

Original**Clinical Statistics for Dysphagia Patients ≤ 18 Years of Age in the Center of Special Needs Dentistry, April 2012–March 2013**

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Abstract : In April 2012, the Center of Special Needs Dentistry (SND) was established at Showa University Dental Hospital to provide function training for children with eating and swallowing disorders. A statistical clinical assessment was performed on new patients ≤ 18 years of age who visited the Center over a 1-year period (April 2012–March 2013) to assess the conditions present at the initial visit. In all, 60 patients (29 boys, 31 girls, mean (\pm SD) age 4.2 ± 4.1 years, range 0-18 years of age) were included in the study. Most patients were < 1 year of age (32%) and most came from one of four cities in the Johnan area (Shinagawa City, Meguro City, Ota City and Setagaya City). The most common primary diseases at the initial visit were cerebral palsy and cleft lip and palate. The third largest patient group was of healthy children with oral function problem. Over 60% of patients attended the Center of SND because of an eating-related complaint. More than 50% of patients were obtaining nutrients via oral intake; the remaining patients were obtaining nutrients via non-oral or a combination of oral and non-oral intake. Because of the young age of the patients and the fact that most were from neighboring areas, it can be inferred that effective community health care is being provided. It is necessary for the Center of SND to continue to provide professional treatment for dysphagia and to contribute to community medicine.

Key words : clinical statistics, the Center of Special Needs Dentistry, dysphagia, oral intake, dysphagia treatment

Introduction

Based on the findings of a survey conducted in 2011 by the Ministry of Health, Labor, and Welfare¹⁾, it is estimated that there are approximately 72,700 disabled children under 18 years

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of age in Japan. Approximately 3.79 million people over 18 years of age are disabled, and this number is increasing annually. According to the 2011 survey¹⁾, an estimated 740,000 people have an intellectual disability. Dentists and healthcare workers involved in dental treatment provide special needs support to a wide range of children and elderly people with disabilities²⁾. To provide treatment for children with disabilities, as well as other patients with special needs, the Center of Special Needs Dentistry (SND) was established in Showa University Dental Hospital in April 2012.

Many disabled children with cerebral palsy and mental retardation have problems swallowing. Recently, the number of dentists capable of providing eating and swallowing training to children with dysphagia has increased. A growing number of medical institutions have specialized dental outpatient clinics that provide this service. In the center of SND, functional eating training for children with dysphagia is provided in the Dental Treatment Unit. The present study was a statistical clinical investigation performed to assess the eating and swallowing training experienced by the patients who visited the Center, and to evaluate the status of this training.

Materials and methods

The total number of outpatients with dysphagia who visited the Center over a 1-year period (April 2012–March 2013) was determined. The following information was collected from the medical records of new patients aged ≤ 18 years attending the Center over this 1-year period: age, geographic origin, referring facility, primary disease, reason for attending the Center of SND, method of nutritional intake (oral intake, oral intake plus tube feeding, or tube feeding), and eating and swallowing function.

The university dental hospital in which the Center is located (in the Johnan area, encompassing Shinagawa City, Ota City, Meguro City and Setagaya City) is an advanced medical facility that provides services to the local community. The geographic origins of patients attending the Center over the study period were classified as either one of the four cities in the Johnan area; one of the other 23 cities in Tokyo, Yokohama City, and Kawasaki City (Kanagawa Prefecture); or “other” regions.

Referring facilities were classified as either the University’s medical and dental hospital, medical facilities outside the University, the Ryoiku Center (a treatment and rehabilitation center for children with disabilities), health and welfare institutions in the local community, or special needs schools. Eating and swallowing functions were assessed and patients were categorized into three groups: (i) those with swallowing problems; (ii) those with oral function problems; and (iii) those with coordination problems of the hand and mouth.

Results

Number of patients

Over the study period, 861 patients visited the Center for treatment (68 for the first time and 793 for follow-up visits). Of the 68 first-time patients, 60 were in the developmental stage of life (0–18 years of age; 29 boys, 31 girls; male :female ratio 1 : 1). Analysis of patient numbers

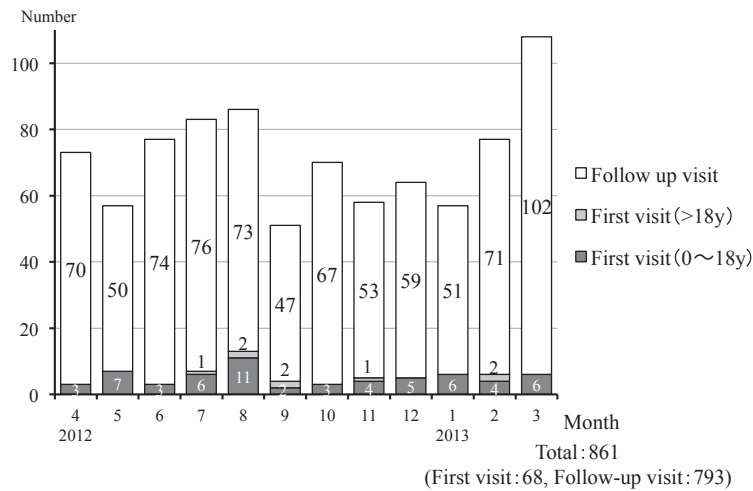


Fig. 1. Number of patients attending the Center of Special Needs Dentistry each month, April 2012–March 2013.

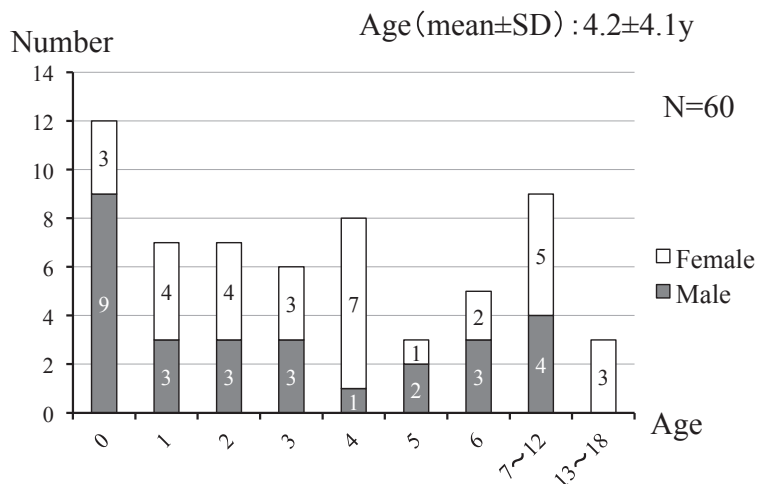


Fig. 2. Age distribution of patients ≤18 years of age attending the Center of Special Needs Dentistry for the first time between April 2012 and March 2013.

by month revealed that the highest number of first-time and follow-up visits occurred in August and March, respectively (Fig. 1).

Patient age

The mean (\pm SD) age of the 60 patients in the 0–18 years age group who visited the Center for the first time was 4.2 ± 4.1 years. Most patients were < 1 year of age (Fig. 2).

Geographic origins and referral facilities

Most patients ($n = 17$) were from one of the four cities in the Johnan area (Shinagawa City,

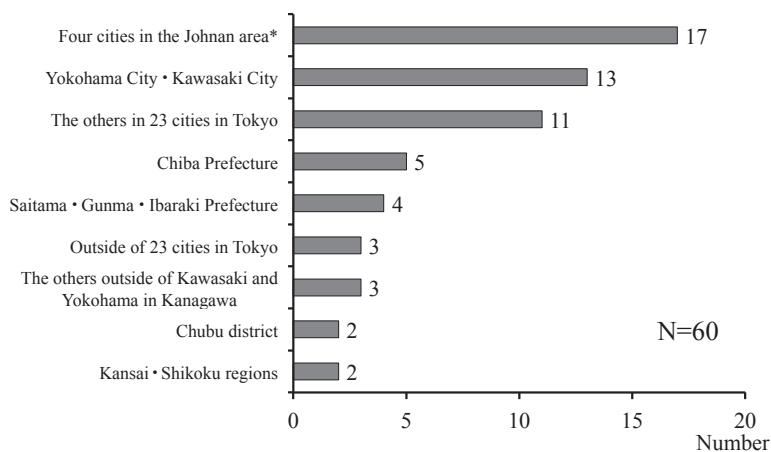


Fig. 3. Geographic origin of patients ≤ 18 years of age attending the Center of Special Needs Dentistry for the first time between April 2012 and March 2013. *The four cities in the Johnan area are Shinagawa City, Ota City, Meguro City and Setagaya City.

Meguro City, Ota City and Setagaya City), followed by Kawasaki City and Yokohama City (Fig. 3). Some of the patients attending the Center came from distant locations, such as the Kansai and Shikoku regions (Fig. 3).

Forty patients had a referral letter. Patients were referred by medical facilities, such as other university hospitals and general hospitals, as well as treatment and rehabilitation centers (Fig. 4). One-third of the referrals were from the Showa University Medical and Dental Hospital (Fig. 4).

Primary disease

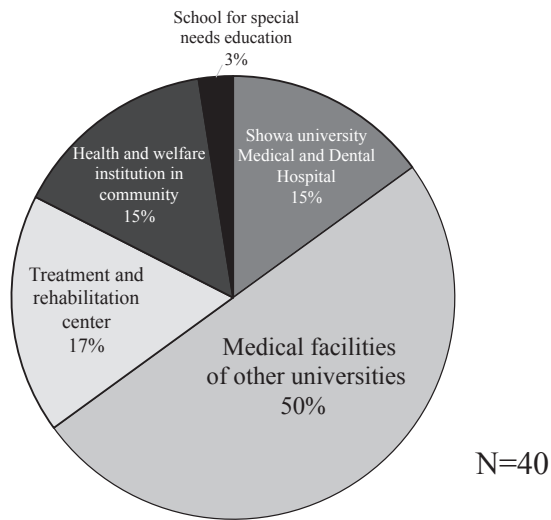
As indicated in Table 1, cerebral palsy and cleft lip and palate were the most common primary diseases. However, patients presented with a wide variety of disorders, including central nervous and muscular system disorders and anatomical impairments. Disorders of the central nervous and muscular systems included Angelman syndrome, Lowe syndrome, and Pitt-Hopkins syndrome. Some patients presented with α -thalassemia mental retardation syndrome, Pallister-Killian syndrome, or CHARGE syndrome. Six children were healthy.

Reason for attending the Center of SND

As shown in Fig. 5, 60% of patients attended the Center because of complaints regarding eating (e.g. requests for eating training). The second most common reason for attending the Center was to obtain a Hotz plate. Other reasons included issues related to malocclusion and requests for a second opinion (Fig. 5).

Nutritional intake

More than half the patients obtained their nutrition via oral intake (Fig. 6). The remaining patients were using oral intake plus tube feeding or tube feeding alone (neogastric feeding tube



The patients only for letter of introduction

Fig. 4. Referring facilities for the 40 patients aged ≤18 years attending the Center of Special Needs Dentistry for the first time between April 2012 and March 2013 who had a letter of referral.

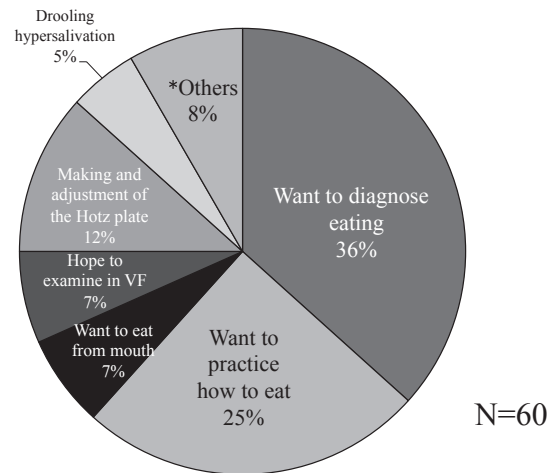


Fig. 5. Reasons for attending the Center of Special Needs Dentistry.
*Note, "Others" includes wanting a second opinion and concerns about malocclusion. VF, video fluoroscopy.

Table 1. Primary disease in the 60 children aged ≤18 years attending the Center of Special Needs Dentistry for the first time between April 2012 and March 2013.

N = 60			
Disease name	N	Disease name	N
Cerebral palsy	8	CHARGE association	1
Cleft lip and palate/Cleft palate	8	Congenital esophageal atresia deficiency	1
Normal child	6	Congenital nasopharyngeal atresia dysfunction	1
Mental retardation	5	Hydrocephalus	1
Down syndrome	4	Lissencephaly	1
Epilepsy	4	Lowe syndrome	1
Chromosome abnormality	3	Microcephaly	1
Pierre Robin syndrome	3	Myotonia congenital decrease	1
Angelman syndrome	2	Nasopharyngeal • Oral atresia dysfunction	1
Hypoxic encephalopathy	2	Pallister-Killian syndrome	1
Ankyloglossia	1	Pitt-Hopkins syndrome	1
ATR-X syndrome	1	Premature baby	1

CHARGE, coloboma, heart defect, atresia choanae, retarded growth and development, genital abnormality, and ear abnormality; ATR-X syndrome, α-thalassemia X-linked intellectual disability.

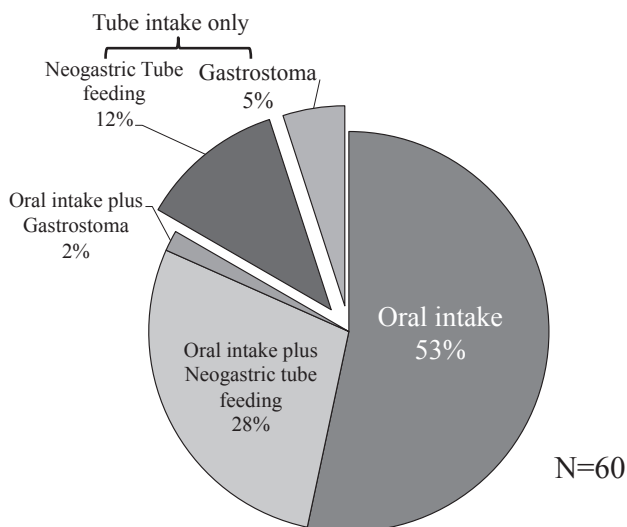


Fig. 6. Method of nutritional intake for the 60 patients aged ≤ 18 years attending the Center of Special Needs Dentistry for the first time between April 2012 and March 2013.

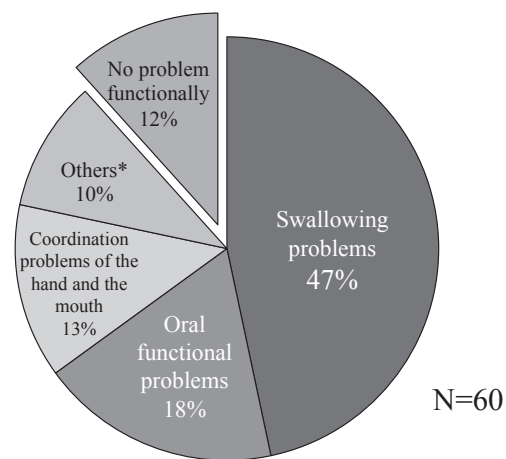


Fig. 7. Eating and swallowing diagnoses for the 60 patients aged ≤ 18 years attending the Center of Special Needs Dentistry for the first time between April 2012 and March 2013.
*“Others” includes problems breastfeeding, suspected esophageal stage disorder, and low saliva secretion.

and gastrostoma).

Eating and swallowing function

Almost half the patients in the study had problems related to swallowing function (Fig. 7). Issues with oral function and coordination problems of the hand and mouth were the second most common problems. Other problems included difficulties with breastfeeding prior to starting solid foods, suspected esophageal stage disorders, and low saliva secretion (Fig. 7).

Discussion

Established in April 2012 for dental patients with disabilities, the Center of SND not only provides treatment for patients with disabilities, but also provides eating training. The two most common primary diseases observed in children who visited the Center over the period April 2012-March 2013 were cerebral palsy and cleft lip and palate. Neonates with a cleft lip and palate exhibit problems breastfeeding immediately after birth^{3,4}. Infants with impaired breastfeeding ability often gain weight very slowly. The cleft lip and palate prevent proper sucking because they prevent the development of negative pressure, even when the gingiva is pressed on the nipple⁴. The use of a Hotz plate separates the oral and nasal cavities, allowing the neonate to press his or her tongue on the nipple and nurse⁴. Many parents attended the Center to request insertion of a Hotz plate in neonates shortly after birth.

Cerebral palsy was equally the most common primary disease in children attending the Center.

Dysphagia is one of the multiple functional disorders associated with central nervous system disorders in children with cerebral palsy or intellectual disabilities^{5,6}, and can be due to anatomical abnormalities, abnormal neurological maturation, sensory disturbance of the oral cavity, or esophageal motility disorders. Dysphagia results in weight loss, impaired growth, aspiration pneumonia due to recurrent aspiration, and lower respiratory tract infections⁷⁻¹¹. To prevent these complications, medical interviews are conducted at the Center to confirm eating status, appropriate examinations are performed (e.g. video fluoroscopy or video endoscopy), and instructions are provided for eating and swallowing with regard to the appropriate posture to adopt while eating and the different types of food, eating utensils, and oral motor function.

The third largest patient group in the present study comprised healthy children. Many of the legal guardians of these children were concerned about their manner of eating or drinking. Awareness of behavioral eating issues has increased among legal guardians in recent years, and one possible explanation for this could be the widespread use of the Internet; legal guardians are able to access the Center's homepage to obtain information about behavioral eating issues.

Most of the patients visiting the Center for the first time were < 1 year of age. This may be because breastfeeding problems in neonates become evident shortly after birth and that dysphagia, complicated by central nervous system disorders, is prevalent among young patients. Treatment of dysphagia during the developmental stage should occur at the earliest age possible. It is more effective to initiate eating and swallowing training early after the loss of feeding reflexes, before children are started on solid foods. Although sucking is an innate motor function, masticatory movements have to be learnt¹²⁻¹⁴. Many infants were brought to the Center for their first visit, indicating that the initiation of early instruction has led to an effective support system in neighboring regions.

With regard to nutrition intake methods, approximately 50% of patients were on non-oral or partial oral intake. Dysphagia appears shortly after birth in neonates with disabilities due to neurological disorders, such as cerebral palsy, and both weight and height gain are lower in these children because of insufficient nutrition. It has been reported that the proportion of children with cerebral palsy of average height and weight at the time of school entry is 2.5%–10%^{5,15}. Thus, tube feeding is used alone or in combination with oral intake in many children with disabilities caused by neurological disorders such as cerebral palsy. Pediatric patients who rely partially or fully on non-oral intake express a desire to have the feeding tube removed. The legal guardians of these children also strongly desire that they “eat through the mouth” and many request eating instructions for their children.

Concerns regarding eating, requests to have a patient's eating method observed, and requests for eating training accounted for 60% of attendances at the Center. Because approximately 50% of patients relied on non-oral or partial oral intake, the primary reason that many pediatric patients attended the Center was related to their desire to eat through the mouth. In addition, the legal guardians seemed to have a strong desire for their children to be able to enjoy eating food.

Eating is essential for maintaining life, growth, and development. Eating through the mouth

is connected to the acquisition of communication and social skills¹⁶⁾. It is also important for emotional bonding with parents^{17,18)}. Oral intake allows mouth hygiene to be maintained and is associated with other functions, such as language. Medical advances have enabled the maintenance of life without oral intake. However, parents consider “eating through the mouth” important. It is necessary for the Center to continue providing professional treatment for dysphagia to contribute to community medicine.

Conflict of interest disclosure

The authors have declared no conflict of interest.

References

- 1) Policy Planning Division, Department of Health and Welfare for Persons with Disabilities, Social Welfare and War Victims' Relief Bureau, Ministry of Health, Labor and Welfare. Heisei 23nen seikatsu no shizurasa nado ni kansuru chosa (zenkoku zaitaku shogaiji sha tou jittai chosa): kekka no gaiyo (Internet). 2011. (accessed 2014 Nov 6) Available from: http://www.mhlw.go.jp/toukei/list/seikatsu_chousa_b.html (in Japanese).
- 2) Morisaki I. Shika iryou ni okeru supesharu nizu. In Japanese Society for Disability and Oral Health ed. Special needs dentistry. Tokyo: Ishiyaku Shuppan; 2009. pp2-5. (in Japanese).
- 3) Turner L, Jacobsen C, Humenczuk M, *et al*. The effects of lactation education and a prosthetic obturator appliance on feeding efficiency in infants with cleft lip and palate. *Cleft Palate Craniofac J*. 2001;**38**:519-524.
- 4) Morris ES, Klein MD. The child who has a cleft lip or palate. In Morris ES, Klein MD. Pre-feeding skills: a comprehensive resource for mealtime development. 2nd ed. Pennsylvania: Harcourt Health Sciences; 2000. pp651-658.
- 5) Calis EA, Veugelers R, Sheppard JJ, *et al*. Dysphagia in children with severe generalized cerebral palsy and intellectual disability. *Dev Med Child Neurol*. 2008;**50**:625-630.
- 6) Del Guidice E, Staiano A, Capano G, *et al*. Gastrointestinal manifestations in children with cerebral palsy. *Brain Dev*. 1999;**21**:307-311.
- 7) Rogers B. Feeding method and health outcomes of children with cerebral palsy. *J Pediatr*. 2004;**145**(2 suppl):S28-S32.
- 8) Dobbelsteyn C, Marche DM, Blake K, *et al*. Early oral sensory experiences and feeding development in children with CHARGE Syndrome: a report of five cases. *Dysphagia*. 2005;**20**:89-100.
- 9) Baujat G, Faure C, Zaouche A, *et al*. Oropharyngeal motor disorders in Pierre Robin syndrome. *J Pediatr Gastroenterol Nutr*. 2001;**32**:297-302.
- 10) Rempel G, Moussavi Z. The effect of viscosity on the breath-swallow pattern of young people with cerebral palsy. *Dysphagia*. 2005;**20**:108-112.
- 11) Reilly S, Skuse D, Poblete X. Prevalence of feeding problems and oral motor dysfunction in children with cerebral palsy: a community survey. *J Pediatr*. 1996;**129**:877-882.
- 12) Hironaka S, Kinoshita K, Yokoyama R, *et al*. A survey of the clinic for eating disorders in Hokkaido University Dental Hospital —1991 to 1998—. *Jpn J Ped Dent*. 2000;**38**:589-594. (in Japanese).
- 13) Arvedson J, Rogers B, Buck G, *et al*. Silent aspiration prominent in children with dysphagia. *Int J Pediatr Otorhinolaryngol*. 1994;**28**:173-181.
- 14) Fucile S, Gisel E, Lau C. Oral stimulation accelerates the transition from tube to oral feeding in preterm infants. *J Pediatr*. 2002;**141**:230-236. Erratum in: *J Pediatr*. 2002;**141**:743.
- 15) Sullivan PB, Juszczak E, Bachlet AM, *et al*. Gastrostomy tube feeding in children with cerebral palsy: a prospective, longitudinal study. *Dev Med Child Neurol*. 2005;**47**:77-85.
- 16) Morris ES, Klein MD. Foundations for a Mealtime. In Morris ES, Klein MD. Pre-feeding skills: a comprehensive resource for mealtime development. 2nd ed. Pennsylvania: Harcourt Health Sciences; 2000. pp7-13.

- 17) Field D, Garland M, Williams K. Correlates of specific childhood feeding problems. *J Paediatr Child Health*. 2003;**39**:299-304.
- 18) Takahashi M, Hagiwara S, Hihara N, *et al.* A study on the effects of professional instructions to outpatients with dysphagia in a regional habilitation center for children with disabilities —swallowing functions at the first examination in comparison with those at the final evaluation—. *Jpn J Dysphagia Rehabil*. 2009;**13**:231-236. (in Japanese).
- 19) Mason SJ, Harris G, Blissett J. Tube feeding in infancy: implications for the development of normal eating and drinking skills. *Dysphagia*. 2005;**20**:46-61.
- 20) Ooka T, Takahashi M, Mukai Y. The relationship between feeding characteristics and feeding function in children with intellectual disability. *Pediatr Dent J*. 2012;**22**:145-154.

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