FEEDING THERAPY FOR CHILDREN WITH FOOD REFUSAL

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ABSTRACT

Disabled children suffer not only from their primary disease, but also from other complications, including food refusal. The purpose of this study was to elucidate the relationship between these conditions and food refusal in disabled children. The effectiveness of feeding therapy in treating food refusal was also examined. The study subjects were 67 disabled children (35 boys and 32 girls; mean age at initial examination: 6.5 years, SD: 6.0 years) who attended the Nippon Dental University Hospital between April 2004 and August 2008. Of them, the 13 subjects who were diagnosed as those who refused food received feeding therapy combined with desensitization therapy for hypersensitivity. Approximately 20% of the subjects showed food refusal symptoms. Primary disease, respiratory impairment and gastroesophageal reflux were not causes of food refusal in this population. There was a significant relationship between food refusal and hypersensitivity (p = 0.021). After receiving feeding therapy, six of the seven subjects with hypersensitivity but without dysphagia at initial examination recovered from food refusal. Food refusal did not significantly correlate with tube feeding. Hypersensitivity and/or tube feeding may induce food refusal. For subjects with these conditions, feeding therapy combined with desensitization therapy is effective in achieving recovery from food refusal.

Key words: Feeding therapy; Food Refusal; Disabled Children, Orofacial Myology, Dysphagia

INTRODUCTION

Orofacial myofunctional treatment is provided in a variety of ways around the world. In many countries, the speech therapist assumes the rehabilitation for oropharyngeal dysfunction; including orofacial myology, and dysphagia. This article is presented to provide insight into one aspect of orofacial myofunctional treatment in Japan. In Japan, many dentists work with oropharyngeal dysfunction. Specifically, many dentists in Japan are very interested in not only in orofacial myofunctional therapy but also integrate this therapy within dysphagia rehabilitation. The Japanese Society of Dysphagia Rehabilitation, which is the largest group for dysphagia rehabilitation in Japan, has approximately seven thousand members. Membership includes, but is not limited to, medical doctors, dentists, speech therapists, nurses, dental hygienists, occupational therapists, physiological therapists, and dietitians. The main members of authors' group consist of dentists and a speech therapist. The authors treat children with eating problems at the university hospital. Orofacial myofunctional therapy is integrated for this population, particularly using techniques which include desensitization. Thus, this study introduces information on eating problems, especially food refusal.

Feeding is a basic ability required for survival in humans. Although the basic function of feeding is developed in early childhood, it is not a simple process. Before eating, normal babies usually pat, poke, smear, smell, and then taste the food because they delight in the sensory exploration of their world (Erhardt, 1993). The decision to eat a new food is based primarily on a baby's sensory perceptions. However, if babies don't like the way food feels when they

touch it, or the way it looks or smells, they may refuse to taste it or have anything to do with it (Morris and Klein, 2000). In due course of time, as their senses become more familiar with different foods, they start eating foods they previously rejected (Birch and Marlin, 1982; Birch et al., 1987). Some disabled patients, however, still refuse to eat, either because they do not want to eat or are not interested in the food. This condition is known as *food refusal* (Morris SE and Klein, 2000; Nicholls and Bryant-Waugh, 2009).

In most cases, there are many reasons why a baby refuses to eat enough food to grow normally (Morris and Klein, 2000). Gastroesophageal reflux and/or respiratory disease are frequent causes of food refusal. Other possible reasons include lack of muscle tone, malstructure of the oral cavity, lack of appetite, having been tube fed, and primary disease or disability (Morris and Klein, 2000). Moreover, discomfort caused by meals is also a possible reason for refusal to eat. Children who refuse to eat usually have a history and experience of complex and multifaceted eating difficulties, and tend to be malnourished or underdeveloped (Morris and Klein, 2000). Reportedly, approximately 50% of children with developmental disorders may have had eating problems caused by their eating misbehavior (Kedesdy and Budd, 1998). A previous study described that the most common cause of refusal to feeding in children aged 2 years or older tended to be behavioral problems (Rommel et al., 2003).

Feeding therapy for food refusal is not straightforward because patients do not want to cooperate. Therefore, treatment of food refusal is complicated for patients as well as their parents/guardians. Thus, there is no established standard treatment for it. There are some reports of the various methods used to treat patients with food refusal, since the condition involves so many background causes (Dawson et al, 2003; Gulotta et al., 2005). Stimulation of appetite is one of the challenges faced in treating patients with food refusal, since appetite is essential for development of the eating function. One particular treatment of moderate to severe food refusal that was effective in developmentally disabled children was the implementation of a reinforcement program in which patients had access to preferred foods at the beginning of treatment, making them more receptive to eating, since they easily and/or incidentally encountered their preferred foods. Later, the swallowing of non-preferred foods together with improvements in food related behaviors, such as consumption rate, food acceptance and decreased expulsion were noted (Riordan et al., 1980). Sevin et al (2002) reported a trial in which foods from each of the four food groups (i.e., protein, starch, vegetable and fruit) were made into a wet ground texture, with approximately 3 grams (g) presented on a spoon during each trial. Therapists rotated food groups across trials in random order and provided continuous attention (e.g., typical conversation) on a response- independent basis. Twenty food trials were presented with an inter-trial interval of approximately 30 seconds (s). Although the inter-trial interval varied during treatment, the maximum session duration was 1 hour, and six to nine sessions were conducted per day. Each bite was presented in the midline, with a verbal prompt (‘‘take a bite’’). Brief praise was provided for acceptance, and a verbal prompt (‘‘show me’’) was delivered 30 s after the bite was deposited, to check for packing. A reversal design was used to evaluate the effects of treatment on food acceptance and the three target behaviors (disruption, expulsion and packing) (Sevin et al., 2002). In another study, packing could be reduced significantly by the simple food redistribution procedure of massaging, resulting in improved eating in babies with chronic food refusal (Gulotta et al., 2005). In another study, the effect of the high probability instruction for food refusal was investigated (Mace et al., 1988; Dawson et al, 2003). The high probability instruction is a simple fine motor response not related to eating (e.g., touching of the ear), and the low probability instruction is a more complex behavior involving multiple steps (e.g., opening the mouth, manipulating the food, and swallowing) (Dawson et al, 2003).

There are other obstacles in the treatment of disabled children, since they have sensory problems as well (Morris and Klein, 2000). Tactile hyperesthesia, or hypersensitivity, in children, particularly disabled children, often affects the development of their eating function

(Morris and Klein, 2000). Hence, in this study hypersensitivity was the focus in the treatment of food refusal. Hypersensitivity includes hyperreaction, sensory defensiveness and sensory overload (Morris and Klein, 2000). Hyperreaction means a child's strong reactions to a specific sensation, especially one that is perceived as unpleasant or negative, often triggering a "fight-or-flight" response (Morris and Klein, 2000). Moreover, children have difficulty in filtering out unnecessary sensory input because of sensory overload (Morris and Klein, 2000). Problems with foreground-background information can interfere with the ability of feeding in a multisensory environment, while treatment of hypersensitivity is expected to be effective in improving food refusal (Morris and Klein, 2000).

Rehabilitation for oropharyngeal dysfunction, e.g. dysphasia and dysphagia, has recently gained popularity. We use dysphagia rehabilitation or orofacial myofunctional therapy in the treatment of children suffering from eating problems. The treatments performed focus on techniques that include desensitization. Desensitization, a type of indirect training, is effective in improving dysphagia (Tamura et al., 2004). As mentioned previously, Japanese dentistry is very interested not only in orofacial myofunctional therapy, but also in its' integration in dysphagia rehabilitation. Dentists number second only to speech therapists in their employment of dysphagia rehabilitation for their patients. Therefore the authors, mostly dentists, routinely treat children with eating problems. We present our experiences with children with eating problems, especially food refusal, in this study, and determine the effect of feeding therapy employing desensitization therapy.

METHODS

Subject

Sixty-seven children, 35 boys and 32 girls (mean age ± SD at initial examination: 6.5 ± 6.0 years), who attended the Rehabilitation Clinic for Speech and Swallowing Disorders at the Nippon Dental University School of Life Dentistry at Tokyo Dental Hospital, between April 2004 and August 2008, for feeding therapy performed by dentists and a speech therapist, for dysphagia caused during the development phase, were initially enrolled in this study. These 67 patients were examined for the presence of refusal to eat. Based on these examinations, 13 of these patients were diagnosed as having refusal to eat and were analyzed and treated in this study.

This study was approved by the Ethics Committee of The Nippon Dental University, School of Life Dentistry at Tokyo Dental Hospital. Before the start of the study, the purpose and protocol were explained to the patients and their parents/guardians in order to obtain their consent. Each area of the function of eating was investigated, based on their medical history, present illness, and course of feeding therapy performed, as described in their medical charts.

Examination

The following conditions were examined: tendency to refuse food, primary diseases (respiratory impairment, gastroesophageal reflux), history of tube feeding, hypersensitivity and dysphagia (Morris and Klein MD, 2000). In the evaluation, report of the subjects' eating behaviors was as observed by their parents/guardians. A diagnosis of food refusal was made when a subject showed certain symptoms during meals at home or at the feeding consultation sessions. The typical symptoms included closing the mouth, wincing, extremely small appetite or vomiting during feeding. *Food refusal* in this study is defined as a tendency to refuse to eat or absolute refusal to eat.

Patients' eating dysfunction was diagnosed by observing them during the meal. Videofluorography and videoendoscopy were also employed for subjects with suspected aspiration. The subjects were divided into four groups, namely, group A: severe dysphagia, *i.e.* aspiration, choking on food, wheezing, and respiratory impairment; group B: mild/moderate dysphagia with oropharyngeal dysphagia, *i.e.* tongue thrust swallowing; group C: masticatory disorders without both dysphagia and swallowing problems; and group D: no dysphagia. Patients with anticipatory problems, who had perceptual-motor dysfunction of hand-

mouth coordination or who could not recognize food due to cognitive deficits or visual impairment, were excluded from this study.

Feeding therapy

Feeding therapy to develop normal feeding/swallowing functions included environmental therapy. Environmental therapy consisted of selecting appropriate utensils, creating a comfortable atmosphere for eating, providing suitable meals, and helping the subjects assume a proper posture during meals, with psychological consideration. To provide suitable meals, modifications of cooking procedures were required, along with careful attention to intake, in order to avoid malnutrition and dehydration.

The therapy also consisted of functional training, which included both direct and indirect training. In direct training with food, the subjects were guided during meals to use the correct and required movements for feeding or swallowing by supporting the subjects' behavior and movements, and by sustaining the subject's face or chin with the trainer's hands. Indirect training without food ingestion included desensitization therapy to treat hypersensitivity, this treatment being started after completing the initial examination (Ayres, 1974; Farber, 1982; Sakamoto and Hanakuma, 1997; Tamura et al., 2004). In desensitization therapy, the trainer just touches a certain area of the subject with his/her bare finger or hand, without patting or massaging. The area touched was exactly the same as the subjects' hypersensitive area, *e.g.* face, lips or intraoral cavity. Hand touching of the sensitive part of the body is done by the application of firm pressure for several minutes. Hypersensitivity occurs in the oral cavity, rather than extra-oral regions, the most commonly affected part causing hypersensitivity being the lingual side of the upper lip and/or upper gums of the anterior teeth. For the treatment, light touch should not be used since babies with hypersensitivity find light touch to be more of a stimulus than is expected. Hence, patients feel more uncomfortable and may reject a light caressing touch (Morris and Klein MD, 2000). Another original method of indirect training includes stimulation of the muscles in the oral region via massage, following the Vangede method (Tamura et al., 2004), but this massage was excluded from our treatment protocols because of its potential to act as a stimulus for the child, making it uncomfortable.

At the beginning of treatment, subjects may be uncomfortable, but they soon feel calm after several minutes of treatment. This desensitization therapy is usually performed once a day or more. The treatment is employed between meals since it may be uncomfortable for the children if they are treated just before or after a meal. Desensitizing therapy enables recovery from hypersensitivity not only through removal of the sensitivity to touch, but also by inducing acceptance of food material in the mouth, since the trainer's finger or hand is assumed to be a food-like foreign body. In this study, parents or guardians were asked to perform the desensitizing therapy for the subject once or more than once a day. When the subject showed willingness to eat continuously, it was assumed their food refusal had resolved.

Data analysis

Correlations between the items were statistically analyzed by the chi-squared test using Windows SPSS Ver. 16. Statistical significance was accepted as $p < 0.05$.

RESULTS

Subjects who refused to eat

Of the 67 subjects with speech and swallowing disorders, 13 refused to eat while 54 had no such symptom, that is, approximately 20% of the subjects had food refusal in this study.

Relationship between food refusal and primary disease

Among the subjects with food refusal, nine (69%) were diagnosed with intellectual disability, two (15%) with cerebral palsy, five (39%) with various syndromes, and two (15%) with no abnormalities. Among the subjects without food refusal, 32 (59%) were diagnosed with intellectual disability, 16 (30%) with cerebral palsy, 19 (35%) with various syndromes, 14 (26%) with other diseases, and one (2%) with no abnormalities. There was no significant relationship between food refusal and primary disease.

Relationship between food refusal and respiratory impairment, gastroesophageal reflux, hypersensitivity and tube feeding

The subjects were evaluated to determine a correlation between food refusal and respiratory impairment, gastroesophageal reflux, hypersensitivity and history of tube feeding (Table 1). No significant relationship was found between food refusal and respiratory impairment or gastroesophageal reflux. On the other hand, there was a significant relationship between food refusal and hypersensitivity (p = 0.021). Food refusal did not significantly correlate with tube feeding.

Relationship between food refusal and dysphagia

Table 2 shows the relationship between food refusal and the presence/severity of dysphagia. It was determined that nine subjects (17%) in group A (severe dysphagia), eight (15%) in group B (mild to moderate dysphagia), 32 subjects (59%) in group C (mastication disorder), and five subjects (9%) in group D (no dysfunction) did not exhibit food refusal despite having variable levels of dysphagia (in groups A, B and C) or behavioral issues (in group D). The relationship between these factors was significant (p = 0.016). Approximately half of the subjects with food refusal had no feeding dysfunction. Of the subjects with food refusal, two subjects belonged to group A (15%), five belonged to group C (39%) and six were in group D (46%). None of the group B patients with mild to moderate oropharyngeal dysphagia exhibited refusal to feed.

Relationship between food refusal and age and sex

No significant relationship was found between food refusal, age and sex. Nor was there any significant relationship between dysphagia, age and sex.

Table 1. Relationship between food refusal and respiratory impairment, gastroesophageal reflux, hypersensitivity and tube feeding

		Respiratory impairment		Gastroesophageal reflux		Hypersensitivity		Tube feeding	
		Yes	No	Yes	No	Yes	No	Yes	No
Food refusal (No. of subjects)	Yes	4	9	2	11	8	5	7	6
	No	13	41	3	51	15	39	14	40
p-value		0.429		0.247		0.021		0.056	

Table 2. Relationship between food refusal and dysphagia

		Subject Groups			
		Group A: *Severe dysphagia*[#]	Group B: *Mild/moderate dysphagia*[##]	No dysphagia	
				Group C: *Masticatory disorder*	Group D: *No dysfunction*
Food refusal (No. of subjects)	Yes	2	0	5	6
	No	9	8	32	5
p-value		0.016			

[#]Severe oropharyngeal dysphagia
[##]Oropharyngeal dysphagia

Outcome of food refusal following feeding therapy

The profiles of the 13 subjects (five boys and eight girls; mean age = 3.7 years, SD = 3.3 years) who had food refusal at the initial examination, and the significant medical backgrounds involved in the development of food refusal are shown in Table 3. Besides hypersensitivity, these conditions might also have been responsible for the food refusal in the study subjects. Many causes of food refusal have been previously reported (Morris and Klein, 2000; Groher and Crary, 2010). In our study, subject **A** had hypersensitivity and cerebral palsy (Groher and Crary, 2010), subjects **A**, **D**, **E** and **H** had intellectual disability (Baranek and Berkson, 1994) and subject **H** had autism spectrum disorders (Groher and Crary, 2010; Stein et al., 2011). Nine of the 13 subjects, **A**, **B**, **C**, **D**, **E**, **F**, **J**, **K** and **M,** had previously undergone one or more surgical procedures, which might be a reason for their food refusal. We also found that six mothers (**E**, **F**, **I**, **J**, **L** and **M**) tried to intimidate their children into eating, which could have resulted in their food refusal (Faith et al., 2008).

Table 4 shows the outcome of treatment in the 13 patients. At the initial examination, eight subjects (average age = 3.3 years, SD =3.3 years) had hypersensitivity and five (average age = 4.4 years, SD = 3.2 years) did not. Among the eight subjects with hypersensitivity, only one subject had severe dysphagia and was classified as group A, while the remaining seven subjects did not suffer from dysphagia (groups C & D). Among these seven subjects, five subjects recovered from their hypersensitivity by desensitizing therapy, requiring 3 (±1.8 months) for the treatment. Moreover, six of the seven subjects mentioned above recovered from food refusal, requiring 12.2 ± 6.9 months (mean ± SD) for the treatment to be effective. On the other hand, among the five subjects without hypersensitivity described above, only one subject in group C recovered from food refusal, recovery taking over 28 months.

Table 3. Profiles of the 13 subjects with food refusal

Groups	Subject	Age	Sex	Diagnosis	Tube feeding
Group A: *Severe dysphagia*[#]	A	0 y. 6m.	F	Cerebral Palsy, Lissencephaly, Intellectual disability	Nasogastric tube
	B	0 y. 6m.	F	Cerebral palsy, Intellectual disability	Nasogastric tube
Group C: *Masticatory disorder*[##]	C	6 y.11 m.	M	Down syndrome, Intellectual disability	Gastrostomy tube
	D	12 y. 5m.	M	FG syndrome, Intellectual disability	History of nasal tube feeding
	E	3 y. 10m.	M	Down syndrome, Intellectual disability	History of nasal tube feeding
	F	3 y. 1 m.	M	Apert syndrome, Intellectual disability	Gastrostomy tube
	G	7 y. 3 m.	F	Rett syndrome, Intellectual disability	None
Group D: *No dysfunction*[#] [#]	H	1 y. 7 m.	F	Sotos syndrome, Intellectual disability, Autism spectrum disorders	None
	I	1 y. 5 m.	F	None	None
	J	1 y. 0 m.	F	Congenital tracheal stenosis	None
	K	3 y. 3 m.	F	Spina bifida, Intellectual disability	Gastrostomy tube
	L	6 y. 11m.	M	None	None
	M	0 y. 6 m.	F	Multiple abnormalities, Intellectual disability, Autism spectrum disorders	Nasogastric tube

[#]Severe oropharyngeal dysphagia

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Table 4. Treatment Outcomes

Dysphagia classification	Subject	Hyper-sensitivity	Outcome of hypersensitivity treatments	Outcome of food refusal treatments
Group A: Severe dysphagia[#]	A	+	Recovered	Failed
	B	-	N/A	Failed
Group C: Masticatory disorder[##]	C	-	N/A	Failed
	D	+	Recovered	Recovered
	E	+	Recovered	Recovered
	F	+	Failed	Recovered
	G	-	N/A	Recovered
Group D: No dysfunction[##]	H	+	Recovered	Recovered
	I	+	Recovered	Recovered
	J	+	Recovered	Recovered
	K	+	Failed	Failed
	L	-	N/A	Failed
	M	-	N/A	Failed

[#]Severe oropharyngeal dysphagia

[##]Without dysphagia

N/A: not applicable

DISCUSSION

Food refusal is one of the factors that inhibit proper development of the eating function. However, only a few reports have analyzed the causes of food refusal using objective and systematic measurements (Morris and Klein, 2000; Tago et al, 2005). Food refusal might be associated with gastrointestinal or respiratory complications (Hyman, 1994; Böhmer et al., 1997; Böhmer et al, 1997; Mathisen et al., 1999; Tago et al., 2005; Haas and Maune, 2009). However, the current study did not find any positive correlation between food refusal and these conditions.

A strong relationship was found between food refusal and hypersensitivity (Tago et al, 2005). In this study, most of the desensitized subjects recovered from food refusal, suggesting that hypersensitivity is closely related to food refusal. The cause of hypersensitivity, however, has not been elucidated. Physically disabled infants, such as those with cerebral palsy, may develop hypersensitivity and tend to resist being touched on their body or near the mouth and oral cavity, which may be due to lack of sensory motor experience (Omoto, 2005). The human central nervous system matures through stimulation of body movement through interaction with the environment after birth (Sakamoto and Hanakuma, 1997). Healthy babies also have hypersensitivity in the neonatal period, which plays a role in protecting the oral cavity from harmful stimuli (Morris and Klein, 2000). Even if they cannot adapt to the surrounding environment, their

hypersensitivity may gradually disappear with repeated sensory and motor inputs induced by tactile stimuli (Morris and Klein, 2000). However, disabled babies who cannot stimulate their tactile sense—for example, who cannot lick their own fingers—are likely to develop dysfunction of sensory integration. Disabled babies have difficulty in adapting to environmental stimuli, and have less potential to learn how to adjust to environmental stimuli (Sakamoto and Hanakuma, 1997). Therefore, hypersensitivity in these babies often affects the development of their eating function, as mentioned above. Although desensitization therapy has been performed to relieve hypersensitivity, in an attempt to improve the function of eating (Tamura et al., 2004), no definite clinical protocols, evaluation methods and standardized training protocols for the treatment have as yet been established.

The application of sensory integrative therapy, which may be indicated for the treatment of sensory integration dysfunction, might be an appropriate desensitization therapy and can lead to recovery from food refusal (Ayres, 1974; Farber, 1982; Sakamoto and Hanakuma, 1997). In this study, it was determined that the acceptance of stimuli in the mouth through desensitizing therapy contributed to recovery from food refusal.

Hament et al. reported on the relationship between n*on-oral feeding* and food refusal (Hament, 2001). Long-term tube feeding or a gastric fistula may suppress development of the normal cycle of fullness and emptiness of the stomach and, hence, decrease appetite, since babies under these circumstances cannot experience eating behaviors, including suckling (Kedesdy and Budd, 1998). Furthermore, the neonatal to infancy period is very important for babies to develop a sense of taste, because lack of the tasting experience may induce hypersensitivity to taste and make perception of food taste difficult (Morris and Klein, 2000). Among the 13 subjects with food refusal in this study, eight had a history of tube feeding, and five of these eight subjects refused to eat. Although tube feeding offers the advantage of providing complete and certain nourishment for children, oral ingestion should be started as soon as possible because of the negative impact of tube feeding. Conversely, attempting to force oral ingestion in order to terminate tube feeding is counterproductive in children who receive tube feeding but do not have a functional disorder. In these cases, food refusal may be caused by an unpleasant experience and/or memory associated with treatment of the primary disease or eating. Higher priority should be given to eliminating unpleasant memories related to oral ingestion. For this purpose, food refusal treatment in children may necessitate a multidisciplinary approach, including psychological and environmental interventions (Fischer and Silverman, 2007). At the time of starting feeding therapy, parents/guardians tend to raise their voice to their children and force them to eat. Delicate attention by parents/guardians not only to their babies or children, but also to understanding the condition, is very important in the treatment of food refusal and in desensitization therapy, which they themselves, the people who are closest to the patient, must perform.

CONCLUSION

Among disabled children who attended our university hospital to receive feeding therapy by dentists and a speech therapist, approximately 20% presented with food refusal. It took approximately 3 months for children with food refusal who had hypersensitivity to be desensitized, and recovery from food refusal was accomplished in approximately 1 year. Hypersensitivity is, reportedly, closely related to food refusal. In our study, feeding therapy combined with desensitization therapy was effective in achieving successful recovery from food refusal. This study also demonstrated the benefits of an interdisciplinary team approach, which integrates orofacial myofunctional treatment methods for dysphagia.

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