Epidermoid cyst mimicking a thyroglossal duct cyst in a pediatric patient: a case report

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This article presents a case of an epidermoid cyst that mimicked a thyroglossal duct cyst in a pediatric patient. An 8-year-old boy was referred for evaluation of a volumetric increase in the median cervical region with an evolution of about 4 years. The skin in the submental region was healthy and normal colored. Palpation revealed a mobile, well-circumscribed nodular lesion of soft consistency. Computed tomography of the neck showed an expansive hypodense formation extending from the base of the tongue to the upper portion of the hyoid bone, suggesting a thyroglossal duct cyst. Considering the diagnostic hypothesis, cystic enucleation via the Sistrunk procedure was planned. However, no ductal structure was identified during the surgical procedure, and the lesion was only near, but not attached to, the hyoid bone. Simple excision of the lesion was therefore performed. At the most recent follow-up examination, about 3 months postoperatively, the patient demonstrated satisfactory clinical progress. The epidermoid cyst close to the hyoid bone presented diagnostic difficulty due to its similarity to a thyroglossal duct cyst. Computed tomography provides limited information for diagnosing this type of lesion, and ultrasonography is the preferred test. In view of the uncertain diagnosis in this case, the extent of the excision was determined during the surgery, and simple excision was a satisfactory treatment associated with a good prognosis.

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Congenital masses in the head and neck region can present as abnormalities of a cystic, solid, or vascular nature and are rarely diagnosed as neoplasms. When present in the midline of the neck, they may represent thyroglossal duct cysts, dermoid or epidermoid cysts, branchial cleft cysts, or teratomas.[1]

Epidermoid cysts are uncommon developmental lesions resulting from the entrapment of ectodermal tissue along the embryonic fusion lines.[2] Although they have a congenital origin, they are usually diagnosed in the second or third decades of life and can develop anywhere in the body.[3] Epidermoid cysts represent about 7% of cysts in the head and neck region and fewer than 0.01% of oral cysts; when they do occur in the oral cavity, the midline of the floor of the mouth is the most frequent location.[4] When present in the anterior cervical region, these cysts are usually located close to the hyoid bone, which may suggest a diagnosis of thyroglossal duct cyst.[5]

Clinically, epidermoid cysts manifest with slow growth, are asymptomatic, are fluctuating or firm on palpation, and rarely cause functional interference; however, in larger dimensions, they can cause changes such as dysphagia, dysphonia, and dyspnea. Microscopically, these cysts are covered with simple stratified squamous epithelium and filled with keratin lamellae. Diagnosis is made by clinical, imaging, and histopathologic examinations. Depending on cyst location, surgical treatment can be performed by an intraoral or extraoral approach and results in a low recurrence rate and good prognosis.[6,7]

This article presents a case of a pediatric patient with an epidermoid cyst in the submental region. The cyst was in close contact with the hyoid bone, mimicking a thyroglossal duct cyst, and was removed via simple enucleation.

Case report

An 8-year-old boy was referred to the outpatient oral and maxillofacial surgery and traumatology clinic of the Mato Grosso Cancer Hospital, Cuiabá, Brazil, for evaluation of a volumetric increase in the median cervical region with an evolution of about 4 years.

On extraoral physical examination of the submental region, the skin was healthy and normal colored. Palpation revealed a mobile, well-circumscribed nodular lesion of soft consistency (Fig 1). Intraorally, the structures were within normal limits. Computed tomography of the neck without contrast enhancement revealed an expansive, 2.5 × 2.0-cm hypodense formation in the submental region (Fig 2). The lesion, which had regular contours and well-defined margins, extended from the base of the tongue to the upper portion of the hyoid bone. These features led to the diagnostic hypothesis of a thyroglossal duct cyst.

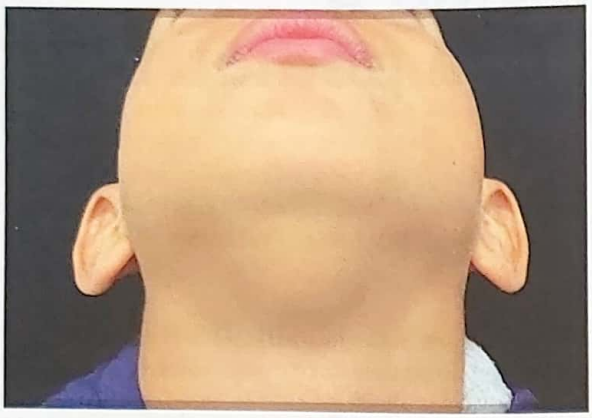

Fig 1. Volumetric increase in the submental region of an 8-year-old boy.

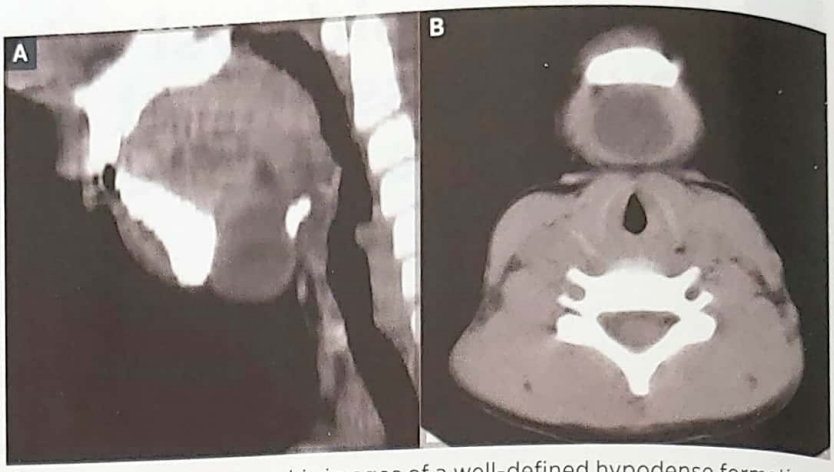

Fig 2. Computed tomographic images of a well-defined hypodense formation, suggestive of a cystic lesion, located between the base of the tongue and the hyoid bone. A. Sagittal view. B. Axial view.

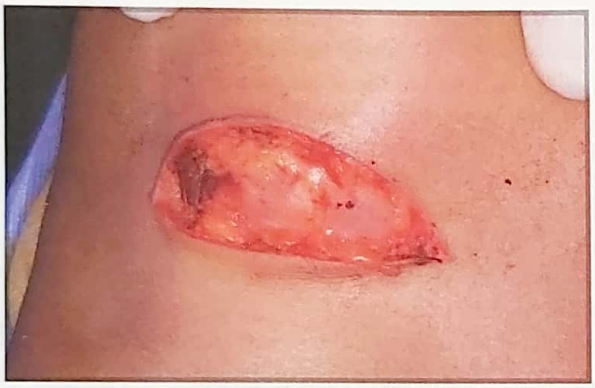

Fig 3. Incision of the skin and subcutaneous tissue.

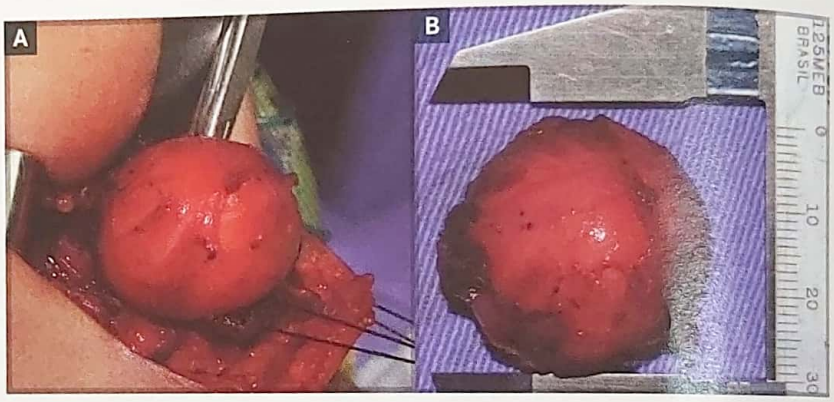

Fig 4. Enucleation procedure. A. Total exposure of the cyst. B. Cyst measuring approximately 30 mm in diameter.

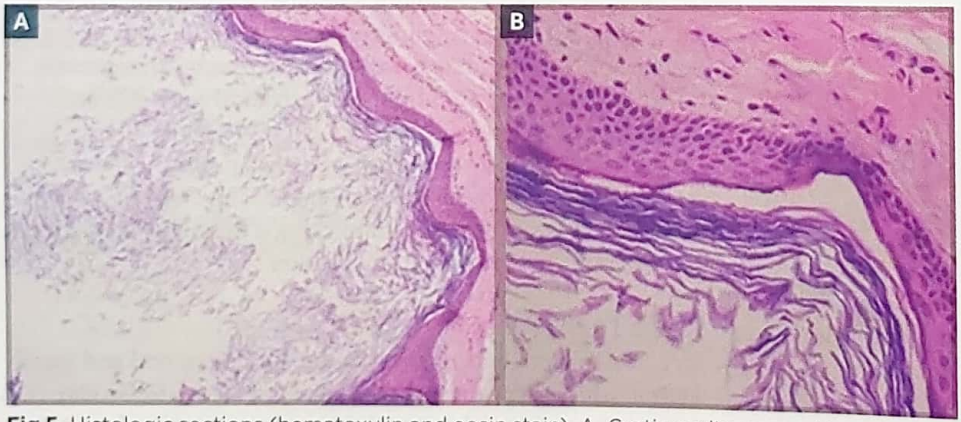

Fig 5. Histologic sections (hematoxylin and eosin stain). A. Cystic cavity covered by stratified squamous epithelium containing keratin lamellae (original magnification ×100). B. Stratified squamous epithelium with keratin lamellae arranged in piles (original magnification ×400).

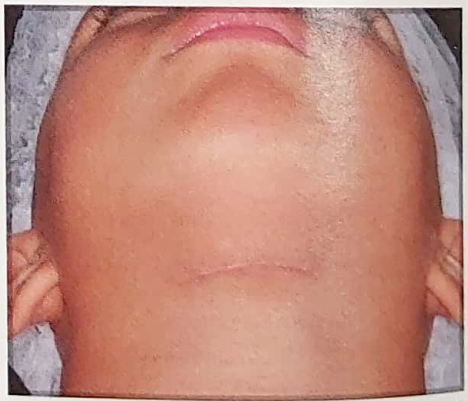

Fig 6. Symmetric submandibular region with a normal-colored and clinically satisfactory scar 40 days postsurgery.

Fine-needle aspiration cytology was performed, and the cytopathologic report described adequate cellularity constituted predominantly by large epithelial cells and a few mononuclear cells, mainly lymphocytes, in an amorphous background, which was compatible with cystic content and absence of malignancy. Considering the data from the clinical, imaging, and cytopathologic examinations, the diagnostic hypothesis of thyroglossal duct cyst was supported.

The planned surgical treatment consisted of the Sistrunk procedure, whereby the thyroglossal cyst is enucleated via dissection of the thyroglossal duct to the base of the tongue and removal of the middle portion of the hyoid bone. After the induction of general anesthesia and orotracheal intubation of the patient, an incision was made in the skin overlying the lesion and the tissue was carefully dissected in layers, avoiding the cyst to preserve its integrity (Fig 3).

No ductal structure that was relevant to the diagnostic hypothesis was identified during the surgical procedure, and it was found that the lesion was only near, but not attached to, the hyoid bone. The cyst was completely removed without any bone, and sutures were placed in layers to minimize the formation of dead space. An intradermal suture was then placed, aiming at a better esthetic result.

Macroscopically, the cyst had dimensions of 3.0 × 3.0 × 2.5 cm and weighed 10 g (Fig 4). The specimen was sent for histopathologic examination, which revealed that the growth was an epidermoid cyst, consisting of a cystic cavity covered by stratified squamous epithelium without atypia and containing keratin lamellae (Fig 5).

On the 7th postoperative day, the surgical wound had a good appearance with scar formation and mild edema in the region. On the 40th postoperative day, the volume of the median cervical region was within the normal range, with a normal-colored and clinically satisfactory scar (Fig 6).

The patient remains under observation and has not presented any other complaints or clinical changes. At the most recent follow-up examination, about 3 months postoperatively, the patient demonstrated satisfactory clinical progress.

Discussion

This paper presents the clinical case of an epidermoid cyst resembling, both clinically and on the imaging examinations, a thyroglossal duct cyst. The absence of a ductal structure and lack of contact with the hyoid bone, observed during the surgical procedure, resulted in a change in the surgical plan from the Sistrunk procedure to simple enucleation.

The epidermoid cyst is one of 3 histologic variants of a lesion primarily called a *dermoid cyst*: (1) an epidermoid cyst derived from the ectodermal tissue, with a squamous epithelial lining, without an attached structure; (2) a true dermoid cyst, originating from ectodermal and mesodermal components, with association of cutaneous attachments; and (3) a teratoid cyst/teratoma, with an epithelial lining derived from the 3 germ layers.[2,4,8] Clinically, they are asymptomatic, slow-growing, and present with fluctuation.[9] The location can be referred to as *sublingual, submental,* or *lateral* depending on the association between the lesion and the oral floor. Cysts located below the mylohyoid muscle may present in a submental or submandibular location, depending on the laterality of the cyst.[2] In the present case, the cyst was painless, with an evolution of about 4 years, a fluctuating consistency on palpation, and a location inferior to the mylohyoid muscle in the midline of the neck (ie, in the submental region).

Considering their similarity to thyroglossal duct cysts when located in the anterior neck region, epidermoid cysts can be distinguished from thyroglossal duct cysts by some important differences. Dermoid/epidermoid cysts arise from elements of the first and second branchial arches, resulting in a location that tends to be more superficial in relation to the subcutaneous tissues and nearer to the base of the tongue, while thyroglossal duct cysts tend to be located more deeply and in close proximity to the hyoid bone.[10] However, these differences are of little help in forming a diagnosis in cases where the mass extends from the base of the tongue to the hyoid bone, as in the present case.

Computed tomography cannot differentiate between the 2 lesions and is associated with exposure to radiation and injection of contrast medium, while ultrasound can be useful for assessing the depth of the cyst and its possible relationship with the hyoid bone.[6,11] A study carried out to determine whether ultrasonography can differentiate dermoid cysts from thyroglossal duct cysts in the midline of the neck concluded that the presence of septa, an irregular wall, and solid components can predict a thyroglossal duct cyst.[12] In the present case, the proximity of the lesion to the hyoid bone on the computed tomographic images suggested the diagnosis of a thyroglossal duct cyst, which was proved to be incorrect during the surgery. This experience reinforces the fact that computed tomography has limited value in these cases, making it difficult to determine the appropriate surgical technique to adopt.

In a review assessing the files from 48 years of an oral pathology service, 13 (0.08%) of 15,387 cases were diagnosed as an epidermoid cyst.[3] The microscopic characteristics of the epidermoid cysts included stratified squamous epithelial lining with fibrous capsule of the salivary glands, adipose tissue, and melanin pigmentation.[3] Additionally, keratin lamellae are present in the cystic lumen.[7] The epithelial lining and cystic content of the lesion in the present case had such characteristics.

The treatment of epidermoid cysts is exclusively surgical. The approach is determined by the location of the lesion in relation to the mylohyoid muscle: extraoral when below the muscle and intraoral when above.[11] Although there is a clinical similarity between epidermoid cysts and thyroglossal duct cysts, and surgical excision is indicated for both cases, the extent of the surgery is different. While thyroglossal duct cysts have a high recurrence rate if the entire tract and the middle portion of the hyoid bone are not excised (Sistrunk procedure), epidermoid cysts can be treated with simple excision without risking recurrence.[12] When the diagnosis is not definitive, the Sistrunk procedure is planned but converted to simple excision if an epidermoid or dermoid cyst is identified.[13] In the present case, considering the diagnostic hypothesis of a thyroglossal duct cyst, excision with the Sistrunk procedure was planned, and, in view of the submental location below the mylohyoid muscle, extraoral access was used.

A retrospective study of 59 cases of patients with congenital lesions in the midline of the neck examined over a 10-year period revealed 33 cases (55.9%) of thyroglossal duct cyst and 14 cases (23.7%) of dermoid cysts.[14] A total of 38 patients with cysts had been admitted with a preoperative diagnosis of thyroglossal duct cyst, but the histopathologic report revealed the presence of a dermoid cyst in 5 patients. Of the 38 lesions with an initial diagnosis of thyroglossal duct cyst, 4 were treated with simple excision. The histopathologic findings in 3 of these cases led to the diagnosis of a dermoid cyst, and the findings in 1 case resulted in the diagnosis of a thyroglossal duct cyst. It is worth mentioning that, in the group that underwent simple excision, there was 1 case of recurrence after 8 months due to the incorrect diagnosis. The decision to treat the lesions with a non-Sistrunk procedure in these 4 cases was made during the operation because the cyst was separated from the hyoid bone and there was no ductal continuity beyond the hyoid bone. The other 34 cysts were treated with the Sistrunk procedure, and histologic analysis revealed that 2 were not thyroglossal duct cysts. The authors concluded that dermoid cysts attached to the hyoid bone must also be treated by the Sistrunk technique.[14]

Conclusion

An epidermoid cyst located close to the hyoid bone presents substantial diagnostic difficulty due to its similarity to a thyroglossal duct cyst. Computed tomography provides limited information for diagnosis of this type of lesion, and ultrasonography is the preferred test. Surgical excision is indicated for both types of cyst, but the extent of the excision differs. When no ductal structure is detected and the cyst is not attached to the hyoid bone, simple enucleation after pericapsular dissection is the treatment of choice. In the present case, in view of the uncertain diagnosis, the extent of the excision was determined during the operation, and simple excision was a satisfactory treatment associated with a good prognosis. The patient did not experience postoperative complications and remains without clinical or imaging evidence of recurrence.

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Disclaimer

The authors report no conflicts of interest pertaining to any of the topics discussed in this article.

References

1. Quintanilla-Dieck L, Penn EB Jr. Congenital neck masses. *Clin Perinatol.* 2018;45(4):769-785.
2. Klibngern H, Pornchaisakuldee C. A large sublingual epidermoid cyst with parapharyngeal space extension: a case report. *Int J Surg Case Rep.* 2020;72:233-236. doi:10.1016/j.ijscr.2020.06.014
3. Santos HB, Rolim LS, Barros CC, Cavalcante IL, Freitas RD, Souza LB. Dermoid and epidermoid cysts of the oral cavity: a 48-year retrospective study with focus on clinical and morphological features and review of main topics. *Med Oral Patol Oral Cir Bucal.* 2020;25(3):e364-e369. doi:10.4317/medoral.23388
4. Utumi ER, Araujo JP, Pedron IG, Yonezaki F, Machado GG, Rocha AC. Extensive epidermoid cyst of the submental region. *Autops Case Rep.* 2016;6(2):51-54. doi:10.4322/acr.2016.031
5. Foley DS, Fallat ME. Thyroglossal duct and other congenital midline cervical anomalies. *Semin Pediatr Surg.* 2006;15(2):70-75.
6. Puranik SR, Puranik RS, Prakash S, Bimba M. Epidermoid cyst: report of two cases. *J Oral Maxillofac Pathol.* 2016;20(3):546. doi:10.4103/0973-029X.190965
7. Kim CS, Na YC, Yun CS, Huh WH, Lim BR. Epidermoid cyst: a single-center review of 432 cases. *Arch Craniofac Surg.* 2020;21(3):171-175.
8. Goins MR, Beasley MS. Pediatric neck masses. *Oral Maxillofac Surg Clin North Am.* 2012;24(3):457-468.
9. Kolomvos N, Theologie-Lygidakis N, Tzerbos F, Pavli M, Leventis M, Iatrou I. Surgical treatment of oral and facial soft tissue cystic lesions in children. A retrospective analysis of 60 consecutive cases with literature review. *J Craniomaxillofac Surg.* 2014;42(5):392-396.
10. Patel S, Bhatt AA. Thyroglossal duct pathology and mimics. *Insights Imaging.* 2019;10(1):12. doi:10.1186/s13244-019-0694-x
11. Zielinski R, Zakrzewska A. Submental epidermoid cysts in children. *Open Med (Wars).* 2014;10(1):77-81. doi:10.1515/med-2015-0013
12. Oyewumi M, Inarejos E, Greer ML, et al. Ultrasound to differentiate thyroglossal duct cysts and dermoid cysts in children. *Laryngoscope.* 2015;125(4):998-1003.
13. Pitner H, Elmaraghy C, Fischer B, Onwuka A, Rabe A, Walz P. Diagnostic accuracy of midline pediatric neck masses. *Otolaryngol Head Neck Surg.* 2019;160(6):1111-1117.
14. Kepertis C, Anastasiadis K, Lambropoulos V, Mouravas V, Spyridakis I. Diagnostic and surgical approach of thyroglossal duct cyst in children: ten years data review. *J Clin Diagn Res.* 2015;9(12):PC13-PC15. doi:10.7860/JCDR/2015/14190.6969