

Treatment of a thyroglossal duct fistula using the modified Sistrunk technique: A case report

Tratamento de fístula do ducto tireoglossal através da técnica de Sistrunk modificada: Relato de caso

Tratamiento de una fístula del conducto tirogloso mediante la técnica de Sistrunk modificada: Caso clínico

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Abstract

During the intrauterine phase, embryological structures such as the branchial arches develop, resulting in structures like the pharyngeal diverticulum, which will later give rise to the thyroid gland and the thyroglossal duct, both vital for human development. However, malformation of these structures can give rise to cysts and fistulas along the thyroglossal duct, resulting in difficulties in moving the tongue, dysphagia, or dysarthria. To treat this pathology, surgical intervention is required, based on the Sistrunk technique, which aims to remove the cysts and fistulas associated with the thyroglossal duct, as well as all its fistular epithelium or cystic capsule, with a safety margin to avoid a recurrent process. This manuscript aims to report a clinical case of a male patient, aged 1 year and 8 months, who had a fistula arising from the thyroglossal duct on the dorsum of the tongue without a path of continuity, resulting in a cleft tongue. The patient was seen at the "Santa Casa de Misericórdia" hospital in Araçatuba City for surgical removal of the lesion associated with the tongue with a safety margin, and it was possible to completely resolve the case, confirmed by the three-month follow-up of the patient, who currently has a completely healed surgical site and control of tongue movement, speech and swallowing. We, therefore, conclude that the more conservative surgical approach, using the modified Sistrunk technique, proved to be effective in this case, promoting the return of function and complete healing of the wound without any recurrences or intercurrents.

Keywords: Thyroglossal cyst; Congenital abnormalities; Oral surgical procedures.

Resumo

Durante a fase intrauterina em que se ocorrem o desenvolvimento de estruturas embriológicas, como os arcos branquiais, que resultam em estruturas como, o divertículo faríngeo, que posteriormente dará origem a glândula tireoide e ao ducto tireoglosso, são vitais para o desenvolvimento humano. Porém, a malformação dessas estruturas pode resultar na origem de cistos e fístulas ao longo do ducto tireoglosso, resultando em malefícios, como dificuldade na movimentação da língua, disfagia ou disartria. Para o tratamento desta patologia, é necessária intervenção cirúrgica, baseada na técnica de Sistrunk, que visa remover os cistos e fístulas associado ao ducto tireoglosso, bem como todo o seu epitélio fistular ou cápsula cística, com uma margem de segurança, a fim de evitar um processo recidivante. O objetivo do presente manuscrito é relatar um caso clínico de paciente, do sexo masculino, de aproximadamente 2 anos de idade, que possuía fístula proveniente do ducto tireoglosso sobre o dorso lingual sem trajeto de continuidade, resultando em uma fenda lingual. O paciente foi atendido no hospital da Santa Casa de Misericórdia de Araçatuba para remoção cirúrgica da lesão associada à língua, com margem de segurança, na qual obteve completa resolução do caso, confirmado pelo acompanhamento de três meses do paciente, que atualmente se encontra com o sítio cirúrgico completamente cicatrizado e com plenos domínios da movimentação da língua, fala e deglutição. Conclui-se que a abordagem cirúrgica mais conservadora, através da técnica de Sistrunk modificada, se mostrou muito eficaz, promovendo o retorno da função e a completa cicatrização da ferida, sem que houvesse recidivas e intercorrências.

Palavras-chave: Cisto tireoglosso; Anomalias congênitas; Procedimentos cirúrgicos bucais.

Resumen

Durante la fase intrauterina se desarrollan estructuras embriológicas como los arcos branquiales, dando lugar a estructuras como el divertículo faríngeo, que posteriormente da lugar a la glándula tiroidea y al conducto tirogloso, vitales para el desarrollo humano. Sin embargo, la malformación de estas estructuras puede dar lugar al origen de quistes y fístulas a lo largo del conducto tirogloso, provocando daños como dificultad para mover la lengua, disfagia o disartria. Para tratar esta patología es necesaria la intervención quirúrgica, basada en la técnica de Sistrunk, cuyo objetivo es extirpar los quistes y fístulas asociados al conducto tirogloso, así como todo su epitelio fistuloso o cápsula quística, con un margen de seguridad, para evitar un proceso recurrente. El objetivo de este manuscrito es comunicar un caso clínico de un paciente varón, de aproximadamente 2 años de edad, que presentaba una fístula surgida del conducto tirogloso en el dorso de la lengua sin trayecto de continuidad, dando lugar a una lengua fisurada. El paciente fue tratado en el hospital Santa Casa de Misericórdia, en Araçatuba, para la remoción quirúrgica de la lesión asociada a la lengua, con margen de seguridad, lo que resultó en la resolución completa del caso, confirmada por el acompañamiento de tres meses del paciente, que actualmente presenta sitio quirúrgico completamente cicatrizado y control total del movimiento de la lengua, del habla y de la deglución. Concluimos que el abordaje quirúrgico más conservador, utilizando la técnica de Sistrunk modificada, demostró ser muy eficaz, favoreciendo el retorno de la función y la cicatrización completa de la herida, sin recidivas ni complicaciones.

Palabras clave: Quiste tirogloso; Anomalías congénitas, Procedimientos quirúrgicos orales.

1. Introduction

Thyroglossal duct cysts and fistulas are an embryological congenital malformation of the thyroid gland, arising on the 17th day of intrauterine life from the maxillary and hyoid arches (1st and 2nd branchial arches). The primordial thyroid (lingual thyroid), through a proliferation of cells, gives rise to cords of cells that gradually form a cavity (Sturniolo et al., 2017). This formation migrates caudally in the form of a diverticulum, which remains connected to the floor of the pharynx (Righini et al., 2016; Montovani et al., 1986). This diverticulum then extends to the anterior surface of the trachea (6th week), dividing into two lobes, giving rise to the definitive thyroid gland (7th week) (LaRivière et al., 2012; Righini et al., 2016).

The descending path of the pharyngeal diverticulum corresponds to the path occupied by the thyroglossal duct. During this migration, the duct is located posteriorly to the 1st pharyngeal arch and anteriorly to the hyoid bone, and from the 5th week its development begins (hyoid bone) (Patigaroo et al., 2017) and then begins to grow and rotate in an anterior direction, compressing and obliterating the thyroglossal duct, resulting in its segmentation into suprahyoid (superior vestige) and infrahyoid (caudal vestige) (Righini et al., 2016; Montovani et al., 1986; Mettias et al., 2023).

In most cases, these segments atrophy and disappear by the end of the eighth week of intrauterine life, resulting in a superior vestige, a depression located at the base of the tongue, at the apex of the lingual “V”, known as the blind foramen, and a caudal vestige, the thyroid lobe (Labouette's pyramid) (Righini et al., 2016; Montovani et al., 1986). However, anomalies

during embryonic development can result in the permanence of the thyroglossal duct (Favarim et al., 2023), which has a fistular path that extends from the blind foramen, at the apex of the lingual “V”, to the thyroid region (Mukul et al., 2016), lined by a defined epithelial membrane (Montovani et al., 1986). For this reason, the formation of cysts along the thyroglossal duct becomes recurrent.

65% of cysts occur at the infrahyoid level, a slightly smaller percentage at the suprahyoid, thyroid and supra sternal levels, while an even rarer percentage can appear on the tongue (called intralingual), which can originate at the level of the blind foramen or project forward, occupying the thickness of the organ and causing a protrusion (Patel et al., 2019; Montovani et al., 2016; Mettias et al., 2023). In patients affected by this pathology, difficulty in moving the tongue, dysphagia and dysarthria can be observed (Montovani et al., 1986), as well as clinical, histopathological and imaging changes that distinguish them from conventional cysts associated with this organ, such as the absence of a capsule, the presence of cystic content or its ovoid or rounded projection, resembling a lingual fistula.

Once this pathology has been correctly diagnosed by means of complementary imaging and histopathological exams, it is important to correctly plan the surgical treatment (Li et al., 2023). This can range from complete surgical removal, with a hyoid bone resection margin (in cysts located in the thyroid, supra- or infrahyoid region, which is based on the technique recommended by Sistrunk in 1920 and modified by him in 1928 (Montovani et al., 1986; Sistrunk, 1920; Sistrunk, 1928; Patigaroo et al., 2017; Rajan et al., 2023).

On the other hand, in cases of lesser extent or severity, surgical removal of the fistular epithelium with a small safety margin becomes necessary, without the need for partial or total removal of the thyroid gland, to avoid the risk of recurrence, when associated with the tongue (Júnior, et al., 2020). Therefore, this study aims to report a clinical case of a thyroglossal duct fistula associated with the tongue, which did not have a path of continuity with the thyroglossal duct. However, the patient had difficulties with tongue movement, dysphagia, and burning in the region of the lesion. Therefore, only the pathological epithelium was surgically removed, with a small safety margin, followed by wound closure by first intention.

2. Methodology

The following study is qualitative and describes a case of thyroglossal duct fistula associated with the tongue, providing relevant information on the etiology, variations, examinations, and treatment performed, as well as the criteria for choosing the surgical approach. The clinical case presented is of a male patient, aged 1 year and 8 months, who had a thyroglossal duct cyst associated with the tongue but without any continuity with the thyroglossal duct. The patient was treated in the “Santa Casa de Misericórdia” hospital in Araçatuba City. This study was approved by the ethics committee, and his guardian signed the Informed Consent Form (ICF) provided by the Araçatuba School of Dentistry, authorizing the diagnosis, treatment, and use of images for publication in newspapers or scientific journals, under the Declaration of Helsinki (Pereira et al., 2018).

3. Results

A male patient, 1 year and 8 months old, was seen at the Santa Casa Hospital in Araçatuba City due to a congenital lesion on the dorsum of the tongue, located in the median sulcus, with no history of similar cases in the family. According to the mother's report, the patient had pain and a burning sensation when eating very hot or spicy food and had difficulty accepting the diet and the process of introducing food. The patient had no systemic comorbidities or history of previous illnesses but was allergic to penicillin. Physical examination revealed deciduous teeth, an anterior open bite associated with parafunctional habits, intact and normal-colored mucous membranes, and an irregular formation on the dorsum of the tongue,

with no signs of inflammation (Figure 1).

Figure 1 - Clinical aspect of the lesion on the lingual dorsum.



Source: Author's images.

To complement the clinical assessment, a cervical and lingual ultrasound (US) was requested, an ideal complementary exam for visualizing the soft tissues. The thyroid ultrasound (7-10 MHz) revealed an eutopic gland, with homogeneous parenchyma and normal lobar volumes (right: 1.0 cm³; left: 0.9 cm³). No ectopic thyroid tissue or communication between the gland and the lingual lesion was identified. Lingual US (7-10 MHz) showed a lesion with irregular contours, internal gas content and a maximum depth of 0.5 cm, with no evidence of solid or cystic components.

In view of the clinical picture and evaluation of the complementary imaging and laboratory tests, surgical excision of the lesion was indicated, based on the modified Sistrunk technique (limited to removal of the fistula or cyst, avoiding excision of the thyroid gland). The procedure was performed under general anesthesia combined with local anesthesia (lidocaine 2% + epinephrine 1:100,000). Initially, extraoral asepsis was carried out with 2% degerming aqueous chlorhexidine and intraoral asepsis with 1% aqueous chlorhexidine. Next, anesthetic infiltration was carried out around the lesion, avoiding tissue deformation and concomitant local hemostasis, allowing greater visualization of the surgical site. The tongue was stabilized with a 5-0 nylon thread at its apex, allowing it to be pulled out, and an ellipse-shaped incision was made around the lesion with a #15 blade. Subsequently, using blunt-tipped scissors, the tissue was carefully divulsed, allowing complete resection of the lesion. After removing the lesion, hemostasis was performed using electro cautery (Figure 2), and internal and superficial sutures were made using absorbable thread (Vicryl 4-0) (Figure 3).

Figure 2 - Trans-operative image of tongue lesion excision after hemostasis.



Source: Author's images.

Figure 3 - Image showing the tongue sutured with 4-0 vicryl absorbable thread.



Source: Author's images.

In the immediate post-operative period, the patient was sent to the Post-Anesthetic Recovery Room (PACU) and discharged after 24 hours, with a prescription for anti-inflammatory (Ibuprofen 100mg/ml) and analgesic (Dipyrone 500mg/ml)

medication. Post-operative instructions were given verbally and in writing to the person responsible. The specimen was placed in a vial with 10% buffered formaldehyde, duly identified, and sent for histopathological analysis (Figure 4).

Figure 4 - Tissue removed from lingual dorsum sent for histopathological analysis.



Source: Author's images.

Histopathological analysis showed polypoid hyperplasia, chronic inflammatory infiltrate, and focal myxoid alterations in the lamina propria. In addition, preserved minor salivary glands and congested vessels were observed. No thyroid tissue, embryonic remains, or signs of malignancy were identified.

Outpatient follow-up showed adequate healing, with intact sutures and no signs of infection or dehiscence. After three months, the tissue had completely healed, with no evidence of recurrence of the lesion, and the patient was able to eat easily and no longer had any painful symptoms or tongue burning (Figure 5).

Figura 5 - Image showing the post-operative result. Wound without dehiscence and no signs of infection. Successful treatment was identified. Source: Author's images.



Source: Author's images.

4. Discussion

Cysts and fistulas of the thyroglossal duct are congenital alterations (Castelan & Lopes, 2012). The thyroid develops at the base of the tongue and migrates to the neck, leaving cellular debris along the way. As the tongue is formed after the thyroid, alterations in this region can lead to the persistence of thyroid tissue (Allard, 1982; Sales Chagas et al., 2012), as in the patient's case, resulting in a lingual fistula of the thyroglossal duct without direct contact with the thyroid tissue, forming a lesion characteristic of a lingual cleft. These fistulas are usually painless, unless there is a secondary infection or lesion, usually caused by local trauma, spontaneous drainage or alterations caused by inappropriate surgery, thus forming fistulas in this region (Allard, 1982). In terms of position, they can be intralingual (2.1%), suprahyoid including submentonian (24.1%), thyroid (60.9%) and suprasternal (12.9%) (Allard, 1982). Cysts and fistulas in the suprahyoid region and especially intralingually can often not generate symptoms, but rather alterations, such as a cleft tongue, which in some cases can generate a feeling of suffocation, dysphagia and dysphonia (Allard, 1982).

As thyroglossal duct cysts and fistulas are soft tissue lesions, radiographic findings are mostly negative (Allard, 1982; Li et al., 2023). When there are doubts as to their extent and ramifications, contrast injection can be carried out, but only in cases of extreme necessity, as in addition to generating a local fistula, this procedure can be very painful (Allard, 1982). Ultrasound (US) of the neck, thyroid and tongue is usually sufficient to identify the structures (Castelan1 & Lopes, 2012; Shahin et al., 2005; Vitorino et al., 2024), as in the case of the treated patient, where an echogenic image of irregular borders with gaseous content inside was identified, with a depth of approximately 0.5cm and no thyroid alterations, without the need for additional tests, complementing the clinical diagnosis of lingual fistula of the thyroglossal duct (degenerated), without thyroid tissue, forming a lesion characteristic of a lingual cleft.

Although 50% of cysts and fistulas subjected to surgical excision contain thyroid tissue in their structure (Vitorino et al., 2024), the histopathological analysis of the sample from the clinical case in question revealed that it did not. Fistulas and cysts generally do not exhibit abnormal changes or grow larger than 4 cm, and they have a low malignancy rate (Castelan & Lopes, 2012; Vitorino et al., 2024; Bao et al., 2023). Therefore, the treatment of these lesions mainly involves complete surgical removal with a safety margin to prevent recurrent infections, given the low risk of malignancy. Simple excision of these cysts and fistulas has recurrence rates ranging from 45% to 55%. The modified Sistrunk technique (1928) is the procedure of choice because it involves excising the fistula from the thyroglossal duct and the surrounding tissue of the thyroglossal tract, without the need for total or partial removal of the thyroid gland if thyroid tissue is not present, thereby reducing the chances of recurrence (Borges et al., 2020; Sistrunk, 1920; Amos J., & Shermetaro C., 2023).

In the presented clinical case, the treatment of the pathology was surgical excision, as recommended by Sistrunk and modified by him in 1928, with surgical removal of the lesion with a safety margin, with a minimum depth of 0.5 cm, as described in the report of the ultrasound performed, associated with correction of the cleft, located on the dorsal surface of the tongue (Vitorino et al., 2024). The recurrence of the lesion occurs in approximately 10% of cases, being associated with incomplete excision, intraoperative rupture, and the surgeon's experience (Amos J., Shermetaro C., Thyroglossal Duct Cyst, 2023). Most complications are mild and present low morbidity, including local infection, seroma formation, hematoma, and wound dehiscence (Thompson LD, Herrera HB, Lau SK, 2016).

Although rare, laryngotracheal injuries and hypoglossal nerve damage are severe complications of the surgical procedure recommended by Sistrunk (1920). Inadequate resection of the thyroid cartilage can compromise the airways, swallowing, and voice, while injury to the hypoglossal nerve can result in partial tongue paralysis. Accurate identification of anatomical structures and resection medial to the lesser horn of the hyoid are essential to minimize these risks (Amos J., Shermetaro C., Thyroglossal Duct Cyst, 2023; Sturniolo et al., 2017). The differential diagnosis of this pathology is

challenging and requires analysis of the patient's age, lesion location, and its relationship to adjacent structures for correct identification.

Thyroglossal duct fistulas, as well as their intralingual variant, although rare, can cause significant symptoms such as difficulty in tongue movement and dysphagia. Accurate diagnosis, complemented by imaging studies (such as ultrasound) and associated with surgical removal, are essential for effective treatment and prevention of recurrence (Rajan et al., 2023). Furthermore, preserving anatomical structures during the surgical procedure ensures proper recovery and minimizes the risk of postoperative complications.

5. Conclusion

Given the presented facts, it can be concluded that the diagnosis, methodology, and technique adopted by the team to conduct the treatment were successful, based on the well-established Sistrunk technique (1920), with a somewhat more conservative approach. Furthermore, because the lesion didn't present malignancy risks or thyroid tissue, it was possible to remove all the pathological tissue from the patient with a small safety margin without any complications or recurrences after a three-month follow-up. This allowed the patient to present tissue with all the characteristics of normalcy and complete wound healing after this period. Thus, it was possible to provide the patient with an improved quality of life by restoring aesthetics, function, speech, mastication, and swallowing without complications or recurrences.

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