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REGULAR ARTICLE

Bone fracture in physically disabled children attending schools for handicapped children in Japan

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Abstract
Objective Very few epidemiologic studies on bone fracture have been conducted in schools for handicapped children (*Yogo* schools). The aim of this study was to clarify the frequency and risk factors of bone fracture in physically disabled children in Japan.
Methods We used a cross-sectional design to examine 525 physically disabled children in 38 *Yogo* schools in the Hokuriku-Koshinetsu District of Japan. The questionnaire surveyed information on participant sex, age, level of physical disability, and bone fracture history. Information on fractures was obtained, including number of fractures over participant lifetime, age when fractures occurred, location, and cause. One-year-period prevalence and lifetime prevalence were defined as the proportion of subjects with incident fractures in the previous year and with a history of fracture, respectively.
Results Participant ages ranged from 6 to 15 years, and 66.3% had cerebral palsy (CP). The 1-year-period prevalence was 3.6% and lifetime fracture prevalence was 9.7%. The 1-year-period prevalence in the age groups of 6–9, 10–12, and 13–15 years was 2/184 (1.1%), 5/171 (2.9%), and 12/164 (7.3%), respectively (P for trend = 0.0031). There were no differences in period prevalence between sexes, and this was not associated with presence of CP. Multiple logistic regression analysis showed that age and presence of one joint contracture in the lower limbs or hip were independently associated with occurrence of bone fracture over participant lifetime.
Conclusions Physically disabled children are at high risk of bone fracture, and further risk factors should be determined.

Keywords Cerebral palsy · Disabled children · Epidemiology · Prevalence · Physical disability

Introduction

In 2006, an estimated 251,000 children, or 2% of the total population of that age in Japan, were reported to be attending classes meant for disabled children or special schools for such children, and this number is increasing [1]. Health and Welfare Statistics in Japan [1] reported that approximately 25% of disabled children have a physical disability, and health problems related to these disabilities appear to be an important issue in child health in schools.

One prevalent health problem in physically disabled children is bone fracture. Children with physical disabilities such as cerebral palsy (CP) have been known to have osteopenia [2] and low bone mineral density [3, 4]. This may be due in part to low or no mobility and to hypovitaminosis D caused by immobility [5].

When a child suffers from a fracture, he or she rests quietly in bed, which causes secondary disabilities such as joint contracture and disuse muscle atrophy [6], which

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leads to subsequent decreases in his or her quality of life. In addition, fractures and related matters could cause physical and mental distress not only to the children themselves but also to their parents, teachers or caregivers who experience stress from child care [7]. In this way, bone fractures in physically disabled children are considered a societal burden.

Studies on bone fracture in disabled children are rare. A few studies from the USA [8–10] have tried to clarify epidemiologic features of fractures in children with CP, and of those, only two were population based [8, 9]. No other epidemiologic studies exist on this issue, and more research is required.

The above background gave rise to our interest in the epidemiologic features of bone fracture in physically disabled Japanese children. The aim of this study was to clarify the frequency and risk factors of bone fracture in physically disabled children in *Yogo* schools (schools for handicapped children) in Japan.

Subjects and methods

Our cross-sectional design targeted 737 children, aged between 6 and 15 years, who attended classes for physically disabled children in 38 *Yogo* schools in the Hokuriku-Koshinetsu District in Japan, including the six prefectures of Toyama, Ishikawa, Fukui, Yamanashi, Nagano, and Niigata. Of the 38 *Yogo* schools targeted, 34 agreed to participate in our study. We sent an anonymous questionnaire through the schools to parents of all 737 children in the 34 schools. Of the 737 questionnaires, 541 (73.4%) were answered and returned to us through the schools. Fifteen questionnaires were incomplete, and one subject had osteogenesis imperfecta, a congenital collagen disorder characterized by easily fractured bones, and thus these subjects were excluded. Ultimately, data from 525 subjects were included in the statistical analysis. Questionnaires were distributed in December 2006, and completed questionnaires were collected in February 2007. Distribution and collection of questionnaires were done in a confidential manner. Informed consent was not obtained due to the anonymous nature of this study. The protocol for this study was approved by the Ethics Committee of Niigata University School of Medicine.

The questionnaire surveyed information on participant sex, age, level of physical disability, and fracture history. It also included items concerning activities of daily living (ADL). Levels of ADL were assessed by items pertaining to mobility, feeding, toilet use, and dressing. For each of these parameters, participants were categorized as 1 (totally dependent), 2 (partially dependent) or 3 (independent). Information on physical disability, including cause of motor dysfunction, location of current palsy, and joint contracture of the children, was obtained. The questionnaire asked parents to specify any and all locations of current palsy in the left upper limb, right upper limb, left lower limb, and right lower limb, and to specify any and all joint contractures of left wrist, right wrist, left elbow, right elbow, left shoulder, right shoulder, left ankle, right ankle, left knee, right knee, left hip, and right hip. Information on previous fractures, including age when fracture occurred, location, cause, and number of fractures in the previous year and earlier, was also obtained.

One-year-period prevalence was defined as the proportion of subjects with an incident fracture in the previous year. Lifetime prevalence was defined as the proportion of subjects with a history of fracture. Fractures caused by high-energy accidents, such as traffic accidents and falls from a place higher than a bed, were excluded from the statistical analysis. Since most of the children had had the physical disability since early in their lives, lifetime prevalence was used as the outcome variable in the association analysis. Lifetime fracture prevalences were compared against presence of possible risk factors and tested by the χ^2-test. A linear trend between age and lifetime prevalence was tested by simple logistic regression analysis. Multiple logistic regression analysis was used to determine independent risk factors associated with the subject suffering a fracture over their lifetime. Qualitative variables were treated as dummy variables in the multiple logistic regression analysis. The SAS statistical package (version 8.02, SAS Institute, Cary, NC, USA) was used for statistical analyses. P-value <0.05 was considered to indicate statistical significance.

Results

Subjects ranged in age from 6 to 15 years, with an average age of 10.9 years (standard deviation, SD 2.7 years). Subject characteristics are summarized in Table 1. More than 60% of subjects had a history of CP as a major disease that caused motor dysfunction. Quadriplegia was the predominant palsy, and about 70% of subjects were totally dependent on others for mobility on level surfaces.

Fracture histories revealed 85 fractures in 56 subjects. Of those, 21 fractures in 19 subjects had occurred in the previous year. Fracture locations are shown in Table 2. Fractures of the lower limbs occurred more often than those of the upper limbs. The most frequent fracture location was the upper legs, followed by lower legs and feet. Causes of fractures are also shown in Table 2. Most frequently, the cause of the fracture was unknown; the next most frequent cause of fracture was related to a fall.

Table 1 Characteristics of study subjects

	Number of subjects	
	Male ($n = 277$)	Female ($n = 248$)
Major disease causing motor dysfunction		
Cerebral palsy	189 (68.2)	159 (64.1)
Chromosomal anomaly	9 (3.2)	18 (7.3)
Traumatic brain injury	14 (5.1)	5 (2.0)
Encephalitis	11 (4.0)	7 (2.8)
Myopathies	12 (4.3)	2 (0.8)
Others	42 (15.2)	57 (23.0)
Current palsy		
Quadriplegia	166 (59.9)	128 (51.6)
Paraplegia	39 (14.1)	31 (12.5)
Hemiplegia	19 (6.9)	20 (8.1)
Triplegia	14 (5.1)	17 (6.9)
Monoplegia	12 (4.3)	12 (4.8)
Unknown	27 (9.7)	41 (16.5)
Mobility on level surfaces[a]		
Totally dependent	185 (66.8)	174 (70.2)
Partially dependent	50 (18.1)	43 (17.3)
Independent	39 (14.1)	29 (11.7)

Percentage values are given in parenthesis

[a] Data missing for 5 subjects

Table 2 Locations and causes of fractures

	Number of observations
Location of fracture	
Hands	7 (8.2)
Forearms (radius or ulna)	2 (2.4)
Upper arms (humerus)	10 (11.8)
Upper legs (femur)	17 (20.0)
Lower legs (tibia or fibula)	16 (18.8)
Feet	15 (17.7)
Others	18 (21.2)
Cause of fracture[a]	
Fall	12 (14.6)
Fall from bed or wheelchair	2 (2.4)
Fall from a place higher than a bed	3 (3.7)
Indoor bump	4 (4.9)
During dressing	5 (6.1)
During bathing	1 (1.2)
Unknown	27 (32.9)
Traffic accident	2 (2.4)
Others	26 (31.7)

Percentage values are given in parenthesis

[a] Data missing for 3 subjects

The 1-year and lifetime prevalence by sex, age, and CP status are shown in Table 3. Overall, there was no significant difference in the 1-year ($P = 0.1568$) or lifetime prevalence ($P = 0.3906$) between sexes. Prevalence was positively associated with age, and this trend was statistically significant. The difference in the 1-year and lifetime prevalences in subjects with and without CP was not significant.

The lifetime prevalence of fracture analyzed according to level of palsy, joint contracture, and ADL is shown in Table 4. Level of palsy was not significantly associated with lifetime prevalence of fracture. Subjects with one joint contracture in the lower limbs or hip had a significantly higher lifetime prevalence of fracture than other subjects. Levels of dependence in feeding and toilet use were significantly associated with lifetime prevalence of fracture; however, levels of dependence in mobility and dressing were not.

Multiple logistic regression analysis was performed to determine the independent variables associated with a history of fracture (Table 5). Significant predictor variables identified in bivariate analyses were considered as candidate variables for the multivariate model, where "joint contracture in the lower limbs or hip" was coded as 1 for one joint and 0 otherwise, and "feeding" and "toilet use" were coded as 1 for independent and 0 for dependent. After adjusting for age, having one joint contracture in the lower limbs or hip was found to be an independent variable associated with a history of fracture. When we performed another multiple logistic regression analysis, in which "joint contracture in the lower limbs or hip" was coded as 1 for one or more joint and 0 for none, age ($P = 0.0105$) was the only significant variable. Moreover, "joint contracture in the lower limbs or hip" ($P = 0.1180$), "feeding" ($P = 0.9244$), and "toilet use" ($P = 0.2982$) were insignificant.

Discussion

Physically disabled children are considered to be at high risk for bone fracture. It is known that various causes contribute to decreased bone mineralization and bone density in children with CP, such as nonambulatory status, immobilization after surgery, anticonvulsant therapy, and poor nutrition [10, 11]. However, there have been only a few epidemiologic studies worldwide on this subject [8, 9], and no such studies have been conducted in Japan. This study is the first description of epidemiologic features of bone fracture in physically disabled children in Japan, and reports several new findings.

Fracture occurrence as it relates to demographic factors in this population can be compared with that in the normal population, because the 1-year period prevalence of this study (defined as the proportion of subjects with an incident fracture in the previous year) is a good estimate of the

Table 3 One-year-period and lifetime prevalence of fracture in subjects by sex, age, and cerebral palsy status

	One-year-period prevalence[a]			Lifetime prevalence[b]		
	Male	Female	Total	Male	Female	Total
Age (years)[c]						
6–9	0/99 (0)	2/85 (2.4)	2/184 (1.1)	3/99 (3.0)	6/85 (7.1)	9/184 (4.9)
10–12	3/92 (3.3)	2/79 (2.5)	5/171 (2.9)	10/92 (10.9)	6/79 (7.6)	16/171 (9.4)
13–15	4/83 (4.8)	8/81 (9.9)	12/164 (7.3)	11/83 (13.3)	15/81 (18.5)	26/164 (15.9)
Total	7/277 (2.5)	12/248 (4.8)	19/525 (3.6)	24/274 (8.8)	27/245 (11.0)	51/525 (9.7)
			P for trend = 0.0031			P for trend = 0.0026
Cerebral palsy						
Present	5/189 (2.7)	6/159 (3.8)	11/348 (3.2)	15/189 (7.9)	17/159 (10.7)	32/348 (9.2)
Absent	2/88 (2.3)	6/89 (6.7)	8/177 (4.5)	9/88 (10.2)	10/89 (11.2)	19/177 (10.7)
			P = 0.4306			P = 0.5735

Five fractures caused by high-energy accidents, such as traffic accidents and falls from a place higher than a bed, were excluded. Percentage values are given in parenthesis

[a] Proportion of subjects with an incident fracture in the previous year

[b] Proportion of subjects with a history of fracture

[c] Data are missing for 6 subjects

incidence rate. Sugimori [12] studied healthy children, and reported that the rate of incidence of fracture (per 100 person-years) was 0.7–1.2 (boys) and 0.5–0.7 (girls) in lower-grade elementary students, 1.5–2.2 (boys) and 1.0–1.3 (girls) in upper-grade elementary students, and 2.2–3.1 (boys) and 0.4–0.9 (girls) in junior high school students. Arai et al. [13] reported that the rate of fracture incidence in elementary school children was 0.4 in boys and 0.3 in girls. Compared with these data, fracture occurrence was higher in our study population, especially in the 13–15 year age group. Children in this age group are relatively physically active, regardless of ADL levels, which may substantially increase their risk of fracture. Fracture prevention should be a priority, particularly in the 13–15 year age group, for physically disabled children. The present study also showed that fracture occurrence did not differ between children with and without CP, suggesting that these results can be generalized to all children with physical disabilities.

Increased risk of fracture in physically disabled children may be associated with their low bone mass. We did not assess bone mass of the subjects, but other researchers have shown that lower bone mass has been reported in children with CP [3, 4]. Likewise, low vitamin D status, which could adversely affect normal bone metabolism, has been reported in children with neuromuscular disorders [5, 14], and thus may also be associated with increased risk of fracture.

The incidence of fracture in boys has been reported to be approximately 2-fold higher than that in girls in healthy children [12, 15]. This finding is interpreted to mean that boys are more physically active than girls. In the present study, there was no significant difference between the sexes, a finding that is not consistent with the results from the general population. This may be explained by the fact that levels of physical activity are similar in both sexes in our study population.

There has been only one epidemiologic study on fracture incidence in physically disabled children. Stevenson et al. [8] reported the rate of fracture incidence in children with CP (mean age 9.3 years) in the USA to be 4.0 per 100 person-years. This value is similar to that obtained in the present study.

Lower limbs were the predominant fracture site (57%, including 20% in the femur) in our study; this is similar to results obtained in previous studies. Brunner and Doderlein [16] investigated 37 CP patients (mean age 12.0 years) who received treatment for fractures diagnosed by radiographs, and reported that 74% of fractures occurred in the femur. Presedo et al. [11] investigated 156 CP patients (aged between 1 and 21 years) who received treatment for fractures diagnosed by radiographs, and reported that 82% of fractures were in the lower limbs. In a population-based setting, Leet et al. [9] investigated fracture histories obtained from medical charts of 763 CP patients (aged between 1.3 and 18 years), and found that 70% of fractures involved the lower extremities. These results are consistent with the finding that bone mineral density (BMD) in the femur is severely diminished in children with moderate to severe CP [3]. Shiragaki et al. [17] also showed that BMD in the lower limbs was lower than other bone sites in

Table 4 Lifetime prevalence of fracture according to levels of palsy, joint contracture, and activities of daily living

Palsy	Lifetime prevalence	χ^2-test ($df = 2$)
Quadriplegia		
None	24/231 (10.4)	$P = 0.6433$
Present	27/294 (9.2)	
Palsy of the upper limbs		
None	15/143 (10.5)	$P = 0.7282$
One arm	9/78 (11.5)	
Both arms	27/304 (8.9)	
Palsy of the lower limbs		
None	7/82 (8.5)	$P = 0.5252$
One leg	8/58 (13.8)	
Both legs	36/385 (9.4)	
Number of joint contractures in the upper limbs		
0	34/382 (8.9)	$P = 0.5705$
1	7/60 (11.7)	
≥2	10/82 (12.2)	
Number of joint contractures in the lower limbs or hip		
0	22/292 (7.5)	$P = 0.0274$
1	9/45 (20.0)	
≥2	20/188 (10.6)	
Mobility on level surfaces		
Totally dependent	35/359 (9.8)	$P = 0.9262$
Partially dependent	8/93 (8.6)	
Independent	7/68 (10.3)	
Feeding		
Totally dependent	31/295 (10.5)	$P = 0.0328$
Partially dependent	6/139 (4.3)	
Independent	12/85 (14.1)	
Toilet use		
Totally dependent	36/366 (9.8)	$P = 0.0180$
Partially dependent	5/108 (4.6)	
Independent	9/47 (19.2)	
Dressing		
Totally dependent	35/357 (9.8)	$P = 0.4475$
Partially dependent	9/121 (7.4)	
Independent	6/43 (14.0)	

Five fractures caused by high-energy accidents, such as traffic accidents and falls from a place higher than a bed, were excluded. Percentage values are given in parenthesis

Japanese children with CP. All of these findings suggest that prevention of limb fractures is of primary importance in physically disabled children.

The causes of fracture were varied, and most frequently (32.9%) the cause was unknown; this has not been observed in normal children [10]. In a previous study, Presedo et al. [9] also observed that causes were unknown for 45% of fractures occurring in CP children. These findings demonstrate that some physically disabled children may have very fragile bones.

The present study shows that having one joint contracture in the lower limbs or hip is an independent risk factor for fracture in physically disabled children. This finding was consistent with that of Leet et al. [9] indicating a significant association between ankle contracture and lower-leg fracture. In general, the presence of joint contracture makes it difficult to care for the child during transfer, dressing, and toilet use. In addition, contracture stiffens the joint, which may then fail to absorb external force to the bones. These mechanisms may explain our finding. It is unknown why one joint contracture in the lower limbs or hip, but not more than one, was associated with fracture. We conjecture that subjects with only one joint contracture in the lower limbs or hips may be more likely to fall than those without such a contracture (due to better balance) and those with multiple joint contractures (due to the fact that they receive better care). Previous studies have suggested that physical factors other than joint contracture, such as quadriplegia and immobility, are risk factors for fracture in physically disabled children. Although we found only joint contracture as a factor associated with fracture, other risk factors should be clarified in future studies.

Some basic characteristics of our population are unique to the Hokuriku-Koshinetsu District, and may not necessarily exist in the general Japanese population. According to Data on Special Support Education, 2006 [18], the number of students (6–15 years old) in *Yogo* schools, relative to total students of the same age, in this area was 602/756,258 (0.08%), which is slightly lower than that of the Japanese population (12,160/10,823,873, 0.11%). Thus, the actual number of physically disable children in the Hokuriku-Koshinetsu District may be lower than the Japanese

Table 5 Results of multiple logistic regression analysis with presence of fracture history as a dependent variable

Candidate independent variables	Odds ratio	95% Confidence interval	P-value
Joint contracture in the lower limbs or hip (1, one joint; 0, others)	2.29	1.02–5.17	0.0456
Feeding (1, independent; 0; dependent)	1.06	0.39–2.88	0.9121
Toilet use (1, independent; 0; dependent)	1.79	0.57–5.61	0.3183
Age (years)	1.17	1.04–1.31	0.0100

Five fractures caused by high-energy accidents, such as traffic accidents and falls from a place higher than a bed, were excluded average. No evidence was found to suggest that the lifestyles of physically disabled children in the Hokuriku-Koshinetsu District were significantly different from those of physically disabled children in other districts. Nutritionally, children in the Hokuriku-Koshinetsu District may have lower levels of vitamin D, due to fewer hours of sunshine in the autumn and winter months, relative to children in other regions. As such, their bone density may be lower. These factors should be considered if these results are extrapolated or generalized to the Japanese population.

While previous studies targeted children with CP or other specific neuromuscular diseases, we studied children with many physical disabilities in *Yogo* schools. Therefore, results obtained from this study can be applied not only to CP children, but to all school children with physical disabilities. However, this study had some limitations. First, we obtained fracture data retrospectively by questionnaire. Underreporting of fractures tends to be a prevalent problem with this method, so the fracture prevalence may have been underestimated. Secondly, we used prevalent fracture (not incident fracture) in the association analyses, and thus were unable to assess relative risks. In addition, this type of study does not assure a causal relationship. A prospective study should be conducted to remedy these limitations. Finally, the sample size in this study was not sufficiently large; a study with a larger sample size could provide further evidence on the risk factors for fracture.

This is the first study focusing on epidemiologic features of fracture in physically disabled children in Japan, and shows that (1) physically disabled children, especially in the 13–15 year age group, have a higher risk of fracture than the normal population, and (2) the presence of a joint contracture in the lower part of the body is an independent risk factor for fracture. Consequently, environmental hazards that could cause fractures should be removed, especially for older students, and prevention of joint contracture from early stage of life should be a main focus. We therefore conclude that children with a physical disability are at higher risk of bone fracture, and further risk factors for this should be determined. The results of our study should be useful in a future fracture prevention program in schools for handicapped children.

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