ORIGINAL ARTICLE

Risk Factors for Long Bone Fractures in Patients with Severe Motor and Intellectual Disabilities: A 6-year Follow-up Retrospective Study

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Objectives: We investigated the clinical situation of fractures that occurred in patients in the severely disabled patients' ward of our hospital. The study aimed to identify risk factors for the occurrence of long bone fractures in the extremities, which pose problems in nursing care. **Methods:** We retrospectively studied fractures that occurred between April 2015 and March 2021 among a total of 126 patients in the severely disabled patients' ward of our hospital. The fracture site, frequency of occurrence, cause of injury, and other parameters were investigated. We statistically compared the fracture group and non-fracture group with respect to age, sex, body position before fracture, motor function, food intake status, body mass index, use of anti-epileptic drugs, hip dislocation, and maximum extension angle and range of motion of elbow/knee joints. **Results:** Among 126 patients, a total of 35 fractures occurred in 28 patients (22%). There were 19 long bone fractures of the extremities in 17 patients. Multiple logistic regression analysis using the occurrence of long bone fractures of the extremities as the objective variable identified the following significant independent variables: age [odds ratio (OR)=1.087, P=0.008], maximum extension angle of the elbow joint (OR=1.039, P=0.023), range of motion of the elbow joint (OR=0.940, P=0.003), and range of motion of the knee joint (OR=0.972, P=0.034). **Conclusions:** This study reveals that older age and flexion contracture of elbow and knee joints are risk factors for the occurrence of long bone fractures in severely disabled patients.

Key Words: fracture; intellectual disabilities; motor disabilities; risk factors

INTRODUCTION

Children with severe motor and intellectual disabilities are known to be affected by bone fragility, and the application of a weak external force can easily result in fracture.[1] Because fractures may occur during routine nursing care despite the absence of a strong external force, the origin of the fracture is unknown in many cases, and establishing prevention and treatment methods is an important issue.[2] In addition, among patients with severe motor and intellectual disabilities (hereinafter "severely disabled patients"), those who are bedridden are considered prone to long bone fractures associated with some nursing care maneuvers. When a fracture occurs, much effort is required to provide treatment and hospital support for the patient, as well as support to the family.

The causes of fractures include bone fragility, the main contributor to which is osteoporosis. Severely disabled patients are known to be predisposed to developing osteoporosis because of a range of factors that includes reduced physical activity, long-term bed confinement, long-term use of anti-epileptic drugs, deficiency in vitamins and trace ele-

Table 1. Patient characteristics

		Total (n=126)
Sex	Male	66 (52.4)
	Female	60 (47.6)
Age, years		43.9 ± 19.8 (3–81)
Causes of severe motor and intellectual disabilities	Cerebral palsy	45 (35.7)
	Encephalopathy	23 (18.3)
	Encephalitis	21 (16.7)
	Cerebral malformation	14 (11.1)
	Chromosomal abnormality	9 (7.1)
	West syndrome	4 (3.2)
	Intracerebral hemorrhage	3 (2.4)
	Neurodegenerative disease	3 (2.4)
	Brain tumor	2 (1.6)
	Traumatic brain injury	2 (1.6)

Data presented as number (percentage). Data for age presented as mean ± standard deviation (range).

ments, and insufficient sun exposure.[2] In addition, severely disabled patients often have severe joint contracture caused by underlying diseases such as cerebral palsy, which may be a pathological factor for fractures. When an external force is applied to the extremities, because the contractured joints cannot move, an excessive force is exerted near the long bone joints, causing fracture.[3] Although the factors causing fractures in severely disabled patients and patients with cerebral palsy have been described in several reports,[1,4–9] there are no reports on the relationship between joint range of motion and fractures in severely disabled patients.

In this study, we investigated the clinical situations of fractures that occurred in patients hospitalized in the ward for severely disabled patients of our hospital. We analyzed the risk factors for long bone fractures of the extremities (humerus, antebrachium, femur, and crus), focusing on the limitation of joint ranges of motion.

SUBJECTS AND METHODS

In this retrospective observational study, we studied 126 severely disabled patients [66 males and 60 females, average age 43.9 ± 19.8 (range 3 to 81) years] who were hospitalized in the severely disabled patients' ward of our hospital as of April 1, 2015 and could be followed for the subsequent 6 years. The causes of severe motor and intellectual disabilities included cerebral palsy (n=45), encephalopathy (n=23), encephalitis (n=21), cerebral malformation (n=14), chromosomal abnormality (n=9), West syndrome (n=4), intracerebral hemorrhage (n=3), neurodegenerative disease (n=3),

brain tumor (n=2), and traumatic brain injury (n=2) (**Table 1**). None of the patients had systemic bone diseases including osteogenesis imperfecta that manifests bone fragility as the main symptom. All the fractures diagnosed by orthopedic surgeons during the 6 years from April 2015 to March 2021 were registered, and each fracture was investigated in terms of frequency, fracture site, cause of injury, and other parameters. All data were collected from the medical charts and incident reports submitted by nurses and other hospital staff members. In addition, to identify the risk factors for the occurrence of fractures in the long bones of the extremities (humerus, antebrachium, femur, and crus), we extracted data for the following 12 items before fracture: age, sex, body position (could maintain sitting position, recumbent position only), motor function (could ambulate alone, show spontaneous movements, few spontaneous movements), food intake status (tube feeding, oral intake + tube feeding, oral intake), body mass index (BMI), use of antiepileptic drug (two or more drugs, monotherapy, none), hip dislocation on plain radiograph (present, absent), and maximum extension angle and range of motion of elbow and knee joints. These items were compared statistically between patients with long bone fracture of the extremities (fracture group) and patients without any fracture (non-fracture group). The maximum extension angle and flexion angle of left and right elbow and knee joints were measured in all patients by physiotherapists. From the values obtained for the left and right joints, the values with greater limitation were used in analyses (**Fig. 1**).

For statistical analysis, univariate analyses were performed using a χ^2 test, a two-sample *t*-test, and the Mann–Whitney
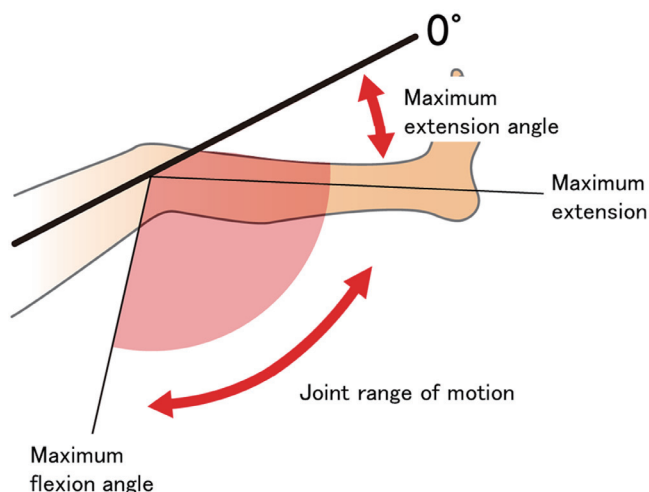
Fig. 1. Measurement of joint range of motion. The maximum flexion angle and maximum extension angle of left and right elbow and knee joints were measured, and joint ranges of motion were calculated. From the results obtained from left and right joints, the value with greater limitation was adopted.

Fig. 2. Sites of 35 fractures in 28 patients.

U test. For multivariate analysis, covariates were entered by stepwise regression. The following covariates were included in the multivariate analysis: age, body position, maximum extension angle of the elbow joint, range of motion of the elbow joint, and range of motion of the knee joint. A logistic regression model was fitted to assess the independent predictors of fracture. In all analyses, the significance level was set at less than 5%. Statistical analyses were performed using StatFlex ver 7 (Artec, Osaka, Japan).

The study was approved by the Ethical Committee of the National Kofu Hospital (approval number R3-2), and patients were given the option to withdraw from the study through our website. Because of the noninvasive, observational nature of our study, the ethics committee waived the need for written informed consent from each patient.

RESULTS

During the 6-year study period, a total of 35 fractures occurred in 28 of 126 patients (22%, mean age 56.5 years). Of these, 3 patients had multiple fractures: 2 patients had 2 fractures and 1 patient had 5 fractures. The annual incidence of fractures, expressed as the annual number of fractures per 100 patients, was 4.63%. By fracture site, there were 2 humerus fractures, 1 ulna fracture, 12 femur fractures, 4 tibia fractures, 5 phalanx (hand) fractures, 8 phalanx (foot)
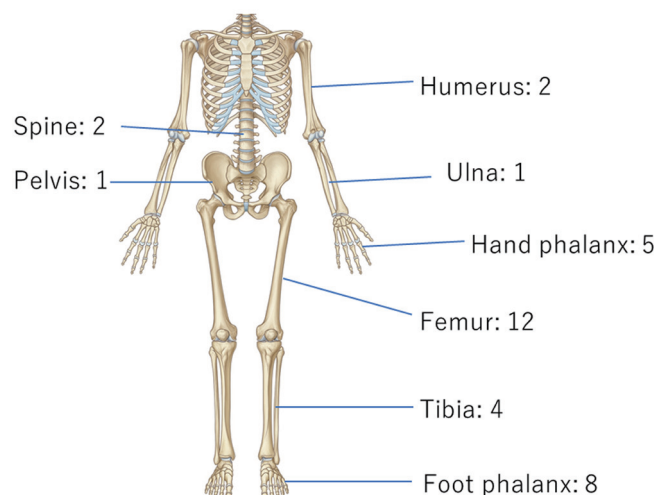
fractures, and 3 spine or pelvis fractures (**Fig. 2**). Of these, 19 long bone fractures of the extremities occurred in 17 patients, and 11 fractures were around the knee joint. The cause of injury was unknown in 22 of the 35 fractures. Six fractures were caused by stress on the joint or affected part while performing the following activities: diaper changing or when changing clothes (3 fractures), crawling on hands and knees (1 fracture), nail care (1 fracture), and during involuntary movement (1 fracture). Four fractures were caused by passive position change performed by a caregiver. One fracture occurred while in a wheelchair. Two fractures were caused by transfer to a wheelchair (**Table 2**). A univariate analysis that compared the fracture group (17 patients with long bone fractures of the extremities) with the non-fracture group (98 patients with no fracture) showed a significant difference in age (P=0.005). However, the analysis showed no significant difference between the groups for sex, body position, motor function, food intake status, BMI, anti-epileptic drug use, or the presence or absence of hip dislocation (**Table 3**). The maximum extension angle of the elbow joint was −75.9 ± 52.8 degrees (mean ± standard deviation) in the fracture group and −43.5 ± 47.3 degrees in the non-fracture group (P=0.014), whereas the range of motion of the elbow joint was 60.0 ± 44.8 degrees in the fracture group and 97.8 ± 42.5 degrees in the non-fracture group (P=0.001). These results showed significant differences between the two groups and indicated severe flexion contracture of the elbow joint in the fracture group (**Fig. 3**). Multiple logistic regression analysis used the occurrence of long bone fractures of the extremities as the objective variable and five items obtained before fracture were used as explanatory variables. Four of these

Table 2. Causes of fracture

Cause	Details	No. of patients
Unknown		22
Stress on joint or affected part	While changing diaper or changing clothes	3
	While crawling on hands and knees	1
	During nail care	1
	During involuntary movement	1
Passive position change by caregiver		4
While in a wheelchair		1
Transfer to a wheelchair		2

Table 3. Univariate comparison of 17 patients in fracture group and 98 patients in non-fracture group

Long bone fracture of extremities		Fracture group (n=17)	Non-fracture group (n=98)	P value
Age (years)		55.1 ± 21.1	40.3 ± 19.2	0.005
Sex	Female	10 (58.8)	44 (44.9)	0.288
	Male	7 (41.2)	54 (55.1)	
Body position	Could maintain sitting position	10 (58.8)	54 (55.1)	0.776
	Recumbent position only	7 (41.2)	44 (44.9)	
Motor function	Could ambulate alone	1 (5.9)	16 (16.3)	0.700
	Had spontaneous movements	12 (70.6)	56 (57.1)	
	Little spontaneous movements	4 (23.5)	26 (26.5)	
Food intake status	Tube feeding	4 (23.5)	53 (54.1)	0.052
	Oral intake + tube feeding	2 (11.8)	1 (1.0)	
	Oral intake	11 (64.7)	44 (44.9)	
BMI (kg/m^2)		14.094 ± 2.688	13.938 ± 2.699	0.826
Anti-epileptic drug use	Two or more drugs	7 (41.2)	23 (23.5)	0.628
	Monotherapy	2 (11.8)	33 (33.7)	
	None	8 (47.1)	42 (42.9)	
Hip dislocation	Present	7 (41.2)	40 (40.8)	0.978
	Absent	10 (58.8)	58 (59.2)	

Data presented as number (percentage) or as mean ± standard deviation.

variables were identified as independent factors associated with the occurrence of long bone fractures of the extremities: age [odds ratio (OR)=1.087; 95% confidence interval (CI): 1.022–1.155; P=0.008], maximum extension angle of the elbow joint (OR=1.039; 95% CI: 1.005–1.074; P=0.023), range of motion of the elbow joint (OR=0.940; 95% CI: 0.902–0.979; P=0.003), and range of motion of the knee joint (OR=0.972; 95% CI: 0.946–0.998; P=0.034). As the fifth explanatory variable, body position showed no significant association with the occurrence of fractures (**Table 4**).

DISCUSSION

Yokoi and Umeki[3] reported that the annual incidence of fractures (annual number of fractures per 100 patients) in the severely disabled patients' wards of 64 facilities in Japan was 2%–3%. Inoue[10] surveyed an average of 113.4 patients per day hospitalized in a severely disabled patients' ward and found seven fractures occurring in eight patients over a period of 10 months (annual incidence of 8.5%). Leet et al.[7] also reported a 12% prevalence of fractures in a study of 418 children with cerebral palsy. The present study of 126 patients showed a total of 35 fractures occurring over a 6-year period, with an annual incidence of 4.6%. These data
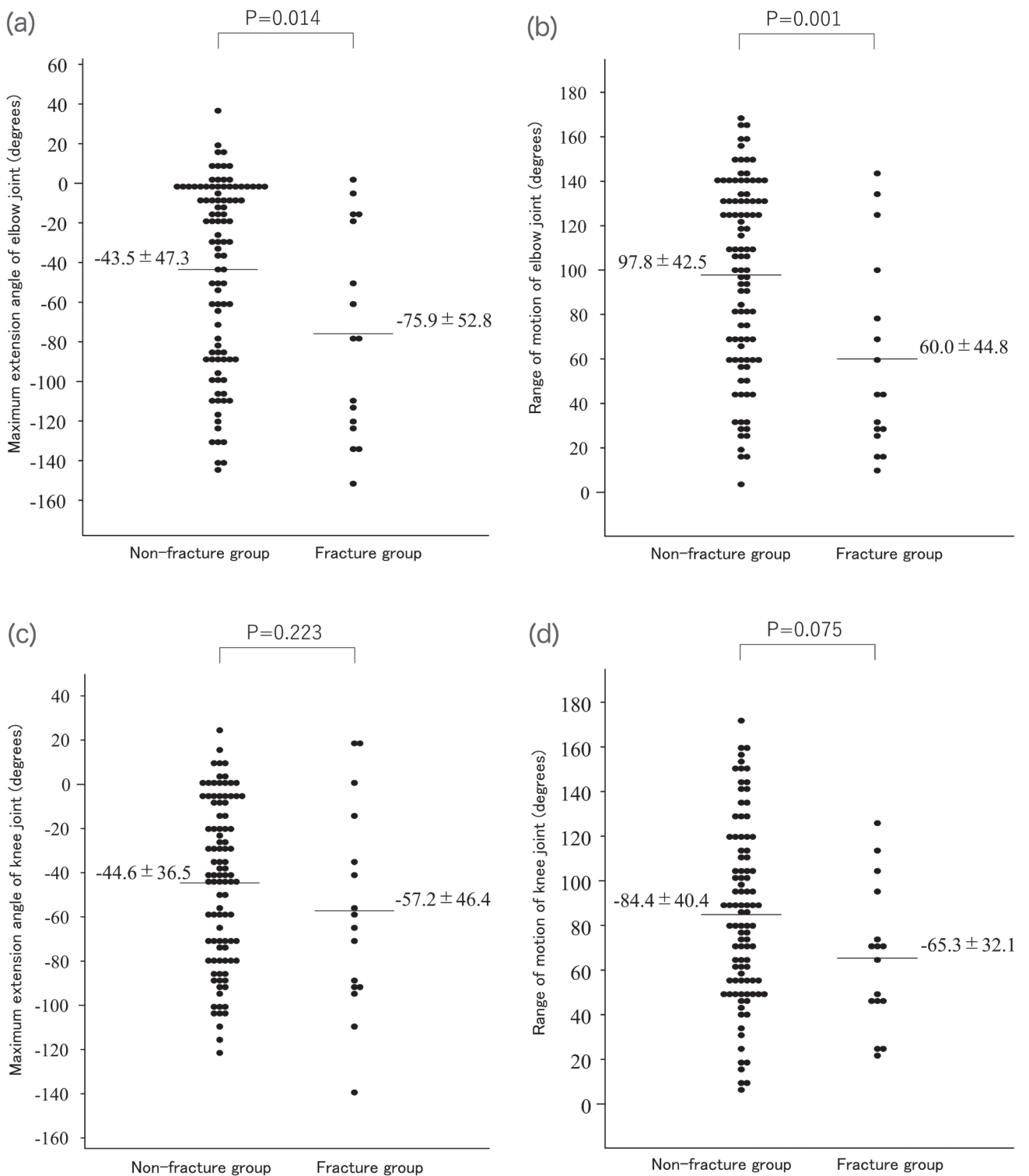
Fig. 3. Comparisons between the fracture group (n=17) and the non-fracture group (n=98) in (a) maximum extension angle of elbow joint (degrees), (b) range of motion of elbow joint (degrees), (c) maximum extension angle of knee joint, and (e) range of motion of knee joint (degrees). Horizontal bars indicate mean values. The mean ± standard deviation for each dataset is shown next to each dot plot.

Table 4. Results of multiple logistic regression analysis

Variable	Odds ratio	95% confidence interval		P value
		Lower limit	Upper limit	
Age	1.087	1.022	1.155	0.008
Body position	7.752	0.806	74.567	0.076
Maximum extension angle of elbow joint	1.039	1.005	1.074	0.023
Range of motion of elbow joint	0.940	0.902	0.979	0.003
Range of motion of knee joint	0.972	0.946	0.998	0.034

clearly show that the frequency of fractures occurring in severely disabled patients is not low. With aging of hospitalized patients in the future, the incidence of fractures is expected to increase along with the impact of age-related osteoporosis.

In terms of fracture sites, Yokoi and Umeki[3] reported that femur fracture was the most frequent in severely disabled Japanese patients, followed by crus fracture and humerus fracture, in that order. Brunner and Doderlein[6] studied 37 cerebral palsy patients with 54 fractures that occurred without significant trauma and found that the majority (74%) occurred in the femoral shaft and supracondylar region. In the present study, 24 of 35 fractures occurred in the lower extremities, with femur fracture being the most frequent. Fractures of the lower extremities, especially femur fractures, require procedures such as surgery and external fixation with a cast, which add great burden to nursing care; hence their prevention is highly desirable.

The predisposition of severely disabled patients to osteoporosis and fractures is attributed to reduced spontaneous movements and other factors such as deficiency in vitamins and trace elements and insufficient sun exposure.[1] Henderson et al.[11] also reported a high prevalence of low bone mineral density in children with moderate to severe cerebral palsy, and noted its significant association with fracture risk. Furthermore, Sakai[12] observed that almost all institutionalized severely disabled patients had severe osteoporosis across all age groups and pointed out the involvement of various factors such as insufficient load and use of anti-epileptic drugs. Therefore, decreased bone mineral density is a risk factor for fractures, and probably all severely disabled patients develop osteoporosis independent of aging. Furthermore, Sakai et al.[13] reported that bone resorption markers in female patients with severe motor and intellectual disabilities increased beyond 50 years of age, possibly representing a sign of primary osteoporosis. Given the increased life expectancy of patients with severe motor and intellectual disabilities, physicians may need to consider the tendency of decreased bone mineral density in older patients. In the present study, the risk of long bone fracture was associated with age, indicating that greater attention is needed in older patients.

In a study of younger patients, Mergler et al.[14] reported the frequent presence of disrupting factors in bone mineral density measurement in children with severe neurological impairment and intellectual disability, most notably movement during measurement, aberrant body composition, small length for age, and scoliosis. They concluded that physicians should be aware of the possible influence of these disrupting factors on the accuracy of dual-energy X-ray absorptiometry. Henderson et al.[11] listed motion artifacts, previous hip or scoliosis surgery, and contractures as factors affecting the reliability of bone mineral density assessment in the proximal femur and occasionally in the lumbar spine. Because of the presence of spinal deformity and joint contracture in severely disabled patients, it is difficult to routinely measure bone mineral density in all such patients. In the present study, we were unable to obtain reproducible bone mineral density data from all patients. Therefore, we conducted this research on the premise that all patients had bone fragility, and we investigated the risk factors for long bone fractures with a focus on joint contractures in the extremities.

Severely disabled patients often have severe joint contractures because of underlying diseases such as cerebral palsy, and the pathological relation with fracture is considered as follows. When an external force is applied to the extremities, the immobility of the joints causes excessive force to be exerted on the long bones, resulting in fracture. Brunner and Doderlein[6] considered that the long, fragile lever arms and stiffness in major joints, notably the hips and knees, were the main causes of fractures. In the present study, we measured the maximum extension angle and the range of motion of elbow and knee joints as indicators of the degree of joint contracture and examined the relationship between joint contracture and fractures in severely disabled patients. Multiple logistic regression analysis identified the maximum extension angle of the elbow joint, the range of motion of the elbow joint, and the range of motion of the knee joint

as independent variables associated with the occurrence of long bone fractures of the extremities. These results clearly indicate that limited range of motion in the elbow joint and the knee joint; that is, contracture, is a risk factor for fracture in this patient population.

Regarding other risk factors for fractures, Kawada[4] found that age, use of sodium valproate, and bone mineral density (percentage of normal levels) significantly affected bone fractures in severely disabled persons. Ko et al.[8] reported low weight and prolonged postoperative immobilization as risk factors for long bone fractures of the extremities in non-ambulatory cerebral palsy children. Patel et al.[9] found that among the risk factors they studied, use of a gastrostomy feeding device was the only variable associated with an increased risk of fracture. In the present study, although analyses revealed that age and limited range of motion were risk factors for the occurrence of fractures, further detailed studies on more factors are needed to prevent the occurrence of long bone fractures in severely disabled patients. Furthermore, there has been no report on the association between the underlying diseases and fractures in patients with severe motor and intellectual disabilities, and further study is required. The current study has a number of limitations, including its retrospective single-center design, a limited number of cases, and the non-quantitative evaluation of bone fragility. These points should be considered in the design of further studies.

CONFLICTS OF INTEREST

The authors report no conflicts of interest.

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