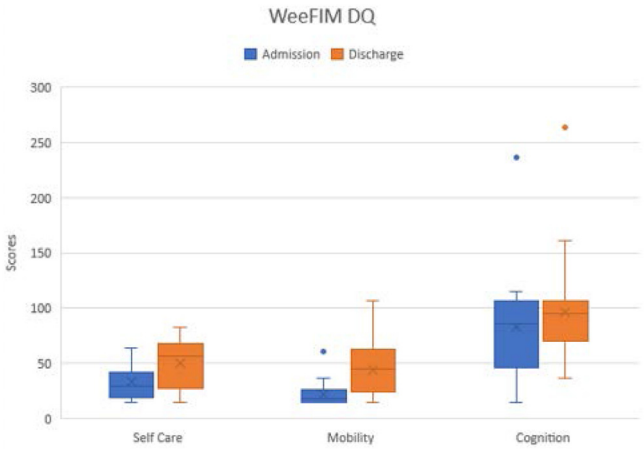

Figure 2. WeeFIM Developmental Quotients for rehabilitation: admission and discharge. Average length of stay was 31 days (S.D. = 45). The color version of this figure is available in the online edition.

The respiratory system appears less resilient, particularly for those who are most affected at onset. Of note, the presence of respiratory or nutritional supports did not prevent children from being discharged home after the acute care setting, but the patients requiring inpatient rehabilitation continued to need more respiratory support at follow-up ($P = 0.04$). Regardless, both cohorts were able to wean respiratory and nutritional supports over time. Subspecialty consultation with pulmonology should be considered for cases with fatigue and reduced endurance but without overt respiratory complaints, as there may be subclinical effects on the respiratory system. There should also be consultation with pulmonology in those children with AFM with thoracic-level neurological involvement regarding pulmonary toileting programs, such as chest physical therapy, insufflation/exsufflation, and scheduled prone positioning, during the acute, rehabilitation, and home phases of care.

During the acute phase, many patients had constipation and urinary retention, necessitating medical intervention. However, the data show that most patients did recover control over their bowel and bladder by follow-up.

In addition to medical and functional recovery, pediatric patients need to return to school. Although specific adaptations were not addressed in this study, social reintegration with peers improves confidence and acceptance. Finally, future studies may consider tracking the incidence of post-polio-like occurrences and impact of AFM on the family as a unit.

Limitations

There are several limitations to our study. The first is the wide range in time between diagnosis and clinical follow-up for each case, which could not be considered when calculating recovery data due to the number of data sources and quality of data available. Another variable is time to recovery, as a child could see functional return between clinical assessments, which could not be captured precisely in a retrospective design with varied follow-up dates. Although strength scores were used in this study, not every individual muscle was tested and standardized strength testing in younger children can be challenging. In addition, there was a small

percentage of patients in each cohort who had complete strength data, which limits subgroup analyses to see if specific populations improve more completely. Finally, due to the small cohort of patients with AFM it is likely that some patients in this study have been reported in other publications.

Conclusions

All children made improvements in strength, with proximal upper extremities remaining weaker than distal segments, whether they were discharged to home or to an inpatient rehabilitation setting. Children who attended inpatient rehabilitation had higher need for ongoing respiratory support at follow-up, whereas recovery of nutrition and bowel/bladder function was similar between groups. Future areas of research include the timing of clinical strength plateau, the long-term trajectory of recovery, considerations for nerve transfers, and the incidence of proximal joint subluxation.

Declaration of Competing Interest

All coauthors have read and agreed to the content of the manuscript and have no conflicts of interest. There was no commercial involvement in the study design or manuscript preparation.

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Supplementary data

Supplementary data related to this article can be found at https://doi.org/10.1016/j.pediatrneurol.2023.04.027.

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