Case Report

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Interdisciplinary approach in the treatment of macroglossia in Beckwith-Wiedemann syndrome: case report

Abordagem interdisciplinar no tratamento da macroglossia na síndrome de Beckwith-Wiedemann: relato de caso

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ABSTRACT

This study aims to report the interdisciplinary management of macroglossia in a Beckwith-Wiedemann syndrome patient during ten years. Clinical follow-up started by the Oral and Maxillofacial Surgery team, followed by Speech Therapy due to feeding difficulties. After clinical and instrumental evaluation, at 8 months old, the speech therapy intervention was indicated, focusing on oropharyngeal dysphagia and orofacial myofunctional therapy. At 1 year and 11 months, no signs of swallowing alteration in the pharyngeal phase and improvement in the posture of the lips and tongue were found. At the age of 3, stimulation to remove oral habits and train masticatory function were initiated. Orthodontic treatment to correct anterior open bite and unilateral posterior crossbite started at age 6. At 7 years and 5 months, there was stability in the nasal breathing mode and adequacy of resting posture of lips and tongue. At the age of 9, due to relapse of the occlusal alterations, surgical reduction of the tongue was indicated, followed by orofacial myofunctional therapy, restarted at the age of 9 years and 3 months. The result was the correction of the posture of the tongue during swallowing and speech adequacy. The association of treatments involving Speech Therapy, Orthodontics and Oral and Maxillofacial Surgery was considered effective in the management of the macroglossia. It resulted in the adequacy and equilibrium of orofacial functions.

Keywords: Beckwith-Wiedemann syndrome; Tongue diseases; Macroglossia; Speech, Language and Hearing Sciences; Orthodontics; Glossectomy

RESUMO

O objetivo deste estudo foi relatar a abordagem interdisciplinar no manejo da macroglossia em um caso de paciente com síndrome de Beckwith-Wiedemann, no período de dez anos. O acompanhamento iniciou pela equipe de Cirurgia Bucomaxilofacial, seguido da Fonoaudiologia, em função de dificuldades alimentares. Após avaliação clínica e instrumental, aos 8 meses de idade, iniciou-se a intervenção fonoaudiológica com foco na disfagia orofaríngea e na terapia miofuncional orofacial. Foi verificado, com 1 ano e 11 meses, ausência de sinais de alteração de deglutição em fase faríngea e melhora na postura de lábios e língua. Aos 3 anos, foram iniciados estímulos para retirada dos hábitos orais e o treino da função mastigatória. O tratamento ortodôntico para correção de mordida aberta anterior e mordida cruzada posterior unilateral iniciou-se aos 6 anos. Aos 7 anos e 5 meses de idade, constatou-se estabilidade do modo respiratório nasal e adequação da postura de repouso de lábios e língua. Aos 9 anos, em função de recidiva das alterações oclusais, optou-se pela redução cirúrgica da língua seguida de terapia miofuncional orofacial, retomada aos 9 anos e 3 meses. O resultado foi a correção da postura da língua na deglutição e a adequação da fala. A associação dos tratamentos, envolvendo Fonoaudiologia, Ortodontia e Cirurgia Bucomaxilofacial foi considerada efetiva no manejo da macroglossia, resultando na adequação e equilíbrio das funções orofaciais.

Palavras-chave: Síndrome de Beckwith-Wiedemann; Doenças da língua; Macroglossia; Fonoaudiologia; Ortodontia; Glossectomia

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INTRODUCTION

Beckwith-Wiedemann syndrome (SBW) is the most common congenital disorder of human growth. It consists of a genomic disorder with a multisystem imprint, varied clinical expression, and complex molecular etiology. It is associated with epigenetic abnormalities in the region of the 11p15 chromosome, which contains imprinted genes that regulate fetal growth. The alterations are often of the mosaic type[1,2], resulting in a spectrum ranging from the classic manifestation, with signs such as macroglossia, omphalocele, and defects in the abdominal wall, to isolated cases of body hemi-hyperplasia or hemi-hypertrophy[2]. Diagnosis is based on a scoring system of cardinal or suggestive clinical features assigned with different predictive values. The most common epigenetic mechanism involves the loss of methylation in IC2 (imprinting center 2), representing the molecular etiology in 50-60% of cases. The incidence is 1:10,000 / 10,500 live births[1,2].

Although macroglossia is the most frequent sign in SBW, a few studies[3-5] have analyzed its impact on the orofacial myofunctional condition and dentofacial characteristics in the long term. Along with other structures of the stomatognathic system, the tongue plays a primary role in vital functions, such as sucking, chewing, and swallowing, as well as in the adapted function of speech. The larger size of the tongue, alterations in the usual posture, and the pressure exerted by the musculature can impair the performance of orofacial functions and dental occlusion[6]. The treatment for macroglossia involves professionals from different areas of Medicine, Dentistry, and Speech-Language Therapy. The criteria that may lead to the indication of surgical tongue reduction (STR) are upper airway compromise, persistent eating difficulties and tongue protrusion, dental or skeletal malocclusion, speech distortions, and psychological impact[2,7].

This study aimed to address the interdisciplinary approach for treating macroglossia for over ten years in a patient who had SBW, covering speech-language therapy, orthodontic intervention, and STR.

CASE PRESENTATION

The caretaker signed the Informed Consent Form and consented to the performance and dissemination of the research and respective results. This case report is part of a larger study, approved by the Research Ethics Commission in Health at the Federal University of Rio Grande do Sul, protocol number 1.472407.

The patient is a male, born at term who presented signs compatible with the clinical diagnosis of SBW, according to the information in the hospital chart: weight above average, macroglossia, omphalocele, and neonatal hypoglycemia. Since his early months, the patient has been monitored by professionals from the areas of Pediatrics, Genetics, Pneumology, Gastroenterology, and Oral and Maxillofacial Surgery. The patient received speech-language therapy from 8 months old until about 9/10 years old, with some interruptions. Chart 1 shows the main objectives, conducts, and results of the speech-language therapy treatment in five clippings over time. The case is reported in five steps, encompassing the follow-up of the Speech-Language Therapy, Orthodontics, and Oral and Maxillofacial Surgery teams.

At 8 months old, the patient was referred for speech-language evaluation due to eating difficulties. It is noteworthy that the indication of a potential STR was conditioned to the responses to speech-language therapy and orthodontic treatment.

Since there was no specific protocol validated for babies at the time, the initial assessment was based on the Orofacial Myofunctional Evaluation with Scores (OMES)[8]. The appearance and postural conditions of the orofacial structures showed no lip occlusion (severe dysfunction); vertical jaw posture with excessive mouth opening; symmetric face; cheeks with increased volume; hard palate without alterations; and tongue interposed between the dental arches with excessive protrusion due to macroglossia. Muscle flaccidity of the lips, tongue, and cheeks were also observed. Presence of digital and pacifier-sucking habits and oronasal breathing. When trying to crush solid foods, he kneaded them, pressing the tongue against the hard palate, which is expected considering that the masticatory function at 8 months old has not yet matured. The clinical evaluation of swallowing revealed suggestive signs of laryngeal penetration and/or tracheobronchial aspiration with liquid. The patient was referred for swallowing videofluoroscopy and otorhinolaryngological evaluation. No obstructive changes were observed in the upper airway. The functional opinion of the videofluoroscopy indicated oropharyngeal dysphagia for liquids, audible aspiration with the use of a high-flow utensil (a cup with a spout), and laryngeal penetration with the use of a bottle.

The evaluations were followed by the speech-language therapy focusing on the treatment of oropharyngeal dysphagia and passive techniques of orofacial myofunctional therapy (OMT).

At the age of 1 year and 11 months, the patient underwent an orofacial myofunctional reassessment. Greater muscle tension of the cheeks, lips, and tongue was observed, which allowed for an improvement in the postural condition of these structures. Although the anterior projection of the tongue during swallowing was observed, clinical signs of dysphagia in the pharyngeal phase were no longer present. The second videofluoroscopy, performed at 2 years and 8 months old, showed adequate oropharyngeal swallowing. Figure 1 shows images of the patient at the time of the initial assessment and after the first phase of the speech-language therapy intervention.

Based on the advances obtained with OMT and the possibility that the oral cavity supports the size of the tongue as it grows and develops, the Oral and Maxillofacial Surgery and Speech-Language Therapy teams decided to continue with the speech-language therapy without performing STR at this follow-up stage. At 3 years old, the speech-language therapy focused on the withdrawal of sucking habits Given the consequences on the orofacial musculature and dental occlusion, at 3 years of age, the speech therapy focused the withdrawal of sucking habits and stimuli for proper chewing.

In this phase, only one professional carried out the speech-language therapy, which continued without interruption until the beginning of the fifth year of life, when it was suspended for an orthodontic evaluation. No therapeutic intervention was performed until the start of orthodontic treatment.

The orthodontic treatment carried out at the same institution as the speech-language therapy intervention began after the interruption of the digital sucking habit, which only occurred at 6 years old.

Chart 1. Speech-language pathology intervention over time

Initial age	Main goals	Conducts	Results
8 months old	- To promote the effectiveness of swallowing protecting the airways. - To improve coordination and muscle tension and enhance the posture of orofacial structures (lips, tongue, and cheeks).	- Use of thin and thickened liquid with different bottle nipples tested during videofluoroscopy, and rhythm control (a pause every eight suctions). - Passive techniques of orofacial myofunctional therapy.	- Lower signs of swallowing alteration in the pharyngeal phase. - Greater muscle tension orofacial, possibility of lip seal at rest, and improved tongue posture.
1 year and 11 months old	- To improve the performance of chewing and swallowing functions.	- Intake of liquid from a glass with rhythm control and application of mandibular control maneuver. - Guidelines for posture and utensils during feeding, and stimulation of chewing through solid foods.	- No signs of swallowing alteration in the pharyngeal phase. - Possibility of sealing lip while chewing some solid foods. - Improved posture of lip, tongue, and cheeks.
3 years old	- To terminate oral habits (digital suction, pacifier, and bottle). - To encourage alternating bilateral chewing. - To stimulate the acquisition of speech sounds.	- Use of recreational resources to terminate oral habits and replace the bottle with a conventional cup. - Training of masticatory function through solid foods. - Use of pictures and children's books to stimulate the acquisition of sounds, according to the order of phonological acquisition.	- Intake of liquid from a conventional glass. - Mastication with seal labialis and the beginning of the lateralization of the foods. - Appropriate phonological acquisition for chronological age; but some sounds were produced with the anterior projection of the tongue.
7 years and 5 months old	- To improve coordination and muscle tension, and adjust the posture of the lips and tongue during rest. - To stimulate nasal breathing.	- Isometric exercises for lips and tongue. - Stimuli to adapt the usual posture of the tongue and lips. - Awareness and training of nasal breathing mode.	- Postural adequacy of lips and tongue at rest. - Consolidation of the mode nasal breathing.
9 years and 3 months old (post-surgical intervention)	- To stimulate language mobility. - To adequate the posture of the tongue in swallowing and speaking. - To reduce headphone distortion [ɾ].	- Dynamic tongue exercises (isotonic). - Swallowing training with different consistencies. - Training in the correct production of the sounds [t] and [d] and the sound [ɾ] in different syllabic positions.	- Appropriate tongue movements. - Adequacy of tongue posture during swallowing and speech. - Fixed sound distortion [ɾ].

Figure 1. Speech-language therapy pre-intervention at 8 months old and after the first stage of speech-language therapy at 1 year and 11 months old

During this period, the patient began to be monitored by the Speech-Language Therapy team at the Orthodontics clinic to determine the ideal time to resume speech-language therapy. The initial orthodontic evaluation detected a mild facial asymmetry, with functional mandibular deviation to the right, right unilateral posterior crossbite, and anterior open bite of about 6 mm. A rapid expansion of the maxilla was performed to correct the posterior crossbite and improve the shape of the upper arch, followed by two attempts to treat the anterior open bite with the lingual arch device with tips. However, the patient did not adapt to it, and fractures at the ends and damage to the device were observed. Therefore, orthodontic buttons were glued directly on the lingual surface of the lower incisors.

At that time, when the patient was 7 years and 5 months old, speech-language therapy was resumed, now focusing on correcting the posture of stomatognathic system structures. This step of the speech-language therapy was carried out by another professional from the Speech-Language Therapy team. The main advancement of OMT, in this phase, was the adequacy of the posture of the lips and tongue during rest (anterior third touching the region of the incisive papilla) and the consolidation of the nasal breathing.

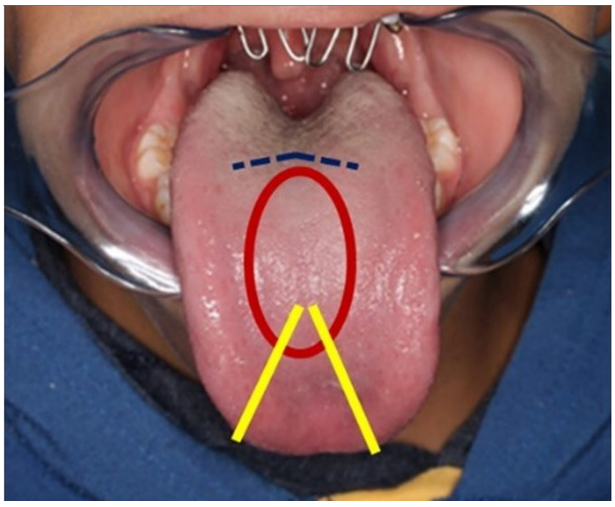

Figure 2. Macroglossia – preoperative image (8 years and 3 months old). Composition of the elliptical (red). and triangular or wedge (yellow) incisions, from the formation of the vallate taste buds (blue) to perform the surgical reduction
Source: Puricelli et al.[10].

However, the volume of the tongue influenced the automation of the correct posture while performing the functions, thus impairing the stability of the results of the orthodontic treatment.

At 8 years and 3 months old, an interdisciplinary reassessment with the Oral and Maxillofacial Surgery, Orthodontics, and Speech Therapy teams allowed the professionals to decide to perform the STR. The surgical intervention was performed after orthodontic correction of the anterior open bite and crossbite. A breaker associated with a palatal crib was used due to the high and forward position of the tongue at rest. From the interdisciplinary reassessment to the moment of the STR, the patient was followed up again by the Speech-Language Therapy team at the Orthodontics clinic.

When the child turned 9 years old, the STR was performed by the Pediatric Oral and Maxillofacial Surgery team at the Santo Antônio Children's Hospital, *Santa Casa de Misericórdia de Porto Alegre*. The purpose of the surgical intervention is to reduce the dimensions of the tongue preserving its shape and functions (motor and sensitive) as much as possible. In the preoperative phase, the analysis of the topographic anatomy of the anterior two-thirds of the tongue must consider that each vascular territory is unilateral and dependent on segments of the tongue artery[9]. Figure 2 illustrates the tongue in the preoperative phase showing the incisions made in the STR.

When the patient was 9 years and 3 months old, a post-surgical speech-language evaluation was performed and the speech-language therapy started. At that time, the consultations were again performed by the speech-language therapist who had started the treatment at 8 months old. The following main aspects were observed in the evaluation applying the AMIOFE protocol[8]: tongue mobility difficulty, mainly lateralizing and raising the apex; tongue posture interposed between the dental arches (dysfunction) during swallowing and alternating bilateral chewing, but with disorganized movements. Regarding speech, after the STR, the patient stopped projecting his tongue during alveolar and palatoalveolar sounds. However, tongue projection continued to occur in the lingual phones [t] and [d]. Distortion of the phone [ɾ] was also observed in all positions, in addition to the omission in some productions involving complex onset.

After 13 speech-language therapy sessions carried out in three months, the patient was reassessed using the same protocol. Tongue mobility was recovered and swallowing and chewing functions became adequate. Figure 3 shows an image of the patient in the frontal view, occlusion, and protruding tongue, one month after the start of speech-language therapy in the postoperative period.

Figure 3. Image in frontal norm, occlusion, and protrusion of the tongue three months after the surgical intervention and one month after the beginning of speech-language therapy (9 years and 4 months old)

Speech alterations were also corrected, except for the phoneme /ɾ/ which, at this moment, was still occurring with distortion. It is important emphasizing that the first three sessions were held in person and the others took place through synchronous teleconsultations. The Telephonoaudiologia resource was used due to the social isolation imposed by the Covid-19 pandemic, which resulted in the suspension of face-to-face consultations. After three months of weekly sessions, the patient began to be monitored monthly. The distortion in the phoneme /ɾ/ was corrected through speech-language therapy six months after the surgery.

DISCUSSION

The macroglossia present in the SBW is recognized and related to tissue overgrowth, which may result in insufficient space in the oral cavity and functional changes[11]. The larger size of the tongue can cause, to a greater or lesser extent, obstructive sleep apnea, dysphagia, sialorrhea, dentofacial and speech alterations, in addition to psychosocial problems[1,2,7,12]. The findings of our initial speech-language evaluation corroborate a previous study[6] with most children presenting impairments in the oral swallowing phase with three consistencies tested and risk of aspiration with liquids. Speech-language therapy intervention through compensatory techniques, such as guidance related to posture, reduction of liquid flow, and modification of textures, are essential to reduce the risk of aspiration, thus improving the feeding of babies[6].

During the early years of life, some authors have suggested OMT as an initial conduct when treating the impact of macroglossia on the orofacial myofunctional[4,13]. The objective should be the acquisition of orofacial praxis and the adequacy of the posture and tonus of the phonoarticulatory organs, aiming to provide satisfactory performance in oral functions[13]. However, only a few studies have addressed less invasive procedures[4,13]. One of the factors that may lead to the indication of STR in older patients is the relapse of occlusal alterations. However, it is important emphasizing that the indication of malocclusion is often relative and not definite[7].

In this study, the habit of prolonged digital sucking favored the development and maintenance of malocclusion. Oral habits cause muscle imbalances and changes in the posture of the structures involved[14]. The absence of tongue contact with the hard palate and the force exerted by the buccinator muscles on the dental arches during the act of suction can cause a lower transverse dimension of the maxilla, resulting in a posterior crossbite[14]. The lower width of the dental arch can also restrict the vertical maxillary development, reducing the overbite and favoring the formation of an anterior open bite[15]. The impact of the prolonged sucking habit and the tongue volume in the oral cavity during the mixed dentition phase might have hindered the results of the speech-language therapy and orthodontic interventions in the long term.

The main occlusion alterations resulting from macroglossia described in the literature are dental Class III, anterior open bite, and mandibular prognathism[7]. In the case presented, the main alterations observed were anterior open bite and bilateral posterior crossbite. The relapse of these occlusal alterations was the main factor for indicating the surgery when the patient was 9 years old. A retrospective study analyzed the relationship between age at surgery and the evolution of anterior open bite in children with SBW[7]. The mean age for an indication of malocclusion in this study was 5.1 years. The two cases described, like this study, underwent the surgery later, when the patients were 13 years old. The authors point out that the ideal age for the indication of STR is still controversial. However, they conclude that the indication of surgery provides good long-term results in the treatment of anterior open bites. Other studies have also shown improvements in speech production with the performance of STR[3,16].

STR is concentrated in the anterior two-thirds of the tongue (free body). Manipulation limited to the dorsal surface with less invasion of the ventral surface is recommended to avoid vascular and nervous injury (lingual and hypoglossal). The vallate papillae limit the posterior advance, both of the incision and of the excision[11]. The basic proposals involve wedge-shaped incisions at the vertex of the tongue and elliptical (oval) in the center, involving the midline, or a combination of both[11]. Through basic surgical techniques, the three-dimensional knowledge of the anatomical territories (muscles, osteofibrous skeleton, vessels, and nerves) allows for creating individualized variants applicable in the apex (length), in the dorsum (width), or both, with predictable depth (thickness). It is worth pointing out that the patient did not report changes in taste after the procedure. Other studies have also reported the preservation of general and gustatory sensitivity in patients submitted to STR[12,16].

After surgery, our patient showed spontaneous improvement in some speech sounds and stopped projecting his tongue on the sounds [s] and [z], remaining the projection on the sounds [t] and [d], which was corrected through postoperative speech-language therapy intervention, with weekly sessions for three months. The distortion in the sound [ɾ] appeared after the surgical procedure and ceased six months after the STR through speech-language therapy. Similar results were reported in a study with a series of ten cases evaluating speech before and after the surgical procedure. Among the participants, three presented speech without distortions after surgery, one child remained with tongue projection in the sounds [s], [t] and [d], and six presented only mild distortions after surgery[16]. It is known that STR can affect the sounds produced by the apex of the tongue against the upper or lower teeth, or the anterior palate[16], which explains the temporary distortion in the production of the alveolar flap [ɾ].

In the post-surgical period, OMT is important to assist in the recovery of movement and functional rehabilitation[3]. The adequacy of orofacial functions is necessary for a prognosis for the results of orthodontic treatment.

More recently, the literature shows only two case reports of patients undergoing STR with a follow-up of over ten years. One study described the follow-up of a patient for 15 years highlighting the importance of managing maxillomandibular growth for establishing a treatment plan with an interdisciplinary team[4]. The other report described the follow-up of a case for 30 years suggesting that the balance of structures and functions of the orofacial complex is the key to satisfactory results and long-term stability[5].

FINAL COMMENTS

In the reported case, performing STR with pre- and postoperative speech-language therapy and orthodontic intervention was essential for adapting the posture of the tongue during functions, which can promote greater stability to the results of orthodontic treatment in the long term. The interdisciplinary treatment, involving Speech Therapy, Orthodontics, and Oral and Maxillofacial Surgery, was effective in managing macroglossia, resulting in the adequacy of orofacial functions. However, it is important to carry out cohort studies with a larger sample size for a more comprehensive understanding of the treatment of the clinical repercussions of SBW on the stomatognathic system.

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