



Packing and Problematic Feeding Behaviors in CHARGE Syndrome: A Qualitative Analysis



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ABSTRACT

Objective: To understand packing and problematic feeding behaviors during mealtime in individuals with CHARGE syndrome. Packing, or holding food in one's cheeks without swallowing, is an adverse feeding behavior that has been described in children with autism and Down syndrome, and in those transitioning from tube to oral feeding. It has never been described in detail in CHARGE syndrome, a genetic disorder with a high prevalence of feeding difficulties, tube feeding, and otorhinolaryngological issues.

Methods: A mixed methods approach used descriptive and qualitative content analysis of interviews with parents of children, adolescents, and adults with CHARGE syndrome. Individuals had previously or were currently experiencing packing or overstuffing one's mouth with food during eating.

Results: Twenty parents completed a phone interview, describing their child/adult's (2–32 years) adverse feeding behaviors. Individuals had a higher proportion of cleft palates (40%) in comparison to the general CHARGE population (15–20%). Parents reported food packing most commonly with bread and pasta (33%), and reported that food was held in cheeks for hours after a meal had ended (35%). Packing was reported to prolong mealtimes for over an hour (30%). Parents were worried about choking during eating (30%). Food packing was also reported in individuals who had never needed G/J tube feeding or feeding therapy, in addition to those who had needed both.

Conclusion: This study provides an in-depth description of parents' experiences with packing and adverse feeding behaviors in individuals with CHARGE syndrome. These feeding behaviors are an important addition to the knowledge of the highly prevalent feeding difficulties in this genetic disorder. Individualized evaluation of feeding behavior should be a part of the standard otolaryngologic and feeding team practice for these patients.

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1. Introduction

CHARGE syndrome is a genetic condition found in approximately 1 in 15,000 live births worldwide [1–4]. Diagnosis of CHARGE syndrome is initially clinical and confirmed by genetic testing [2,3]. Due to the broad range of clinical symptoms, extensive multidisciplinary treatment and management is needed, including assessment of all major organ systems and behavioral management [4]. Treatment and interventions can involve medical and surgical management including otolaryngology, deaf/blind services, occupational therapy, physiotherapy, speech/language

therapy, cochlear implant, behavior therapy, psychological counseling, and more [4,5].

Children with CHARGE syndrome commonly experience feeding difficulties, with up to 90% reporting gastroesophageal reflux, poor sucking and swallowing skills, and G/J tube feeding [4,6–8]. Many parents report “odd” eating behaviors in their children with CHARGE syndrome when they transition from tube to oral feeding, such as taking large bites of food, overstuffing food into one's mouth before swallowing, and holding food in cheeks [6,7]. Severe feeding difficulties persist over the entire lifespan in this genetic condition, lasting throughout adolescence and adulthood [9–11].

Holding or pocketing food in ones' cheeks and mouth without swallowing has been described as ‘packing’ [12]. Packing has been identified as a problematic feeding behavior in children with autism spectrum disorders and children with feeding disorders [12–15]. Pocketing of food, or packing, has also been identified in Down Syndrome and described as leading to

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choking [16]. It has not previously been described in detail in CHARGE syndrome.

Packing, in addition to poor sucking and swallowing skills, may lead to an increased risk of aspiration, malnutrition, choking, and even death [16]. Aspiration and abnormal swallowing, including poor bolus mobility and pooling of secretions, have been previously identified in 60–80% of children with CHARGE syndrome [8]. Choking while eating has resulted in death in an eight-year-old child with CHARGE syndrome [17]. One of the authors (KB) is also aware of other individuals with CHARGE syndrome who had severe consequences due to choking on food. These consequences were anoxic cerebral palsy and a fatality due to choking on a piece of broccoli. Feeding difficulties, in addition to breathing difficulties and reflux, were significantly more prevalent in children with CHARGE syndrome who died before age 10 than those who survived longer [17].

Otorhinolaryngological and gastrointestinal tract surgeries are the most common surgeries in CHARGE syndrome [18]. Dental procedures and diagnostic scopes (nasopharyngoscopy, laryngoscopy, and bronchoscopy) are the next most common surgical procedures reported in children, adolescents, and adults with this genetic disorder. [18]. Excess oral secretions have also been identified as an issue in CHARGE syndrome [19]. Individuals may need to undergo botulinum toxin injections into their salivary glands to reduce oral secretions [19]. Numerous surgical procedures on the airway and gastrointestinal tract and excess salivary gland secretions can contribute to the feeding difficulties, dysphagia, and high risk of aspiration in this population [6,18,19]. ENT is one of the most common medical specialties that continues to follow individuals with CHARGE syndrome, with over 85% of adolescents and adults reporting ENT involvement in their care [9].

Despite the potential negative consequences of packing and the high prevalence of feeding difficulties in this genetic disorder, this problem has received little attention in the literature and has not been investigated in CHARGE syndrome. The objective of this study was to understand and describe packing and other problematic feeding behaviors in individuals with CHARGE syndrome through in-depth parent interviews. We aimed to understand the problematic feeding behaviors from the perspective of the parents themselves.

2. Methods

2.1. Demographics survey

Information regarding demographics as well as the major and minor phenotypic features of CHARGE syndrome was collected (Appendix A) [5,20].

2.2. Semi-structured interview

Participants completed a semi-structured interview consisting of open-ended and closed-ended questions (Appendix B). The interview questions explored general eating, problematic feeding behaviors (e.g., packing), chewing, swallowing, choking, and feeding therapy. The interview questions for the semi-structured interview were developed from the current literature on food packing [11,17–19], and feeding difficulties experienced in CHARGE syndrome [6,8,12]. Several interview questions were based on the Pediatric Assessment Scale for Severe Feeding Problems [20]. Clinicians and experts in pediatric feeding issues reviewed and revised the interview questions. Reviewers included a general pediatrician whose clinic includes individuals with CHARGE syndrome, a speech language pathologist specializing in feeding, a feeding specialist from a pediatric feeding team, an

occupational therapist, a pediatric gastroenterologist, and a parent of an older child with CHARGE syndrome.

2.3. Participants

Parents of individuals with CHARGE syndrome who were currently experiencing or had previously experienced food packing, overstuffing one's mouth during eating, or both feeding behaviors, were included in this study. Individuals had to have a confirmed genetic or clinical diagnosis of CHARGE syndrome and be older than 1 year.

2.4. Procedure

Approval for this study was obtained through the I.W.K. Health Centre Research Ethics Board. Invitations to participate were sent via CHARGE syndrome organizations in the USA, Canada, Australia, and the UK. Members of these international and national organizations included parents of individuals with CHARGE syndrome. The invitations included information about the study and eligibility criteria.

Informed consent was obtained from each parent before participation. Participants completed the Demographic survey and the pre-set interview questions during a recorded phone interview with the primary author [AH]. Interviews were conducted until data saturation was reached for the pre-set questions. The primary author [AH] conducted all of the interviews. Interviews were audio recorded and transcribed verbatim. The transcriptions were checked for accuracy.

2.5. Analysis

Interviews were analyzed using a qualitative content analysis of open-ended questions and a descriptive analysis of closed-ended questions. Continuous variables were expressed as mean (M) ± standard deviation (SD) and categorical variables were expressed as frequency (N) and percentage (%). All analyses were done in SPSS Version 20.0.0 (IBM® SPSS® Statistics, 1 New Orchard Road Armonk, New York).

The primary author reviewed the responses to the open-ended questions and created content categories, with definitions for each category. The data was coded by these categories. The number of responses in each category was summed. Separate categories were created for each of the open-ended questions. All analyses were done in MAXQDA Version 11.2.1 (Udo Kuckartz Berlin®). Direct quotations from the interviews were taken to support the categories. A second coder independently reviewed and validated the coding and content categories.

3. Results

3.1. Participants

Between April and August 2015, 20 parents (95% mothers) of individuals with CHARGE syndrome (12 female, 60%) completed a phone interview. Interviews took an average of 44 min (SD = 11 min). Fifteen individuals (75%) had a confirmed genetic diagnosis of CHARGE syndrome (CHD7 mutation positive). One individual (5%) did not demonstrate the CHD7 gene mutation and four individuals (20%) had not undergone genetic testing, but all clinically met the criteria for typical/definite CHARGE syndrome using both the Verloes criteria (2005) [15] and the Blake criteria (1998) [5].

The average age of individuals with CHARGE syndrome was 9.9 years (SD = 8.6 years) and ranged from 2 to 32 years. Individuals were from USA ($n = 11$, 55%), Canada ($n = 4$, 20%), Australia/New

Table 1
Frequency of major and minor phenotypic CHARGE syndrome features in the study population.

	Count (%) (n = 20)	General CHARGE syndrome population frequency [4]
Major CHARGE syndrome features		
External ear abnormalities	19 (95)	95–100%
Middle ear abnormalities	15 (75)	
Inner ear abnormalities	19 (95)	90%
Abnormal semicircular canals	11 (55)	
Coloboma (Retina/Iris)	15 (75)	75–90%
Choanal stenosis/Atresia	13 (65)	65%
Cranial nerve dysfunction		
CN I (Difficulty smelling)	11 (55)	
CN VII (Facial palsy)	13 (65)	50–90%
CN VIII (Difficulty hearing)	19 (95)	
CN IX, X, XI (Difficulty swallowing)	17 (85)	
Minor CHARGE syndrome features		
Distinct CHARGE facial features	19 (95)	
Developmental delay	20 (100)	70%
Major/Minor heart defect	17 (85)	50–85%
Cleft lip or Palate	9 (45) ^a	15–20%
Cleft lip	6 (30)	
Cleft palate	8 (40)	
Genital hypoplasia	10 (50)	50–70%
Growth deficiency	11 (55)	70–80%
Tracheoesophageal fistula	1 (5)	
Urinary tract problems	6 (30)	

^a Indicates a higher frequency than reported in the general CHARGE syndrome population [4].

Zealand ($n = 4$, 20%), and Europe ($n = 1$, 5%). Table 1 displays the frequency of major and minor phenotypic CHARGE syndrome characteristics in this study sample and the reported known frequencies of these characteristics in the general CHARGE syndrome population.

A higher proportion of the individuals with CHARGE syndrome in this study ($n = 8/20$, 40%) had a cleft palate in comparison to the general CHARGE population frequency of 15–20%. Four other individuals (20%) had a medically diagnosed high palate and one individual (5%) had a sub-mucosal cleft palate.

3.2. Feeding characteristics

Parents' reports of their child/adult's most common feeding and swallowing characteristics are presented in Table 2. All individuals had parent-reported difficulties with chewing their food. The majority of individuals needed to be closely supervised during eating. The majority of parents had to closely supervise and remind their child/adult to chew and swallow while eating. Additional feeding difficulties reported by parents were choking or coughing while eating and food accidentally falling out of the mouth.

The parent-reported feeding history, including tube-feeding duration, medication use for gastroesophageal reflux, and surgeries to manage feeding and/or swallowing is displayed in Table 3.

Eighteen parents (90%) reported that their child/adult had experienced swallowing difficulties over their life. Fourteen of these 18 parents (70%) reported that the swallowing difficulties were currently ongoing. Parents reported difficulties with aspiration ($n = 5/18$, 28%), a weak swallow ($n = 3/18$, 17%), and problems swallowing certain food textures ($n = 3/18$, 17%).

Chewing problems was the most prevalent feeding difficulty, reported by all parents. Seven parents (35%) reported an abnormal or uncoordinated chewing motion. Parents also reported an abnormal bite ($n = 4/20$, 20%) and low muscle tone that interfered with chewing ($n = 3/20$, 15%).

Table 2
Feeding and swallowing characteristics of the twenty individuals with CHARGE syndrome.^a

	Count (%) (n = 20)
Chewing problems	20 (100)
Parents cut food for child/Adult	18 (90)
Parents have to tell to swallow when eating	16 (80)
Parents have to tell to chew when eating	14 (70)
Have aspirated or coughed while eating	13 (65)
Food falls out of mouth when eating	10 (50)
Difficulties with food texture	
Liquids	8 (40)
Puree	0 (0)
Mashed/Lumpy	5 (25)
Chewable/Solid	13 (65)
Current oral aversion	7 (35)
Eats a meal unattended	7 (35)
Parents feed the child/Adult	6 (30)
Prefers to eat non-food items	0 (0)

^a Parents described their child/adult's typical feeding difficulties during a meal through a series of pre-set questions (see Appendix A).

Table 3
Medical feeding history of the twenty individuals with CHARGE syndrome.

	Count (%) (n = 20)
Never tube fed	3 (15)
Previously tube fed	12 (60)
Currently supplements oral feeding with tube feeding	5 (25)
Duration of tube fed (previous and current)	
<1–5 years	11 (55) ^a
6–10 years	5 (25) ^b
13 years	1 (5)
Gastroesophageal reflux medication use ^c	16 (80)
Surgery to manage feeding/swallowing problems ^d	10 (50)
Nissen fundoplication	5/10 (50)

^a Four of these 11 individuals still use tube feeding to supplement oral feeding.

^b One of these five individuals still uses tube feeding to supplement oral feeding.

^c Includes Lansoprazole, Ranitidine, Omeprazole, Nizatidine.

^d Includes a vascular ring repair, cleft palate repair, choanal atresia repair, esophageal atresia repair, esophageal dilatation, adenoid and tonsillectomy, and sternocleidomastoid release.

Seven parents (35%) reported difficulties with their child/adult's tongue movement (e.g., not able to move the tongue forward out of the mouth). Three parents (15%) reported that their child/adult previously had a tongue-tie (ankyloglossia) that was surgically released.

3.3. Food packing

Fifteen parents (75%) reported that their child/adult with CHARGE syndrome would pack food in their cheeks. Bread, pasta, and crackers were the most common reported texture of packed food. Non-smooth food textures (e.g., rice) and meat were the next most common reported textures of packed food. The parents' descriptions of the food packing included describing the behavior as being 'chipmunk-like' ($n = 3/20$, 15%), as a result of over-stuffing one's mouth ($n = 3/20$, 15%), and that food would also pack in their palate ($n = 2/20$, 10%).

Seven parents (35%) reported that there would be food packed in cheeks for prolonged periods of time after a meal had ended. Three of these 7 parents (15%) reported that the food would be packed for at least an hour, and two parents (10%) reported packing for two hours after the meal. Three parents (15%) reported that their child/adult would also pack saliva in their cheeks. Three parents (15%) reported that they were worried about poor teeth hygiene and cavities as consequences of food packing.

3.4. Problematic feeding behaviors

Nineteen parents (95%) reported that their child/adult would over-stuff their mouth with food while eating a typical meal. Parents highlighted that the excess food had to be spit out or removed by the parent ($n = 5/20$, 25%), and that the child/adult would choke or cough ($n = 4/20$, 20%). Three parents (15%) described that this feeding behavior of over-stuffing resulted in negative behavioral interactions with the parent. These individuals were in late adolescence and early adulthood (age 16, 17, and 20 years). The negative interactions were reported as getting into arguments with the parent when prompted to swallow, trying to feed the parent to increase the speed of the family mealtime, and using mouth over-stuffing as a tool to choke oneself when upset.

Two parents (10%) reported that the mouth over-stuffing feeding behavior interfered with their adolescent's (age 17 and 20 years) social interactions with peers. The parents reported that the adolescents were at risk of social isolation due to their feeding behavior and that they threw out friends' lunches because they had not finished their meals as quickly as them. Two parents (10%) described their child/adult's mouth over-stuffing feeding behavior as manipulative, such as over-stuffing all foods except for sweets and chocolate bars, and learning how to drink a liquid chaser (a liquid given to drink with every bite of food to reduce food packing) without removing the food packed in their cheek.

Twenty-five percent of parents ($n = 5/20$) reported that their child/adult would not mix liquids and solid food during mealtime. Six parents (30%) reported that their child/adult needed to finish all food and have an empty plate. Four parents (15%) reported that the same routine had to be followed at each mealtime (e.g., needing to eat the same foods at each meal). Three parents (15%) reported anger and aggression from the child/adult if the plate of food was taken away or not available to them, which their parents were doing in an attempt to reduce food packing.

3.5. Food textures

Parents' reports of their child/adult's feeding difficulties with different food textures are displayed in Table 2. All participants reported that there was no adverse feeding difficulties with puree food textures. Three parents further detailed that puree was "the favorite texture". The majority of parents reported that mashed foods caused no eating issues although four of these parents indicated that the texture had to be mashed very smoothly with no lumps. Individuals had the most reported feeding issues with liquids and solid food textures. Six parents specifically identified that meat was a very problematic food. Two thirds of individuals had trouble with drinking water out of an open cup and/or controlling the speed of drinking. Only one parent reported that their child/adult was able to eat hard vegetables and fruit.

3.6. Time to eat a typical meal

Qualitative analysis of parents' descriptions of a typical mealtime included both short and prolonged mealtimes. Four parents (20%) described that their child/adult with CHARGE syndrome ate very quickly because they were stuffing their mouths and finishing the food in a short amount of time (15 min or less). Conversely, six parents (30%) reported that packing feeding behaviors slowed the eating process down and led to an increased time of eating (60 min or more). Half of the individuals ($n = 10/20$) ate a typical meal in the span of 20 to 45 min.

Table 4

Therapies employed for feeding difficulties.

	Count (%) ($n = 20$)
Feeding therapy use ^a	18 (90)
Speech language pathologist	15 (75)
Occupational therapist	13 (65)
Psychologist	2 (10)
Reflux medication use	16 (80)
Liquid or solid chaser use when eating	15 (75)
Past use of modified utensils when eating	14 (70)
Surgeries for eating and swallowing	10 (50)
Current use of modified utensils when eating	8 (40)
Therapy to reduce saliva (i.e., Botox therapy)	5 (25)
Have to remove food from cheeks when eating	2 (10)

^a Individuals reported any past/current feeding therapy from one or more of the following therapists: speech language pathologist, occupational therapist, and psychologist.

3.7. Oral cavity hyposensitivity

When asked to give a general description of their child's/adult's mouth food packing and problematic feeding behaviors, three parents (15%) reported that they thought these feeding behaviors were a cause of a lack of sensory feedback in the mouth. Four parents (20%) described their child's/adult's mouth as "under-sensitive". Descriptions included not feeling food leftover on their lips and not noticing the food in their cheek until the parent physically pressed on it with their hand. One parent remarked that "overstuffing and pocketing—it is only in her facial palsy side. Her side that works, she does not pocket food whatsoever".

3.8. Choking

Six parents (30%) reported worrying that the food packing and mouth over-stuffing feeding behaviors would cause their child/adult to cough and choke. Overall, the majority of the individuals with CHARGE syndrome in this study ($n = 13/20$, 65%) had reportedly coughed or choked excessively and recurrently during oral feeding. Four parents (20%) reported coughing and choking occurring while their child/adult was exhibiting food packing and mouth over-stuffing feeding behaviors. The majority of parents ($n = 13/20$, 65%) reported that close supervision during meals was a necessity.

3.9. Specific feeding interventions

The majority of parents reported that their child/adult had received therapy for feeding/eating issues from a speech-language pathologist, occupational therapist, or psychologist ($n = 18/20$, 90%). Therapy and interventions for feeding difficulties and problematic feeding behaviors are displayed in Table 4. Parents reported the top three interventions used at mealtime to reduce the food packing and adverse feeding behaviors (Table 5). The common presenting features of food packing and other feeding difficulties in CHARGE syndrome are summarized in Table 6.

4. Discussion

This is the first study to investigate packing and adverse feeding behaviors in individuals with CHARGE syndrome. Qualitative analysis of parent interviews provide a comprehensive view of these problematic eating behaviors and how they affect both the individual and their parents' lives [21,22]. It is important to understand the parents' experiences of their child/adult's adverse feeding behaviors—information that may not be captured in quantitative studies.

Table 5Main themes: Parent's top interventions to reduce their child's/adult's packing and adverse feeding behaviors^a

Intervention theme	Number of parents who reported (%)	Specific examples
Verbal prompts during eating	8 (40)	<ul style="list-style-type: none"> • Have a gestural prompt or cue (i.e., touch his hand) when starts over-stuffing • Tell or sign to chew and eat before give more food
Soft foods	7 (35)	<ul style="list-style-type: none"> • Cook to a soft consistency • Soft diet • Blending and pureeing the food • Give textures that they can handle easily
Include in the family dinner	7 (35)	<ul style="list-style-type: none"> • Sit at the dinner table with everyone • Eat similar foods as the family
Close supervision	5 (25)	<ul style="list-style-type: none"> • Be present during feeding • Have an occupational therapist watch him/her eat once a month
Distractions	4 (20)	<ul style="list-style-type: none"> • Use an iPad to distract while eating • Watch a favorite TV show during eating • Use favorite foods as incentives • Create a calm environment
Liquid chaser	4 (20)	<ul style="list-style-type: none"> • Give plenty of liquids • Encourage to take a sip of water often
Cutting the food	4 (20)	<ul style="list-style-type: none"> • Cut it to the preferred shape • Cut into tiny pieces
Limit food on utensil	4 (20)	<ul style="list-style-type: none"> • Give one item at a time • Put a small amount on the fork • Move the plate away
Practice eating	3 (15)	<ul style="list-style-type: none"> • Have 3 meals and 2 snacks a day to help practice eating • Have solid and puree options to practice chewing and swallowing • Maintain the same structured order of eating for each meal
Use modified utensils	3 (15)	<ul style="list-style-type: none"> • Use a smaller spoon
Model the eating process	2 (10)	<ul style="list-style-type: none"> • Model eating, chewing, and swallowing during the meal • Bring a mirror to the table to watch self-eating
Use favorite foods as incentives	2 (10)	<ul style="list-style-type: none"> • Give rewards • Use food they like

^a Parents reported their top 3 interventions to reduce their child/adult's mouth over-stuffing and food pocketing during oral eating (see Appendix A)

Packing has been investigated in individuals with autism and Down syndrome, and has been identified as a frequent behavior in children with feeding disorders [12,15,16]. Abnormal tongue movement has been identified as a cause of pocketing food (packing) in Down syndrome, which was also described in our study population [16].

A recent review by Kerzner et al. [23] on the classification and management of feeding difficulties described that pocketing (food retained in cheeks) can occur in children with a fear of feeding, such as those who were tube fed. They specified that these children might feel threatened when transitioning from tube to oral feeding [23]. This adds support to the 'packing' feeding behavior occurring in the CHARGE population, of which 90% or more are tube fed [6].

Previous studies on packing hypothesized that children with feeding problems may pack food because they do not have the oral

motor skills to consume more textured food (e.g., ground beef) [24]. They found that packing occurred more with high textured food (e.g., bread) than low texture food (e.g., puree) [24]. Parents reported this same phenomenon in our population of CHARGE syndrome. Difficulties with chewing and swallowing have been identified as severe oral motor skill deficits in this genetic condition [6,7]. These difficulties are thought to be due to cranial nerve abnormalities, which are a major feature of CHARGE syndrome [25]. Abnormalities of cranial nerve V and cranial nerves IX, X, and XI could contribute to dysphagia, abnormal chewing, packing and adverse feeding behaviors [6,25].

Packing feeding behaviors were present even in individuals who had never undergone G/J tube feeding or feeding therapy. This highlights the need to be aware of adverse feeding behaviors in all individuals with CHARGE syndrome, even those who had never previously presented with the need for feeding intervention. These problematic feeding behaviors were also seen in individuals with a wide range of texture tolerance, from those who only ate puree-textured foods, to those who ate all food textures. ENT is one of the most common medical specialties to follow individuals with CHARGE syndrome over their life, therefore they should be aware of the potential for these adverse feeding behaviors to occur in any individual with this genetic disorder [9].

Food packing has been identified as a feeding behavior that increases the risk of choking [12,16]. Previous studies on packing in individuals with autism noted aspiration may be a severe consequence of packing food in cheeks for extended periods of time [14,24,26]. Many parents in this present study expressed concern over their child's/adult's risk of choking while eating. Consequently, mealtimes require a large investment of effort, resources, and supervision for the individuals with CHARGE syndrome throughout their lives. In this study, mealtimes were often reported to take over an hour due to packing.

Adolescents may become defensive about the lack of the resultant feeding independence. Strategies to alleviate these feeding behaviors may help reduce the need for close supervision

Table 6Common presenting features of feeding difficulties in CHARGE syndrome^a.

Suggestive symptoms/signs of feeding difficulties
Prolonged mealtimes (>45 min)
Very fast mealtimes (<10 min)
Failure to advance food textures (e.g., purees to solid foods)
Necessity of close supervision while feeding
Necessity of a reminder or prompt to swallow when feeding
Necessity of a liquid chaser during feeding
Organic red flags^b
Dysphagia
Weak suck/swallow
Gastroesophageal reflux
Aspiration
Behavioral red flags^b
Over-stuffing one's mouth with food
Food packing
Holding food in cheeks after a meal has finished
Coughing or choking during eating

^a Adapted from Kerzner et al. [23].

^b Suggesting a need for feeding therapy intervention.

and increase independence and autonomy while eating as the individual ages. Stroke patients often suffer from similar dysphagia and feeding difficulties due to neurological and cranial nerve dysfunction [27]. Research has shown that neuromuscular electrical stimulation has been shown to improve their feeding difficulties and aspiration, and lead to increased autonomy while eating [28,29]. These techniques may be applicable to individuals with CHARGE syndrome, and warrant future research.

Food packing may also increase the risk of tooth decay and cavities. Little is known about dental issues in CHARGE syndrome, but periodontal disease caused by poor oral hygiene has been recently identified as a common problem in this genetic condition [30]. Periodontal disease can lead to pain with eating and can further exacerbate poor chewing skills in children with genetic syndromes [16]. Food packing can contribute to poor oral hygiene, especially in cases of food staying hidden in cheeks for hours after a meal. Parents should regularly check their child's/adult's cheeks and mouth or have the child/adult check their own cheeks and mouth after a meal to ensure there is no remaining packed food. Combined with an oral aversion to brushing ones' teeth, which is common in individuals with CHARGE syndrome, food packing may lead to poor dental hygiene [6].

Tongue and oral cavity dysfunction has been identified as a major source of feeding problems in children with genetic syndromes. Specifically, abnormal tongue movement has been identified as a cause of food packing in Down syndrome [16]. Abnormal tongue movement has been linked to poor sucking skills and choking, and macroglossia has been linked to oral food loss and difficulties swallowing in children with genetic syndromes [16]. Tongue dysfunction has not been well described in CHARGE syndrome, yet individuals are well known to have hypotonia and cranial nerve dysfunction, including cranial nerve XII (hypoglossal nerve), which is responsible for tongue movement [6,7,25]. Many of the parents in this present study described tongue abnormalities such as impaired mobility and ankyloglossia. In the individuals with ankyloglossia, food packing continued to be a problem even after surgical correction. Tongue mobility abnormalities, macroglossia, and ankyloglossia may cause or exacerbate problematic feeding behaviors such as food packing, and may often be overlooked due to the multitude of serious health conditions these individuals face.

Craniofacial abnormalities and other dysfunctions seen in CHARGE syndrome, including gastroesophageal reflux, periodontal disease, oral hypotonia, and uncoordinated suck and swallowing, have also been identified as causes of retention of food in children with genetic syndromes [16,31]. This present study population had a much higher prevalence of cleft palate/lip than has previously been reported in the general CHARGE syndrome population [4]. This midline craniofacial abnormality may interfere with normal feeding development and result in a weak or ineffective sucking ability [4,7,31]. Our study population also reported sub-mucosal clefts and high palates, with several individuals packing food up into their high palate in addition to their cheeks. These subtler midline abnormalities may often be overlooked. Other structural defects of this syndrome such as choanal atresia/stenosis and tracheoesophageal fistula could interfere with sucking and swallowing during feeding, and may contribute to food packing behaviors [6,16,31]. A clinician that deals with midline abnormalities may be an important addition to the multidisciplinary management of these individuals.

Complex feeding difficulties in children have been most commonly classified as a combination of behavioral, neurological, and structural problems [23]. Individuals with CHARGE syndrome experience issues with physical structures, oral-motor skills, and sensation [3,4]. Cranial nerve abnormalities are commonly seen in the CHARGE syndrome population [6,25]. In particular, cranial nerve VII (facial nerve) dysfunction is seen in over 50% of individuals [4]. Cranial nerve dysfunction can lead to problems

with auditory, olfaction, and swallowing function in this population [25]. Abnormal cranial nerve function contributes to many of the motor and sensory impairments in CHARGE syndrome, and is thought to play a major role in feeding difficulties [6,25]. In this present study, cranial nerve dysfunction was reported in 95% of the individuals with CHARGE syndrome who were experiencing abnormal feeding behaviors. This study adds support to the hypothesis that cranial nerve dysfunction in this genetic disorder plays a crucial role in feeding difficulties experienced [25,32]. We hypothesize that individuals with CHARGE syndrome may engage in food packing and adverse feeding behaviors (e.g., over-stuffing one's mouth) as a way to receive increased sensory feedback.

Lower motor neuron cranial nerve weakness is likely a contributing factor in food packing, as they control both oral cavity sensation and muscle movement. Cranial nerve VII (facial) is one of the most commonly affected nerves, manifesting in facial palsy and/or weakness in up to 90% of individuals, and previous imaging has shown that this nerve has an abnormal course in individuals with CHARGE syndrome [4,25]. Cranial nerve V (trigeminal), innervating the muscles of mastication, may also interfere with proper chewing and food bolus movement. Cranial nerves IX and X (glossopharyngeal and vagus) are also hypothesized to contribute to the highly prevalent swallowing difficulties reported in this present study and previous studies in CHARGE syndrome and other congenital genetic syndromes [6,25,31]. A full examination of cranial nerve function in these individuals is warranted. Cranial nerve weaknesses combined with a behavioral etiology, or either occurring in isolation, are potential causes of food packing in this syndrome.

Previous studies have indicated that feeding difficulties in CHARGE syndrome tend to improve with age [6]. However, the definition of feeding difficulties has often included only the necessity for tube feeding, the risk of aspiration, and the presence of gastroesophageal reflux. We recommend that packing and other problematic feeding behaviors be added to the definition of the feeding difficulties experienced in individuals with CHARGE syndrome. Many of the individuals in this study were no longer tube fed, or on medication to treat reflux, and had safely progressed to oral feeding with complex food textures (i.e., mashed, lumpy, and chewable).

Current evidence on treatment of packing feeding behaviors highlights the need to combine procedures. Simple interventions such as using a 'Nuk' brush instead of a spoon to eat has been found to reduce packing [33]. One study found that using a toothbrush or flipped spoon to re-distribute (removing packed food from the mouth and placing it back on the tongue) and facilitate swallowing reduced packing [34]. Another study found that using a chaser, a liquid or solid to consistently swallow with each bite, reduced packing behaviors [13]. Combining all three interventions of re-distribution, swallow facilitation, and a chaser has been shown to be effective [12]. These interventions may help reduce packing and problematic feeding behaviors in CHARGE syndrome.

These feeding behaviors can affect individuals with CHARGE syndrome at any age in their life. If food packing presents early and it is not addressed then it can continue to persist despite undergoing feeding therapy for other feeding difficulties. Almost all individuals in this present study had undergone feeding therapy with a speech-language-pathologist, occupational therapist, or psychologist, therefore raising awareness of food packing feeding behaviors in CHARGE syndrome can help improve prognosis. Packing can also occur when individuals may have a fear of feeding due to an episode of choking, a painful oral procedure, transitioning from tube-feeding, introduction of oral foods, or in those who lack experience with oral feeding [23]. Therefore, packing may improve with time if other existing feeding difficulties are also improved. Severe feeding difficulties can lead to mortality, but this may manifest more in individuals at the extreme end of the

spectrum [1,17]. If food packing and other adverse feeding behaviors are identified, treatment options such as behavioral treatment, feeding therapy including a focus on tongue mobility, and nerve stimulation are hypothesized to reduce these feeding difficulties and avoid associated adverse outcomes such as poor oral hygiene, choking, and aspiration.

5. Limitations

The present study relied on parental recall of their child/adult's feeding difficulties so there is a possibility of recall bias and recall errors. However, many of the child/adult's feeding difficulties were ongoing, so details were likely salient. Although this study involved a relatively small sample, interviews were performed until no new information or themes were acquired, thus reaching data saturation. Parents of individuals with more severe feeding difficulties may have been more inclined to participate. However, our final study sample showed a wide range of feeding difficulties, including individuals with very mild difficulties who never required tube-feeding or feeding therapy as well as those with very severe feeding difficulties. The clinical features were representative of the general CHARGE syndrome population with the exception of increased frequency of individuals with cleft palates. The type and prevalence of feeding difficulties that the individuals with CHARGE syndrome faced in addition to their food packing were similar to those that have been reported in previous studies, indicating that this study sample likely was a representative population [6,7].

Future directions

Future research should explore the efficacy of feeding therapy interventions on reducing packing in individuals with CHARGE syndrome. In addition, future research should examine whether altering the time it takes an individual with CHARGE syndrome to finish eating a typical meal would be beneficial in reducing these problematic feeding behaviors. Caregiver anxiety in parents of individuals with CHARGE syndrome who experience feeding difficulties should be explored.

6. Conclusion

This study provides a deeper understanding of packing and other problematic feeding behaviors in individuals with CHARGE syndrome. The information from this study can be useful for pediatric otorhinolaryngologists and feeding therapy teams who are often involved in the care of these individuals starting in infancy. These problematic feeding behaviors can have severe health consequences such as choking and tooth decay thus should not be overlooked despite the multitude of health issues children and adults with CHARGE syndrome experience. This study provides the parents' perspectives on their child and adult's oral packing behaviors, the types of difficulties they encounter, and the mealtime interventions they try at home and at school. The information from this study can help inform individualized feeding evaluations and interventions to prevent food packing and other adverse feeding behaviors.

Conflict of interest

The authors declare no conflict of interest.

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Appendix A

Demographics survey

(1) Demographic Information

Name of individual with CHARGE syndrome:

Date of birth (dd/mm/yyyy):

Age:

Gender (male/female):

Age at diagnosis:

Ethnicity:

Country you live in:

(2) CHARGE Syndrome Characteristics Major Features: Please circle the appropriate answer

Gene CHD7 demonstrated CHARGE Syndrome

Yes

No

Not tested

Coloboma of the iris

Yes, both sides

Yes, one side

No

N/A

Coloboma of the retina

Yes, both sides

Yes, one side

No

N/A

Choanal atresia (complete nasal obstruction)

Yes, both sides

Yes, one side

No

N/A

Choanal stenosis (narrowing of nasal passage)

Yes, both sides

Yes, one side

No

N/A

Ear anomalies

External ear (floppy, lop shaped): yes/no/N/A

Middle ear (bones, infections): yes/no/N/A

Inner ear (sensory neural hearing loss): yes/no/N/A

Abnormal CT/MRI scan of temporal bone (semicircular canals):
yes/no/not tested

Cranial nerve dysfunction

C1 (difficulty smelling): yes/no/N/A

C7 (facial palsy/weakness): yes/no/N/A

C8 (hearing problems): yes/no/N/A

C9, 10, 11 (swallowing problems): yes/no/N/A

(3) CHARGE Syndrome Characteristics Minor Features: Please circle the appropriate answer

Heart defect major (requiring surgery): yes/no

Heart defect minor (patent ductus arteriosus or small hole in heart ventricular septal defect): yes/no

Cleft lip: yes/no

Cleft palate: yes/no

Genital hypoplasia: yes/no
 Growth deficiency: yes/no
 Developmental delay: yes/no
 Tracheoesophageal fistula: yes/no
 Distinct CHARGE facial features: yes/no
 Urinary tract problems: yes/no

Appendix B

Interview Questions

- (1) Eating
 - (a) Please describe how the eating process has changed for your child from infant to now.
 - (b) Please describe your child's transition from tube feeding to oral feeding and the age of tube removal. Please indicate how long your child was on tube feeding (nasogastric or G/J tube feeding) and the challenges of the transition.
 - (c) Please describe any oral aversion or defensiveness your child displays to oral feeding (i.e., Not accepting the bottle, cup or spoon).
 - (d) Does your child ever prefer to eat non-food items instead of food? (i.e., Preferring objects, rocks, or dirt rather than food).
 - (e) What amount of your time does your child require, on average, to finish eating a typical meal?
 - (f) Please describe any specialized or modified utensils your child uses during feeding.
 - (g) Does your child require any assistance during the eating process? (i.e., Putting food in their mouth, cutting their food, etc.)
 - (h) Does your child ever have to be told to chew or swallow during the eating process?
 - (i) Does food ever fall out of your child's mouth while they are eating?
 - (j) Does your child ever eat a meal while unattended?
- (2) Mouth over-stuffing during eating
 - (a) Some parents report over stuffing of their child's mouth, odd eating behaviors, and choking. Please describe any of these experiences with your child.
 - (b) Has your child had any difficulties related to eating due to the amount of food they put in their mouth?
 - (c) Has your child ever stored food in his/her cheeks (pocketing of food) while eating?
 - (d) If your child has ever pocketed food in his/her cheeks while eating, can you please describe the texture and size of the food they pocketed?
- (3) Chewing
 - (a) Please describe any challenges with chewing during eating experienced throughout your child's life.
 - (b) Please describe any eating challenges your child experiences with the following food textures: liquids, purees, mashed/lumpy, and chewable/solid.
- (4) Choking
 - (a) Has your child ever experienced choking while feeding? Please describe the severity and frequency of choking episodes while eating throughout your child's life.
- (5) Swallowing
 - (6) Please describe any swallowing difficulties your child experienced during eating throughout your child's life.
- (7) Therapy
 - (a) Has your child received any therapy from a speech language pathologist for eating issues? If so, please describe the therapy that was done and the results (i.e., symptom relief, complications, etc.).
 - (b) Has your child received any therapy from an occupational therapist for eating issues? If so, please describe the therapy

that was done and the results (i.e., symptom relief, complications, etc.)

- (c) Has your child received any therapy from a psychologist for eating issues? If so, please describe the therapy that was done and the results (i.e., symptom relief, complications, etc.)
- (d) Has your child undergone any surgeries to better manage his/her swallowing or eating? If so please describe what procedures have been done, the date of the procedure, and the results (i.e., symptom relief, complications, etc.)
- (e) Please describe the top three interventions that you use during meals to aid your child's eating process (i.e., Hold a mirror in front of them while eating, etc.).
- (f) Do you ever remove the food from your child's cheeks and place it on their tongue to help the eating process?
- (g) Do you ever give your child a liquid or solid to swallow consistently to reduce mouth over-stuffing/pocketing of food during the meal (i.e., a chaser)?
- (h) Please describe any medications your child has taken to treat gastro-esophageal reflux (i.e., reflux acid suppressant) and the time period they were taken.
- (i) Has your child benefited from any therapy to reduce saliva? (i.e., Botox injections)

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