Scholars Journal of Medical Case Reports
Abbreviated Key Title: Sch J Med Case Rep
ISSN 2347-9507 (Print) | ISSN 2347-6559 (Online)
Journal homepage: https://saspublishers.com

OPEN ACCESS

Radiology

Teratoid Cyst: A Rare Lingual Lesion, About a Case

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DOI: 10.36347/sjmcr.2022.v10i10.009 | **Received:** 01.09.2022 | **Accepted:** 08.10.2022 | **Published:** 12.10.2022

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Abstract **Case Report**

Dermoid cysts are dyssembryogenic tumors of ubiquitous location. Their histological aspects are varied. The tongue is a very rare location; about 20 cases have been reported in the literature. Only 6 of them were teratoid cysts (characterized by the presence of respiratory ciliated epithelium). The authors report a new observation of this rare entity in a 5-year-old male child who presented with macroglossia with speech impairment. The CT scan showed a cystic lesion on the dorsal surface of the tongue. The treatment consisted of a complete removal of the cyst. From this observation, we discuss the epidemiological, clinical, radiological, histological and therapeutic aspects of this extremely rare entity, whose diagnosis must be evoked before any lingual swelling in children.

Keywords: Dermoid cyst, macroglossia, child.

INTRODUCTION

Dermoid cysts are benign congenital dyssembryogenic tumors that can develop anywhere in the body. In the head and neck, they most often occur in the midline from the sternal fork to the submental region [1]. Dermoid cysts of the tongue are rare [2]. It can have 3 different histological aspects. The teratoid variety is extremely rare with only 6 observations reported in the literature [2]. We report a new observation.

OBSERVATION

This was a 5 year old male child whose parents brought him to the clinic for macroglossia with speech impediment. This macroglossia had been noted since birth. According to his parents, it was increasing regularly in size, hindering speech and eating but without respiratory consequences.

Clinical examination revealed a painless cystic mass of the tongue, measuring 5 cm long and mobile in both planes, with a firm consistency, without inflammatory signs and without any disturbance of the lingual mobility or palpable adenopathies (figure 1).

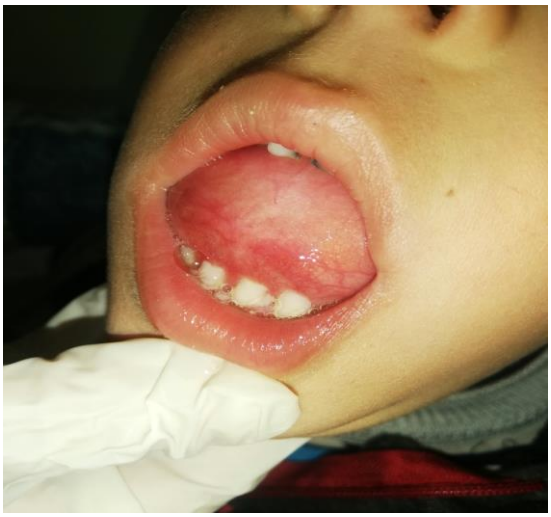

Figure 1: Cystic mass of the tongue measuring 5 cm in long axis

- An ultrasound of the subchinion region revealed a cystic formation of roughly oval shape, well limited with fine echoes and measuring 42x34 mm, with a well limited hyperechoic formation with a cystic center not lighting up on color Doppler and measuring 17.5x 13 mm, (Figure 2).

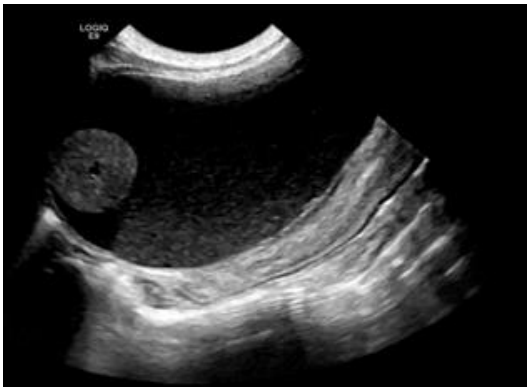

Figure 2: Ultrasound image revealing a cystic formation with fine echoes and a hyperechoic intracystic formation

- The CT scan showed a well-limited oval formation of fluid and fat density on the dorsal surface of the tongue, annularly enhanced after injection of contrast medium and measuring 15x25 mm, and a thin-walled rounded formation of fluid density annularly enhanced after injection of PDC and measuring 11x10 mm. The thyroid gland was in place with a normal appearance (Figure 3).
- MRI showed a cystic formation of the floor of the mouth measuring 56 x 42 x 42 mm with a liquid signal in T1 hypo signal, T2 hyper signal and an intracystic formation, heterogeneous, located on the left antero-lateral wall, measuring 15 x 15 mm in T1 frank hyper signal with T2 intermediate signal, enhancing in the periphery after injection of Gadolinium (figure 4).
- Topographically, it bulges downward at the submental level, medial to the anterior belly of the digastric muscle, with respect for the fatty separation line and without bone lysis in front of it. It discreetly pushes back the base of the tongue with respect for the fatty separation line and remains at a distance from the oropharynx.

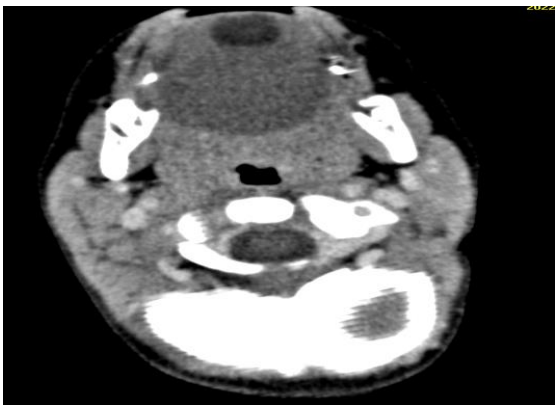

Figure 3: Facial CT scan c+ in axial section: ***oval formation of fluid and fat density on the dorsal surface of the tongue annularly enhanced after injection of contrast medium, site of a rounded formation of thin-walled fluid density annularly enhanced after injection of PDC***

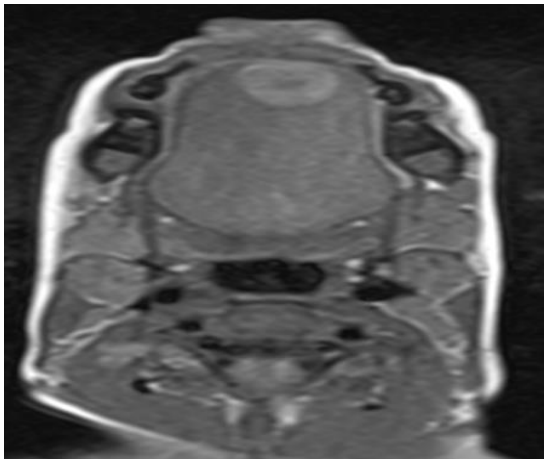

Figure 4: Facial IMR, axial section in T1 sequence: ***Cystic formation of the floor of the mouth with a liquid signal in hypo signal, seat of an intracystic formation, heterogeneous of left antero-lateral mural location in frank hyper signal.***

These data, both clinical and imaging, pointed to the diagnosis of an embryonal benign cystic lesion and the patient underwent complete surgical excision.

Macroscopically, the excisional specimen corresponded to a cystic formation measuring 16g and 4.2x3.5x1 cm. On opening, its content was sebaceous and its wall was thin. Absence of endo and exo cystic vegetations.

On microscopic examination, it was a fibrous wall bordered by a regular squamous epithelial lining. This lining was invaginated in places, giving rise to regular pilosebaceous follicles. Cartilaginous tissue, salivary glands and a regular respiratory mucosa were also present. The diagnosis retained was therefore that of a mature multi-tissue teratoma.

DISCUSSION

Dermoid cysts are congenital benign tumors that can occur anywhere in the body. In the face and neck, they are mainly located on the midline [1]. Johnsen and Erich, in a series of 1495 dermoid cysts, found 7% of cervico-facial localizations, 23% of which were on the floor of the mouth [3]. Localization in the tongue is very rare. Their presence is explained by embryogenesis. They develop from embryonic ectodermal remnants that have undergone ectopic migration between the 2nd and 5th week of embryonic life [4].

The clinical symptomatology is revealed by a painless lingual swelling or macroglossia, with difficulties in eating or infectious episodes [5]. The latter would result from contamination of the cystic contents from the oral flora or following trauma [6]. In case of late diagnosis, the increase in cystic volume, usually progressive, may become more rapid in the pubertal period due to sebum secretion, leading to submental deformity, permanent oral hollowness,

difficulties in mastication and speech, and even breathing [7, 6].

Ultrasound is the first-line examination to confirm the cystic nature of the mass, but the fatty component may go unnoticed and the deep extension into the base of the tongue may be difficult to specify because it is masked by calcific anatomical structures [1, 8].

Computed tomography (CT) plays a role both in the positive diagnosis, by demonstrating the fatty component and the possible calcific component within the cystic lesion, and in the accuracy of the lesion extension by assessing the location and depth of the mass and the assessment of associated bone lesions [9].

MRI always offers a better tissue characterization by showing the fat content, which is quite characteristic with short relaxation times in T1 and long relaxation times in T2, resulting in a T1 hyper signal and a T2 hyper signal identical to the subcutaneous fat, which is extinguished on the fat suppression sequences. It also allows a very precise assessment of extension thanks to the sagittal and coronal sections and to the better tissue contrast of the different structures in the region [9].

The list of differential diagnoses of a painless swelling of the midline of the tongue is long: hygroma, cystic lymphangioma, neurofibroma, hemangioma, lingual thyroid... [10, 11].

The clinical and para-clinical data can only guide to the diagnosis which is only made by the anatomopathological examination of the excision specimen.

This examination shows a cystic wall with a structure identical to that of the dermis with hair follicles, sweat and sebaceous glands and squamous epithelium. The contents of the cyst are composed of epithelial cell degradation products (cholesterol and keratin), glandular secretions and hair [12], which explains the partly fatty content that characterizes this lesion on imaging. Based on histological criteria, Meyer [13] showed that all dermoid cysts of the oral cavity should be classified into 3 groups:

- Squamous cysts: covered by a keratinized squamous epithelium with supporting connective tissue;
- True dermoid cysts: a cavity with an epithelial border containing sweat glands, sebaceous glands or hair follicles (skin appendages).
- Teratoid cysts covered by a squamous epithelium with which coexist a ciliated respiratory epithelium, a characteristic element. The different layers present cutaneous islands and mesodermal, endodermal and ectodermal derivatives. This last variant to which our observation belongs is extremely rare, with only six cases reported in the literature [2].

Surgical treatment is curative and consists of complete excision of the cyst [14]. Post-operative complications and recurrences are rare and occur when the removal is incomplete [12].

CONCLUSION

The dermoid cyst of the tongue is a rare congenital malformation that must be evoked before any lingual swelling in children. The teratoid variety, characterized by the presence of a respiratory ciliated epithelium, is exceptional. Early diagnosis allows adequate therapeutic management and avoids infectious, functional and morphological complications.

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