

Dermoid Cyst of the Floor of the Mouth

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Dermoid cysts are congenital lesions composed of tissues with different origins: ectoblastic, mesoblastic, or endoblastic, caused by a defect in the fusion of the embryonic lateral mesenchymatic mass. A true dermoid cyst is a cavity covered with epithelium showing keratinisation and presenting identifiable dermal appendices.

We present a case of a male patient aged 53 years presenting tumouration on the floor of the mouth with an evolution of about 30 years. A bibliographic revision of this pathology is also presented.

Key words: Dermoid cyst. Floor of the mouth. Congenital lesions.

Quiste dermoide de suelo de boca

Los quistes dermoides son lesiones congénitas constituidas por tejidos de varias procedencias: ectoblástico, mesoblástico o endoblástico. Tienen su origen en un defecto de fusión de las masas mesenquimatosas laterales embrionarias. El quiste dermoide verdadero está recubierto de epitelio que muestra queratinización y tiene apéndices de piel identificables.

Presentamos el caso de un varón de 53 años de edad que presenta una tumoración en suelo de boca de unos 30 años de evolución. Se realiza una revisión bibliográfica de dicha patología.

Palabras clave: Quiste dermoide. Suelo de boca. Lesiones congénitas.

INTRODUCTION

Dermoid cysts are development lesions found inside normal organs or tissues as a result of the inclusion of tissue from diverse sources (ectoblastic, mesoblastic or endoblastic) when a merger defect occurs in the lateral mesenchymatous masses of embryos (mainly the 1st and 2nd arcs) during the fifth week of embryological development¹⁻³.

Depending on their location, dermoid cysts are divided into medial and lateral cysts. Dermoid cysts on the floor of the mouth are often relatively soft unfluctuating masses, frequently adhered to the child's hyoid bone. In adults, the cyst on the floor of the mouth is subdivided into sublingual or genioglossal when it is located between the geniohyoid and mylohyoid muscles. It brings about an upward displacement of the tongue and, depending on the size of the cyst, will give rise to certain symptoms. When located between the mylohyoid muscle and the neck's cutaneous muscle, it is referred to as a geniohyoid cyst with displacement outwards and resembling a double chin⁴.

In a series of 1,495 dermoid cysts collected over 15 years, only 1.6% are located in the oral cavity, with cysts on the floor of the mouth representing 0.01%^{5,6}.

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In 1955, Meyer updated the concept of dermoid cyst to describe three histological variants: the true dermoid cyst, the epidermoid cyst and the teratoid variant. True dermoid cysts are cavities lined with epithelium showing keratinization and with identifiable skin appendages such as pilous follicles, and sudoriparous and sebaceous glands on the cyst wall. Epidermic cysts are lined with simple squamous epithelium with a fibrous wall and no attached structures. The lining of teratoid cysts varies from simple squamous to a ciliate respiratory epithelium containing derivatives of ectoderm, mesoderm and/or endoderm.

All three histological types contain a thick, greasy-looking material.

CASE STUDY

We present here the case of a 53-year-old male who attended the otorhinolaryngology clinic because of a tumouration on the floor of the mouth that had existed for about 30 years, although he had not noted any significant increase in size over the last few years. He had been pressured by his family into coming to the clinic because of an increase in snoring in the last two years and he believed that the tumouration on the floor of his mouth was the cause of the snoring. He reported mild discomfort in swallowing and speaking, well tolerated.

On examination, an unfluctuating tumouration was observed on the midline of the mouth floor, adhered to the



Figure 1. Tumouration on the floor of the mouth displacing the tongue upwards. The mucosa surrounding it is in appearance normal.

deep planes, painless and displacing his tongue upwards. When pressure is applied to the submaxillary region, the tumouration is displaced upwards, giving the impression of an increase in the tumouration's size, on the floor of the mouth. The appearance of the mucosa on the floor of the mouth is normal (Figure 1). The size and appearance of the orifices of Wharton's ducts are normal. Hypertrophy is observed on the veil of the palate and diffuse thickening of the pharynx. By means of fibre optic rhinolaryngoscopy, Müller's manoeuvre was performed and posterior displacement was seen in the base of the tongue, with collapse of the hypopharynx and closure of the soft palate, the most likely cause of the obstructive sleep apnoea syndrome. The larynx was normal. On inspection, his neck was seen to be short and thick. The tumouration cannot be palpated at the submentonian level and there are no laterocervical adenopathies.

The computerized tomography showed an encapsulated cystic mass without calcification (Figure 2).

The patient was informed that it was necessary first of all to perform an exeresis of the cyst and then to study the obstructive sleep apnoea syndrome. The tumouration on the floor of the mouth was not considered to be related to his snoring.

The cyst was located between the geniohyoid and the mylohyoid muscles and corresponds to a sublingual or genioglossal cyst. The exeresis of the cyst was performed using the buccal route (Figure 3). The cyst measured 8 × 5 cm (Figure 4).

The anatomical pathology showed a cystic structure containing keratinous material. The cavity was very focally delimited by a keratinized polystratified epithelial plane and, for the most part, by an inflammatory reaction comprising scant lymphocytes and abundant mononucleate and multinucleate histiocytes. The histiocytary cell elements phagocyte keratin laminae. There are frequently pilous structures among the inflammatory component, surrounded by generally multinucleate histiocyte cells. Peripherally, a fibrous reaction was observed with moderate inflammatory infiltrate of lymphocytes. It was possible to observe pilous



Figure 2. Computerized tomography, cystic tumouration of the floor of the mouth, well delimited.

structures in the lumen of the cystic space and among the keratinous component.

An immunohistochemical study was performed and showed abundant CD68-positive cellularity. The lymphoid component corresponded to B lymphocytes (CD20-positive) with a lower proportion of T lymphocytes (CD8). The anatomical pathology diagnosis was a dermoid cyst (Figure 5).

Two months after removal of the cyst, a further rhinofibrolaryngoscopy was performed, with results similar to those prior to surgery, along with a polysomnograph test. This test showed that he spent 33% of the time with saturations of O₂ < 90% and recorded 209 obstructive apnoeas, 188 hypopnoeas and an AHI of 50 per hour. The patient is currently receiving instrumental treatment with CPAP at 8 cm H₂O throughout the hours of nocturnal rest and there are no sequelae from the extirpation of the cyst.

DISCUSSION

Most patients are in the range between 10 and 35 years of age⁷. In a series of 16 cases, the mean age is 27.8 years and the ratio of men/women is 3:13, although previous papers have found no difference by gender^{9,10} while others have found a predominance of women^{11,12}. Growth of the cyst may be constrained by hormonal stimulus during puberty,

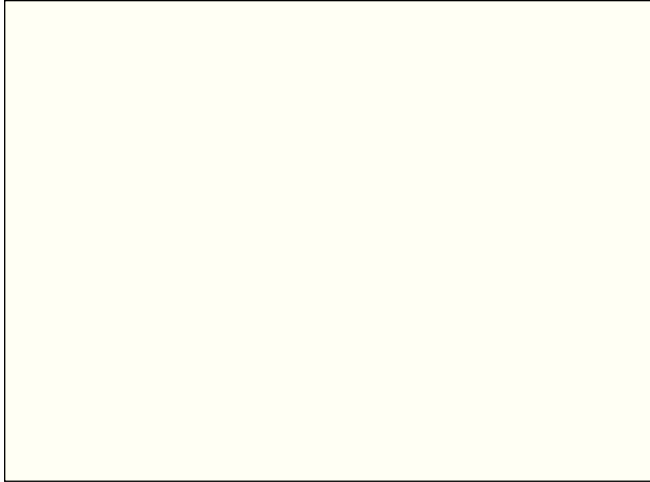


Figure 3. Exeresis of the cyst using the buccal route

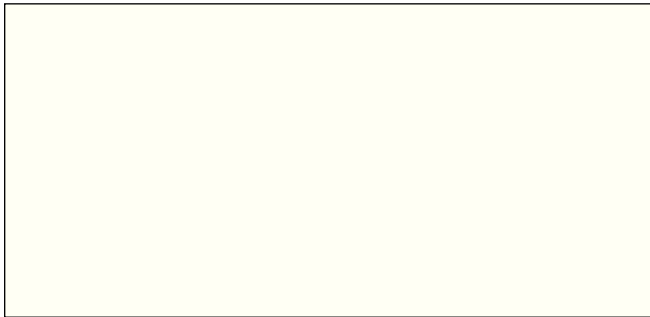


Figure 4. Image of the cyst measuring 8 × 5 cm.

producing a hypersecretion of fat, which would explain the greater incidence in the young adult stage (16-40 years of age). The cyst's fast growth is associated with infectious processes^{10,13}.

Size varies; descriptions published report up to 12 cm in diameter. The size and location of the cyst are the cause of the clinical manifestations. Dermoid cysts on the upper plane of the floor of the mouth, sublingual or ginioglossal cysts grow above the genioid muscle, displacing the tongue upwards. Depending on these two factors, patients may be asymptomatic or suffer from alterations in the mobility of their tongues, manifested by alterations in pronunciation and in chewing. Other symptoms are dyspnoea, dysphagia and obstructive sleep apnoea. In our patient the alterations in pronunciation and chewing were resolved; the obstructive sleep apnoea did not resolve with surgery and he is currently under instrumental treatment with CPAP.

For diagnostic purposes, it may be sufficient to have a computerized tomography indicating the cystic nature of the tumour, its size and anatomical relations. FNAP may be negative because the content is very thick¹⁴.

A differential diagnosis must be made bearing in mind other processes with similar characteristics and situation, such as sublingual ranula, schwannoma, lipomas, Ludwig's angina, etc.

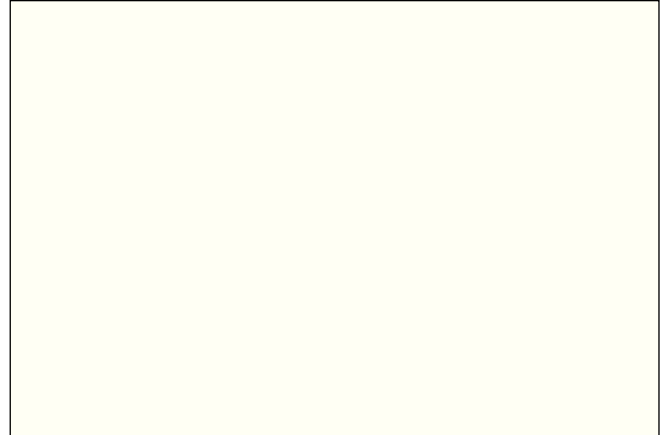


Figure 5. Histological image of the cyst: the black arrow points to a cystic cavity with keratinous material. A wall can be seen with abundant mononucleate and multinucleate histiocyte cells surrounding pilous structures (white arrow).

Treatment is surgical with exeresis of the cyst with its entire capsule in order to avoid local recurrences. The approach route differs depending on location: the cysts in the upper plane of the floor of the mouth can be accessed for exeresis through the medial sublingual raphe, with minimal risks of haemorrhage and damage to nerves. Cysts in the lower plane of the floor of the mouth can be extirpated by medial cervicotomy.

Prognosis is good if the cyst has been entirely eliminated. Local recurrences have been observed with incomplete exeresis of the lesion. No malignant transformations have been seen in the floor of the mouth, although these have been described in 5% of the dermoid cysts in other locations¹⁵.

REFERENCES

1. Contencin P. Fistulas y quistes congénitos del cuello. Encyclopédie Médico-Chirurgicale- E-20-860-A-10. Paris: Elsevier; 2000.
2. Anderson JR. Muir's Textbook of Pathology. 12^a ed. London: Edward Arnold; 1985. p. 12-48.
3. Sapp J, Eversole L, Wysocki G. Patología oral y maxilofacial contemporánea. Harpourt Brace, ed., 1998.
4. Nancy Ph, Boris S, Manac'h Y, Aumont C. Fistule de la 4.^a poche endobranchiale. Rev Laryngol Otol Rhinol. 1984;105:491-4.
5. De Ponte FS, Brunelli A, Marchetti E, Bottini DJ. Sublingual epidermoid cyst. J Craniofac Surg. 2002;13:308-10.
6. New GB, Erich JB. Dermoid cyst of the head and neck. Surg Gynecol Obstet. 1937;65:48-55.
7. Lima SM Jr, Chrcanovic BR, de Paula AM, Freire-Maia B, de Souza LN. Dermoid cyst of the floor of the mouth. Scientific W J. 2003;3:156-62.
8. Longo F, Maremonti P, Mangone GM, De Maria G, Califano L. Midline (dermoid) cysts of the floor of the mouth: report of 16 cases and review of surgical techniques. Plast Reconstr Surg. 2003;112:1560-5.
9. Gibson WS, Fenton NA. Congenital sublingual dermoid cyst. Arch Otolaryngol. 1982;108:745-8.
10. King R, Smith B, Burk J. Dermoid cyst in the floor of the mouth. Review of the literature and case reports. Oral Surg. Oral Med Oral Pathol. 1994;78: 567-76.
11. Miles LP, Naidoo LC, Reddy J. Congenital dermoid cyst of the tongue. J Laryngol Otol. 1997;111:1179-82.
12. Wiersma R, Hadley GP, Bosenberg AT, Chrystal V. Intralingual cysts of foregut origin. J Pediatr Surg. 1992;27:1404-6.
13. Solazzo L, Madaro E, De Giovanni PP, Berrone S. Dermoid Cyst of the oral floor. A clinical case report and review of the literature. Minerva Stomatol. 1994;43:171-7.
14. Gil E. Quiste dermoide sublingual. Anales ORL Ibero-Am. 2000;27:215-21.
15. Riera C, Doménech E, Valladares J. Quiste dermoide del suelo de la boca. Acta Otorrinolaringol Esp. 1999;50:78-80.