

Awareness-Raising Activities to Identify Children with Short Stature

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Background: Although short stature is sometimes treatable in children, family members do not always realize that their children have short stature. To develop better educational materials for identifying short stature, we conducted a questionnaire survey on children with short stature. Using the results of the survey, we revised educational activities regarding short stature.

Methods: To assess the effectiveness of the revised activities, we examined changes in the numbers of consultations before and after the changes to the educational activities, the height of children examined after such changes, the test implementation rate, and the test results.

Results: After the start of direct promotion for school nursing staff in 2015, the number of outpatients with short stature who visited the hospital significantly increased (16.1/year before 2014 vs. 68.8/year after 2015; $p = 0.02$). The number of patients hospitalized for a growth hormone secretion stimulation test also significantly increased, from 9.3/year before 2014 to 47.0/year after 2015 ($p = 0.02$). However, 35% of families did not want to subject their child to a growth hormone stimulating test, even if their child was extremely short.

Conclusions: Our revised educational activities for short stature among school nursing staff, school physicians, and nurses at health centers were more effective than conventional activities consisting of public relations magazines and lectures for the general public. It is important to provide proper explanations to enable a better understanding of hormone therapy. (J Nippon Med Sch 2024; 91: 410–416)

Key words: short stature, promotion activity, school nursing teacher, school physician, school nurse

Introduction

Although some cases of short stature are treatable during childhood, family members sometimes do not realize that their children have short stature, that short stature can be treatable, and that, in cases of short stature caused by growth hormone deficiency, growth hormone treatment cannot be given after closure of the epiphyseal line. At our institution, we have experienced many cases in which treatment was not possible because consultations were delayed until after epiphyseal closure.

Since 2002, the Department of Pediatrics at Nippon Medical School Chiba Hokusoh Hospital has been actively raising awareness of short stature through activities such as lectures for the general public, publications in preschool magazines, and educational materials for school nurses and practitioners¹. However, these efforts have not been satisfactory, and consultations thus con-

tinue to be held after epiphyseal closure for children with short stature. To date, there have been few reports on educational activities regarding short stature^{2–5}.

To develop educational materials that are better in identifying short stature, we conducted a questionnaire survey that targeted children with short stature and their guardians and aimed to increase awareness of short stature and referrals to medical clinics for treatment⁶. The survey results revealed that the rate of children with short stature (>2.0 SD below the mean) was 90% among those who learned about short stature from a school nursing teacher and 100% among those who learned about short stature from a public health nurse. These results indicate that a high percentage of children with short stature had been referred by a school nursing teacher or public health nurse.

Physical measurements are actually taken and recorded

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https://doi.org/10.1272/jnms.JNMS.2024_91-411
Journal Website (<https://www.nms.ac.jp/sh/jnms/>)

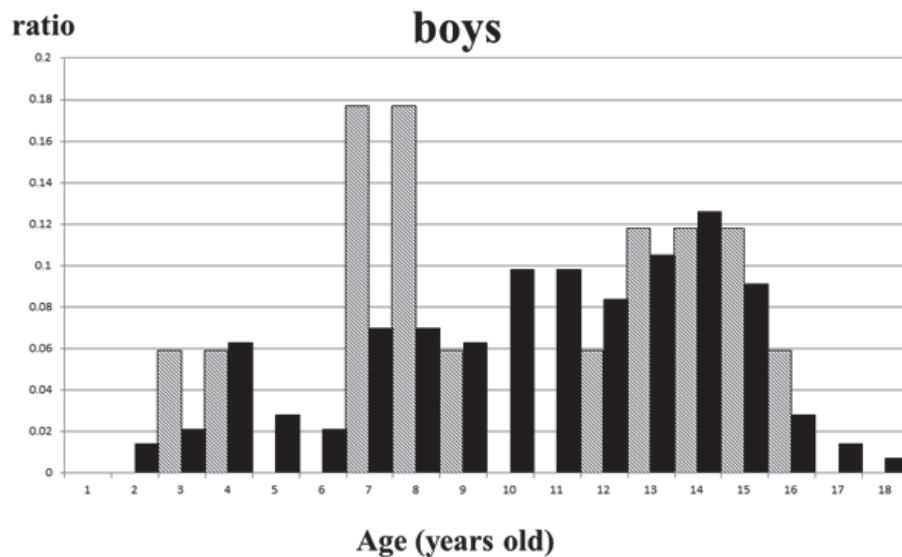


Fig. 1a Age distribution ratio of referral of boys

Before revision of the materials, the age of referred boys was concentrated during the period before and after the development of secondary sexual characteristics and upon entering elementary and junior high school. However, after the revision of the materials, the age of referral for boys with short stature was evenly distributed, although there was no significant difference in age distribution using the two-sample Kolmogorov-Smirnov Test ($p=0.21$). In particular, the number of referrals increased around the time before the development of secondary sexual characteristics, with the highest peak around the time of entrance to junior high school, and the second highest peak after a health checkup at age 3 years.

The ratio was calculated as number of referrals divided by the total number of referrals. Referrals included all referrals for which the main complaint was short stature. Grey bars represent the number of referrals of boys before 2014; black bars represent the number of referrals of boys after 2015.

several times a year in elementary schools, junior high schools, preschools, and nursing homes, so we suspected that short stature could be identified with certainty by school nursing staff and public health nurses. In 2015, PC software (child health management program) created for school nurses was distributed to schools across Japan. Furthermore, since 2016, the revised Japan School Health and Safety Law has required measurement and recording of physical data and interpretation of the values, for more effective utilization⁷. These actions might improve awareness of short stature among school nursing staff. In addition, the subsidy system for pediatric medical expenses in each local government was enhanced, and hurdles for consultation and treatment for short stature were lowered.

Thus, we decided to revise educational activities regarding short stature by using the results of a questionnaire survey. We terminated activities such as lectures for the general public and sending of materials and started conducting face-to-face workshops for school nursing staff and public health nurses. In addition, we held

workshops approximately twice a year and asked the local board of education to hold a 2-hour presentation and a question-and-answer session for school nursing staff at elementary and junior high schools, during which we explained how to interpret the height curve, possible medical conditions and their characteristics, and the consequences if such conditions are left untreated. In this study, we examined changes in the numbers of consultations before and after revising the educational activities, the height of children examined after such changes, the test implementation rate, and the test results.

Methods

We retrospectively examined the subsequent consultation status, the height of children examined after the changes, the test implementation rate, test results, numbers of outpatients with short stature, hospitalization examinations for growth hormone stimulating testing, and children starting hormone therapy before and after changing educational activities from medical charts. This study was approved (no. 588) by the Ethics Committee of Nippon

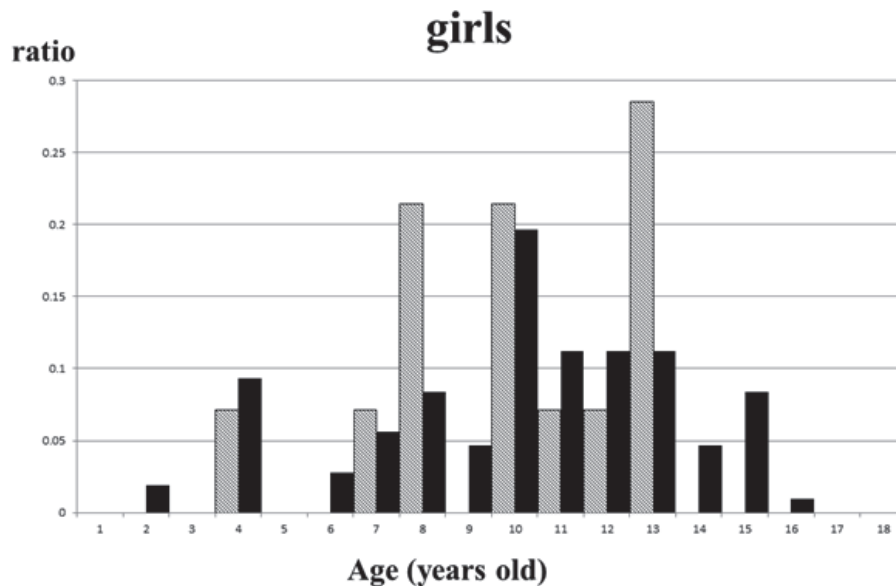


Fig. 1b Age distribution ratio of referral of girls

Before revision of the materials, the age of referred girls was concentrated during the period of entrance to elementary and junior high school and around the time of the development of secondary sexual characteristics. The most frequent age of referral for girls was around the age of 10 years, although there was no significant difference in age distribution using the two-sample Kolmogorov-Smirnov Test ($p=0.33$).

The ratio was calculated as number of referrals divided by the total number of referrals. Referrals included all referrals for which the main complaint was short stature. Grey bars represent the number of referrals of girls before 2014; black bars represent the number of referrals of girls after 2015.

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Statistical Analysis

The Mann-Whitney U-test was used to determine the significance of differences between groups. The chi-square test for independence or Fisher exact probability test was used for comparisons within groups. The software package Kaleida-Graph (Synergy Software, Reading, PA, USA) was used for correlation coefficient analysis. Histograms (Fig. 1) were analyzed with the two-sample Kolmogorov-Smirnov test.

Results

Numbers of Outpatients, Hospitalization Examinations, and Children Starting Hormone Therapy after the Start of the Direct Promotion for School Nursing Staff (Table 1)

After the start of direct promotion for school nursing staff, in 2015, the number of outpatients with short stature who visited the hospital significantly increased (16.1/year before 2014 vs. 68.8/year after 2015; $p < 0.01$). The percentage of patients with short stature who came directly to our hospital was 7.9% before the change in awareness activities and 2.7% after the change, but the

difference was not significant. The number of patients hospitalized for a growth hormone secretion stimulation test significantly increased, from 9.3/year before 2014 to 47.0/year after 2015 ($p < 0.01$), as did the number of patients undergoing growth hormone therapy, from 6.0/year before 2014 vs. 22.8/year after 2015 ($p < 0.01$). These findings indicate an increase in the number of patients eligible for hospitalization for growth hormone secretion stimulation tests. The ratio of patients eligible for hormone therapy to those hospitalized for a growth hormone secretion stimulation test was 63% before 2014 vs. 48% after 2015 ($p = 0.04$), and the ratio of outpatients who received hormone therapy to those presenting for treatment of short stature per year was 35% before 2014 vs. 33% after 2015 ($p = 0.60$). We did not investigate the change in the number of patients with short stature who may have been delayed in consulting a doctor.

Changes in Triggers for Consultation

After the promotion of specialized information for school nursing staff, most cases were referrals from the school principal, school nursing staff, or family doctor. Before the revision of the materials, boys were usually referred before or after the development of secondary sex-

Table 1 Numbers of outpatients, hospitalization examinations, and children starting hormone therapy after the start of direct promotion for school nursing staff

year	1994-2014		2015-2019	
	Mean Median (Mann-Whitney U-test)	Standard deviation	Mean Median	Standard deviation
Number of outpatients (per year)	16.1 12 ($p<0.01$)	15.9	68.8 61	26.1
Number of inpatients for examination (per year)	9.3 5 ($p<0.01$)	10.5	47.0 36	17.6
Number of children undergoing de novo growth hormone therapy (per year)	5.6 3 ($p<0.01$)	5.9	22.8 17	9.1
Number of inpatients for examination/number of outpatients	52.0% 50.0% ($p=0.19$)	22.0%	69.0% 66.4%	27.0%
Number of children undergoing de novo growth hormone therapy/number of outpatients	35.0% 33.3% ($p=0.60$)	21.0%	33.0% 31.8%	6.0%

After the start of direct promotion for school nursing staff in 2015, the number of outpatients with short stature who visited the hospital increased, as did the number of patients hospitalized for growth hormone secretion stimulation testing. Number of outpatients includes new and returning patients.

ual characteristics and upon entering elementary and junior high school (Fig. 1a). However, after the revisions, age at referral for boys with short stature was evenly distributed. In particular, there was an increase in referrals during the period before the development of secondary sexual characteristics. The highest peak was around the time of entrance to junior high school, and the second highest peak was after a health checkup at age 3 years, although the two-sample Kolmogorov-Smirnov Test showed no significant difference in age distribution ($p=0.21$).

Before dissemination of the revised materials, girls were usually referred around the time of entrance to elementary and junior high school and around the time of the development of secondary sexual characteristics. The most frequent age of referral among girls was approximately 10 years, although the two-sample Kolmogorov-Smirnov Test showed no significant difference in age distribution ($p=0.33$)(Fig. 1b). Because a longer duration of treatment for short stature might result in a better outcome, the younger age at consultation after the materials revision was considered a positive change in terms of ensuring better treatment for short stature.

Distribution of Height Growth SDs among Children Referred for a Checkup after the Start of the Direct Promotion for School Nursing Staff and Nurses

A total of 127 patients attended consultations, and 91 had a height SD of -2.0 or less (71.7%). In addition, six children had height growth of -1.5 SD or less over 2 years, even though the SD values were larger than -2.0 . We performed a growth hormone secretion stimulation test in all six cases, and growth hormone deficiency (GHD) was diagnosed in three. Precocious puberty was diagnosed in five children. The height of 10 children measured at our hospital ranged from -1.8 to -2.0 SD. Of the 91 children with an SD less than -2.0 , a growth hormone secretion stimulation test was performed in 52 (57.1%), 21 of whom had GHD and four were small for gestational age (SGA).

Distribution of Height SDs among Children Referred by a Family Doctor after Revision of the Materials

In total, 112 children were referred from family doctors, among whom, 63 (56.2%) had a height of -2.0 SD or less. Among 49 children with a height greater than -2.0 SD, eight with height growth less than -1.5 SD over 2 years and five with GHD underwent growth hormone stimulation testing. Three children with SGA, as indicated by birth records, were deemed ineligible for growth hormone treatment because their current height was

Table 2 Diagnoses by height

Body height (Standard deviation)	number of boys	number of girls	Number of inpatients for examination	diagnosed as GHD	diagnosed as SGA	diagnosed as precocious puberty
<-3.00 SD	6	5	10	6	2	0
-2.50 SD~-3.00 SD	24	22	28	8	3	0
-2.00 SD~-2.50 SD	51	51	65	29	0	0
-1.50 SD~-2.00 SD	34	24	16	9	0	0
>-1.00 SD	10	9	0	0	0	6

GHD: growth hormone deficiency

SGA: small for gestational age

greater than -2.5 SD. Moreover, 47 (74.6%) of 63 children with a height of -2.0 SD or less underwent a growth hormone stimulation test: 18 were diagnosed as having GHD and one as having SGA.

Diagnosis by Height (Table 2)

Height at first visit of -3.00 SD or less

Among 11 children (6 boys, 5 girls) with a height at first visit of -3.00 SD or less, 10 were examined. Six were diagnosed with GHD and two with SGA. One patient did not attend the examination even after being informed of his short stature. No other cause of short stature, such as inherited bone disease or other underlying disease, was identified.

Height at first visit of -2.50 to -3.00 SD

Among 46 children (24 boys, 22 girls) with a height at first visit of -2.50 to -3.00 SD, 28 were examined. Eight were diagnosed with GHD and three with SGA. Eighteen children were not tested, for the following reasons: the child's developmental condition (5 cases), age of referral younger than 3 years (5 cases), parental refusal (4 cases), closure of the epiphyseal line (2 cases), pending examination (1 case), and unknown (1 case).

Height at first visit of -2.00 to -2.50 SD

Among 102 patients (51 boys and 51 girls) with a height at first visit of -2.00 to -2.50 SD, 65 were examined and 29 were diagnosed as having GHD. In total, 37 children were not tested, for the following reasons: refusal by the patient (30 cases), closure of the epiphyseal line (6 cases), and age of referral younger than 3 years (1 case).

Height at first visit of -1.50 to -2.00 SD

Among 58 children (34 boys, 24 girls) with a height at first visit of -1.50 to -2.00 SD, examinations were performed on 16 (all of whom had height growth less than -1.5 SD), nine of whom were diagnosed as having GHD. The reasons for not performing an examination were as follows: the physicians decided on follow-up only (32

cases), refusal by the patient (7 cases), SGA diagnosed but treatment not indicated (3 cases), and closure of the epiphyseal line (1 case).

Greater than -1.0 SD

Among 19 children (10 boys, 9 girls) with a height at first visit greater than -1.0 SD, six (5 girls, 1 boy) had precocious puberty. Precocious puberty was recognized as early growth spurts by school nurses.

Discussion

Detecting unnoticed treatable short stature is an important task for pediatricians. However, parents, school nursing staff, and school physicians are not well aware that short stature in children can be treated and that the treatment window is limited. In addition, consultations for short stature often take place after closure of the epiphyseal line. We focused on educational activities regarding children with short stature that targeted school nursing staff and public health nurses with access to the children's height data. We hypothesized that this process would identify more children with short stature and enable timelier treatment at medical institutions.

Since 2015, we have been focusing on direct educational activities for school nursing staff in our area. As compared with previous educational activities, the present educational program has significantly increased the numbers of outpatients complaining of short stature, hospitalizations for testing to stimulate growth hormone secretion, and children starting growth hormone treatment.

To date, no systematic study has attempted to identify children with short stature in need of medical treatment. The present study—the first to use the School Health and Safety Law Enforcement Regulations⁷—revealed that the use of revised educational activities increased the number of referrals of short stature children. However, several problems emerged.

First, 56 (35.2%) of 159 children were not examined,

even if their height was less than -2.0 SD. When we asked families the reason for refusal, they responded that they were most concerned about the practice of administering hormonal drugs to their children via daily injection for many years. Parents and children have different perceptions of short stature, so care is needed by medical professionals when emphasizing the disadvantages of short stature, as this may lead to discrimination. In the future, we believe that academic societies will need to distribute guidelines regarding appropriate explanations of short stature and the need for psychological follow-up.

Second, 11 of 16 children who were examined because their annual height growth was less than -1.5 SD, even though their current height was greater than -2.0 SD, had GHD. It is critical to determine whether children with short stature are pathologically short based on their height at one point. However, to determine height growth, family members need to provide accurate historical data on growth, which is somewhat complicated. In the future, when individual medical data are electronically aggregated in Japan, it will be important to incorporate physical measurement data at schools, preschools, and nursing homes. Some schools lack physicians familiar with children's growth. In such cases, schools need to build a system of cooperation with related organizations, such as boards of education, medical associations, care hospitals, specialist associations, and school health and lifelong health organizations.

Third, 19 (50%) of 38 children with a height less than -2.5 SD were not diagnosed as having GHD or SGA. Recombinant growth hormone injection is the only medical treatment for short stature currently covered by Japan's national health insurance system. Only a few studies have investigated the pathology of short stature, such as dysfunction of growth hormone receptors due to genetic abnormalities, and little work has been done on the cause of GHD. Future studies should attempt to elucidate the genes and single-nucleotide polymorphisms responsible through comprehensive genetic analyses of children with short stature in genome-wide association studies. In addition, individualized genetic treatments need to be developed.

Conclusion

Our revised educational activities for school nursing staff, school physicians, and nurses at health centers were more effective in addressing short stature than were conventional activities consisting of public relations magazines and lectures for the public. Because 35% of families

declined growth hormone stimulating testing, even when their child was extremely short, it is important to provide explanations that improve understanding of hormone therapy. In addition, because many children with a height growth of -1.5 SD or less were diagnosed as having GHD, even if their current height was not less than -2.0 SD, the need to collect longitudinal height data in future educational activities should be emphasized.

Author contributions: MT collected the patient data. TA performed the statistical analysis. MT wrote the first draft of the manuscript, and TA and MT wrote the subsequent drafts. All authors reviewed, edited, and approved the final version of the paper.

Acknowledgements: The authors sincerely thank all members of the Department of Pediatrics of the Nippon Medical School Chiba Hokusoh Hospital for their contribution to this work. This study was supported in part by grants from the Promotion and Mutual Aid Corporation for Private Schools of Japan and the Science Research Promotion Fund.

Conflict of Interest: The authors declare no conflict of interest.

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(Received, April 23, 2024)

(Accepted, May 20, 2024)

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