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CASE REPORT

Assessment of the tongue frenulum in Beckwith-Wiedemann syndrome: Pre- and post-frenectomy findings

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Abstract

Aim: To report the pre- and post-frenectomy findings of a patient with Beckwith-Wiedemann syndrome.

Methods and Results: Clinical case report of a patient with a confirmed genetic-molecular diagnosis of the referred syndrome. The minor was evaluated and reassessed by the protocol for the evaluation of the tongue's frenulum for babies in two moments: pre-surgical and 2 months after the frenectomy. The surgical procedure was performed using the traditional technique and, after the procedure, the minor was breastfed and received photobiomodulation with a red laser. The minor obtained 16 points in the neonatal tongue screening test, indicating the need for a frenectomy. Thus, she was referred to a dentist for surgery. After the surgical procedure, macroglossia was observed as a maternal complaint (previously not mentioned). The wound healing was satisfactory, and the total score obtained in the reapplication of the protocol (five points) showed functional results of improvements in sucking and tongue mobility, justifying the importance of the frenectomy.

Conclusion: Frenectomy, despite showing macroglossia related to the Beckwith-Wiedemann syndrome, allowed anatomical and functional advances of the tongue in the present clinical case.

KEYWORDS
oral health, oral motor function, oral surgery

1 | INTRODUCTION

Beckwith-Wiedemann syndrome (BWS) (OMIM #130650) was described by Beckwith in 1963 and by Wiedemann in 1964, thus giving rise to the name of the syndrome.[1] It is a genetic condition characterized by overgrowth in the presence of congenital anomalies.[2,3] There is also a predisposition to germ cell tumors and autosomal recessive diseases.[4]

BWS is caused by an alteration in the short arm of chromosome 11 (11p15) and manifests as a triad comprising omphalocele (congenital umbilical hernia), gigantism, and macroglossia.[5,6] Omphalocele is evident on fetal ultrasound examination at the onset of the prenatal period.[7] Macroglossia is the most common manifestation of the syndrome and is present in 82%–99% of affected individuals, and may also be associated with the presence of craniofacial changes.[8] One literature report[9] described an individual with referred syndrome associated with ankyloglossia, submucosal fissure, and bifid uvula.

Macroglossia is associated with a variety of congenital syndromes and is regarded as confirmed in individuals with excessive enlargement of the tongue or when space in the oral cavity is insufficient for the organ. Macroglossia is a fundamental feature of BWS and can compromise function, including breathing, chewing, swallowing and speech, and facial aesthetics.[10,11] Treatment is usually interdisciplinary, involving various health specialties including pediatrics, orthopedics, cardiology, otolaryngology, dentistry, and/or speech therapy.[12] The present report describes findings before and after lingual frenectomy in a patient with BWS.

2 | MATERIAL AND METHODS

The present investigation was a descriptive, exploratory, retrospective, clinical case study based on the medical records of a patient with BWS. This study was approved by the local ethics committee (CAAE 20786719.1.0000.5546).

The clinical case involved a child who underwent interdisciplinary evaluation. The subject was female and was 9 months of age when she was diagnosed with BWS using molecular genetic techniques. According to a report from a geneticist, the patient presented with macrosomia, macroglossia, hemangioma on the face and scalp, umbilical hernia, furrows behind the ear, and recurrent hypoglycemia after birth. The mother experienced complications of pre-eclampsia during delivery, which was premature (32 weeks). The patient had a history of exclusive breastfeeding up to 6 months of age and was complementary to the present day. She was also administered vitamin D and ferrous sulfate supplements, and underwent multidisciplinary follow-up.

The child underwent speech screening of the sublingual frenulum, based on a validated protocol from the neonatal tongue screening test,[13] in which scores are attributed to clinical history (0–8 points) and anatomofunctional evaluation (part I, 0–12 points; part II, 0–5 points), with a total possible score of 25 points. Higher scores indicate a worse prognosis. After screening, the patient was referred to the school clinic of the specialization course in pediatric dentistry, where lingual frenectomy was performed. Because this location does not have a speech therapist on the team, the patient was referred to the speech therapy school clinic.

3 | RESULTS

At 9 months of age, the patient underwent a tongue frenulum assessment test. The guardian denied the existence of a family history of BWS; however, she reported that there were records of metabolic disorders (hypertension) and ankyloglossia (maternal grandfather of the patient). Regarding breastfeeding, particularly remarkable features included short intervals between breastfeedings (≤1 h), the child suckled "just a bit," then slept, and that she used to bite the nipple. The patient scored 5 points in this section of the test.

Clinical speech-language examination (total, 10 points) revealed open lips at rest and tongue positioned low during crying, with the tip of the tongue resembling the shape of a "heart." It was possible to visualize the sublingual frenulum with tongue elevation and posteriorization maneuvers. Furthermore, the examination revealed that the frenulum was thin and fixed at the apex of the tongue and its insertion between the caruncles.

On functional evaluation of suction (part II), it was not possible to observe non-nutritive sucking because the patient did not allow the therapist place her finger in her mouth. In nutritive sucking, the patient performed only a few suction movements with long pauses, with adequate coordination between sucking, swallowing, and breathing. Nipple bites and tongue clicks during sucking were not apparent, yielding a partial score of 1 point.

Of the total score (i.e., 25), the patient scored 16. Frenulum interference in tongue movement(s) can be considered in individuals scoring ≥ 13. Thus, dental evaluation was recommended for lingual frenectomy. At this assessment, the patient was 1 year, 2 months of age, 77-cm tall, and weighed 10 kg. Extraoral examination revealed an enlarged nasal dorsum with flattening of the alar cartilages, a shallow orbital floor, abnormal grooves in the earlobes, shortened middle-third of the face, mandibular and lingual projection, a mouth breathing pattern, and absence of lip sealing. Intraoral examination of soft tissues revealed an upper labial frenulum with low insertion, an interincisal diastema, ankyloglossia with thick and short lingual frenu-

lum, and macroglossia, with the tongue exhibiting teeth marks on the lateral edges.

On assessment of hard tissues, the patient exhibited incomplete primary dentition, an atresic upper arch, a parabolic and protruding lower arch, and anterior crossbite. The upper and anterior teeth exhibited extrinsic stains due to the use of ferrous sulfate and white stains related to initial carious lesions without cavitation. Before frenectomy, laboratory investigations, including blood count, prothrombin time, partial activation of thromboplastin time, and thrombin time, were all within normal limits. In addition, a cardiologist classified the patient as American Society of Anesthesiologists class I.

The lingual frenectomy procedure, which was performed 2 months after the evaluation, involved protective stabilization with pedi-wrap, head stabilization, and a mouth opener. Topical anesthesia with 20% benzocaine was applied using a flexible cotton swab, followed by infiltrative anesthesia at the base and apex of the tongue with 2% lidocaine diluted 1:100,000 with a vasoconstrictor using an anesthetic tube. Tongue removal was performed with the aid of the child's tentacannula and an incision made in the lingual frenulum in the anteroposterior direction using a No. 15 scalpel blade. Tissue divulsion was then performed using straight-tip scissors and simple suture with 5.0 resorbable thread. At the conclusion of the procedure, the patient was breastfed, and photobiomodulation using a red laser was performed (energy 2 J; power, 100 W; application time, 20 s per point).

In the postoperative period, ibuprofen (100 mg/ml) was prescribed, with 5 drops administered every 6 h for 24 h, as per post-surgical recommendations. She was then referred to speech therapists who reviewed the frenectomy and requested a return to speech therapy sessions. In the dental evaluation after surgery, proper healing and improved mobility of the tongue were noted. Regarding macroglossia, it was decided by her guardian to observe the child's growth and development for indications prompting the need for glossectomy. In the same session, prophylaxis was performed, fluoride varnish was applied, instructions on oral hygiene were given, and a labial frenectomy procedure was scheduled (labial frenulum with insertion in the papilla and ischemia region, in addition to reports from the guardian regarding difficulties with cleaning teeth in this region), which was performed in the subsequent session.

Post-frenotomy reassessment was performed when the patient was 1 year, 6 months of age (i.e., 2 months after the surgical procedure). The mother reported improvement in breastfeeding and tongue movements, as described below. For reassessment, the same protocol[13] was used.

Clinical records regarding family history remained the same. Regarding breastfeeding, in the reassessment, the mother reported that the daughter increased the interval between breastfeeds without exhibiting tiredness during feeds, and not releasing and biting the nipple and, sometimes, suckled for a while and slept. These features yielded 1 of a maximum of 8 points.

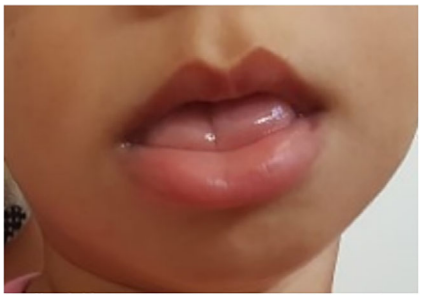

FIGURE 1 The habitual posture of lips

Clinical speech-language examination revealed habitual posture with open lips closed and the tongue remaining low, usually positioned on the lower lip (Figure 1). The examination revealed a slight crack at the apex, although it was not possible to observe the sublingual frenulum with an elevation and posteriorization maneuver of the tongue because the patient did not allow it. However, when the patient willingly raised her tongue, it was possible to visualize a thin fixation of the frenulum to the sublingual face of the tongue in the middle-third, and the insertion on the floor of the mouth, from the sublingual caruncles.

In the functional evaluation, it was not possible to observe non-nutritive sucking because the patient did not allow it. Nutritive sucking was assessed by breastfeeding, in which the suction rhythm was characterized by several suctions followed by short pauses, with good coordination between sucking/swallowing/breathing, without a "bite" to the mother's nipple or crackling tongue sounds during suction. Mobility of the tongue was observed throughout the evaluation process; it was verified that the patient was able to lateralize, protrude (Figure 2), and elevate it. In addition, palpation verified sagging lips and cheeks, as well as decreased tongue tension.

From the total sum of the protocol, 5 points were obtained (of a total of 25 points), and when a score is ≥13, one can consider the interference of the frenulum in the movements of the tongue. In the previous evaluation, the patient scored 16 points, corresponding to an improvement of 56%.

Regarding oral language, the mother reported that lallation started between 3 and 4 months of life, and that at 1 year, 6 months the child emitted /ma'mã/ for mom, /pa'pa/ for dad, /go'gɔ/ for gogó (baby bottle), /'aw' aw/ for dogs, /'kuko/ for juice, /'ai/ for more and /'awa/ for water. On the day of screening, emissions of /'tia/ for aunt and /'nãw/ were not observed.

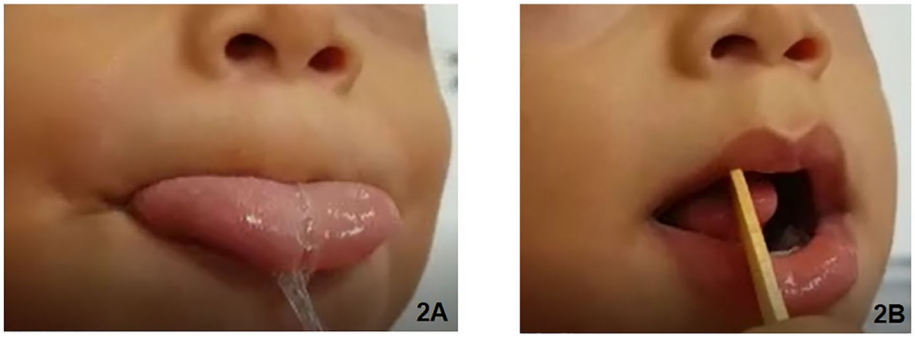

FIGURE 2 Spontaneous tongue movement: protraction and lateralization

4 | DISCUSSION

BWS is a rare disease, with the present article describing such a case. We believe it is of high value because it provides insights into fundamental mechanisms in addition to assisting professionals involved in the treatment of other patients affected by this condition.[3]

Variable degrees of macroglossia have been observed in patients diagnosed with BWS. A previous study demonstrated that one of the main features of macroglossia is the protrusion of dentoalveolar structures, resulting in an open bite and prognathic mandibular appearance secondary to an abnormally obtuse gonial angle and increased effective mandibular length.[1] Early correction of macroglossia through partial glossectomy can promote reduction in the anterior open bite and mandibular prognathism. However, changes that occur during growth and development of tongue shape and dentofacial morphology can support the premise that early partial glossectomy should be postponed or abandoned, leaving aside cases in which tongue reduction is considered to be mandatory. Nevertheless, this type of treatment is challenging and requires customization in the absence of a standard protocol(s) or guidelines for such a rare disease.[15] Thus, in the present case, together with the child's guardian, it was decided to wait for a more advanced stage of growth and development of the child to perform partial glossectomy.

According to the literature, wider and bulkier tongues can lead to functional changes in the stomatognathic system, thus affecting chewing, swallowing and speech, and facial aesthetics,[10,11] and the size and shape of the dental arch, speech intelligibility, and recurrent lingual trauma. In addition, it can hinder the stability of orthodontic treatment(s),[16] in addition to apnea,[17,18] requiring interdisciplinary collaboration to minimize its effects.

In the first interview with the individual responsible for the patient described in this report, the size and volume of the tongue were not considered as maternal complaints, even after the team became aware that macroglossia was part of the syndrome and that surgery would favor its visualization. Due to release of the tongue frenulum, these aspects were later reported as complaints by the guardian, and it was noted that the perception of her daughter's disability can generate social stigma.[19] Therefore, health professionals need to be attentive and actively listen to complaints to be able to intervene appropriately and facilitate acceptance by the family and society of craniofacial disorders resulting from genetic syndromes.

In the present case, the tongue test was not performed at birth, with ankyloglossia being symptomatic (more specifically, when it compromises the function of the tongue[20]), visible, and easy to perceive. If such a procedure had been performed earlier, there would certainly be impact on the patient's weight gain and breastfeeding habits, as highlighted in the literature,[21,22] in addition to enabling the follow-up of the recommendations made by the World Health Organization[23] and the Ministry of Health of Brazil[24] regarding exclusive breastfeeding up to 6 months and up to 2 years of age with the introduction of complementary feeding. This recommendation is important because the literature has pointed out that ankyloglossia can lead to early weaning, future speech changes,[25] difficulties with cleaning the oral cavity, social embarrassment,[22] and changes in craniofacial growth and development.[26] There is also the possibility that ankyloglossia could have repercussions on breathing, which is a risk factor for obstructive sleep apnea.[27] However, according to a Clinical Consensus Statement on Ankyloglossia in Children,[22] there is still insufficient evidence regarding this issue.

Among mothers of children with ankyloglossia who breastfeed, there may be complaints of pain when breastfeeding, the presence of ulceration, nipple bleeding, and mastitis due to the baby's sucking difficulties and, consequently, incomplete emptying of the breasts, in addition to anxiety, frustration, feelings of failure, or maternal inadequacy.[28] As such, this study confirmed previous literature reports[28,29] because the patient's mother reported that she experienced pain while breastfeeding, especially when the breasts were "full" and her daughter did not

breastfeed sufficiently to empty them. Together with pain, she felt anxious about the difficulties presented by her daughter during breastfeeding, which can be explained by the presence of ankyloglossia. A previous study[30] added to this situation the possibility of maternal mastitis and the baby's signs of frustration. It appears pertinent that this problem should be critically analyzed by health professionals who attended to the mother-baby dyad during the breastfeeding process.

Maternal insistence on breastfeeding enabled the baby to continue the process; however, there were several difficulties, as evidenced in the first evaluation. To minimize such difficulties, surgery was performed. Such a procedure can be conventional when the frenulum is cut using a scalpel blade or when it is performed using electrocautery or a high-power laser.[25] In this case, the procedure was performed using conventional methods because of the surgical opportunity. In addition, photobiomodulation using a low-level laser was performed to aid with analgesia, inflammation modulation, and tissue repair in the postoperative period.

Ankyloglossia is a congenital anomaly that may be present in some syndromes such as BWS,[9,17] thus highlighting the presence of this characteristic in the present clinical case. In addition, familial ankyloglossia may be possible given that there were similar cases in the patient's family history. Changes in the frenulum of the tongue in the family have been reported in inheritances linked to the X chromosome and in autosomal dominants with incomplete penetrance patterns based on the analysis of family lineage,[31] with genetic evaluation in these cases being relevant.

It is noteworthy that descriptions of changes in the frenulum of the tongue in syndromes are scarce and may occur due to *TBX22* gene mutation, resulting in microvariation in the fixation of the genioglossus muscle and cleft palate linked to chromosome Xq21 (OMIM # 303400).[32] In Joubert syndrome (OMIM # 213300), the cerebellum and brain stem do not fully develop due to genetic flaws; a clinical case involving a 4-year-old girl with ankyloglossia has been described in the literature.[33] A 15-year-old male adolescent with Van der Woude syndrome with a short frenulum[34] and a family with eight members exhibiting ankyloglossia was also reported, determining autosomal dominant inheritance.[35] Concern with a short and anterior frenulum is based on limitation of movements of the tongue in the various functions it performs, such as suction, speech, swallowing, and chewing and in oral hygiene.

Thus, frenectomy may result in better tongue mobility and, consequently, functions of the tongue, as observed in this case and confirmed in the literature.[36] Longitudinal monitoring of clinical cases is important because there may be changes in chewing, speech, and sleep due to the previous existence of a short tongue frenulum, which, after frenectomy, can improve considerably.[37] Therefore, we recommend extensive training of health professionals on the subject to provide better assistance with maternal and child health.

FINAL CONSIDERATIONS

Despite the presence of BWS-related macroglossia, frenectomy facilitated anatomo-functional advances in the tongue of the patient described in this clinical case.

CONFLICT OF INTEREST

The authors declare that there is no conflict of interest that could be perceived as prejudicing the impartiality of the research reported.

ETHICS STATEMENT

The Ethics Committee (CAAE 20786719.1.0000.5546) of the Federal University of Sergipe, Prof. Antônio Garcia Filho Campus, located in Lagarto, Sergipe, approved this research.

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