CASE REPORT

Floor of mouth dermoid cysts: a pediatric case series

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ABSTRACT

Tumors in the oral cavity in the pediatric age usually correspond to the persistence or alteration in embryonic formation. They are frequently benign and asymptomatic neoplasms, which present clinically as an increase in the volume of the soft tissues at the level of the floor of the mouth that can even protrude through the submental region. When they have considerable volumes, it can cause swallowing, speaking, or breathing limitations. We present the case of six pediatric patients with a disease duration of up to 8 years, characterized by a tumor at the submental level, neither visible nor palpable in the oral cavity, completely asymptomatic. All underwent surgery. With a quick recovery, without complications, and presenting an adequate aesthetic and functional result, the patients have returned to their social activities without any problems. After one year of follow-up, there was no evidence of recurrence.

Keywords: Dermoid Cyst; Pediatrics; Mouth Floor; Surgery, Oral (Source: MeSH)

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Quistes dermoides de piso de boca: una serie de casos pediátricos

RESUMEN

Los tumores en cavidad oral en edad pediátrica suelen corresponder a la persistencia o alteración en la formación embrionaria. Frecuentemente son neoplasias benignas y asintomáticas, que se presentan clínicamente como un aumento de volumen de las partes blandas a nivel del piso de la boca que pueden incluso protruir a través de la región submentoniana. Cuando tienen volúmenes considerables puede llegar a ocasionar una limitación en la deglución, habla o respiración. Presentamos el caso de seis pacientes en edad pediátrica con tiempo de enfermedad de hasta 8 años, caracterizados por una tumoración a nivel submentoniana, no visible ni palpable por la cavidad oral, completamente asintomática. Todos fueron sometidos a una intervención quirúrgica. Con una recuperación rápida, sin complicaciones, y presentando un resultado estético y funcional adecuado, los pacientes han reingresado a sus actividades sociales sin mostrar inconvenientes. Posterior a un año de seguimiento, no se evidenció la presencia de recidivas.

Palabras clave: Quiste Dermoide; Pediatría; Suelo de la Boca; Cirugía Bucal (Fuente: DeCS)

INTRODUCTION

Floor-of-mouth cysts are tumors that occur in the oral cavity with extension to the submental region and are usually located in the midline. Their origin can be congenital or acquired. The congenital form originates from embryonic cells of the first or second branchial arch. The acquired form may be due to trauma or iatrogenic causes. However, tumors at this level may be due to different pathologies, such as ranula, neurofibromas, hemangiomas, and lymphangiomas (1,2).

Patients with cysts of the floor-of-mouth are usually asymptomatic according to their size, although large tumors cause dysphagia, odynophagia, speech limitation, or dyspnea. The main sign is the presence of the tumor that the patient presents on physical examination, even showing a double chin. There is no gender preference, and it has been reported more frequently

in adolescents. Congenital cysts of the floor of the mouth, derived from the fusion of germinal lines, correspond to benign tumors trapped from the remnants of the germinative layers of the ectoderm, mesoderm, or endoderm due to a fusion defect between the first and second pharyngeal arch around the fifth week of embryonic development (1,3).

Gordon *et al.* (4) distinguished three histologic variants of congenital cysts of the floor of the mouth: the epidermoid cyst, which is lined by stratified squamous epithelium without dermal appendage; the dermoid cyst which has a cavity lined by stratified squamous epithelium, with dermal appendages (hair follicles, and sweat and sebaceous glands) and the teratoid cyst whose lining varies from simple squamous epithelium to respiratory ciliated epithelium and contains derivatives of ectoderm, mesoderm and endoderm (muscular, osseous, cartilaginous, dental tissue, etc.). However, macroscopically, all three histological types contain a thick material with a greasy appearance and yellowish coloration (1,2,4).

They usually occupy one or three areas of the submental space (between the anterior bellies of the digastric muscles and below the mylohyoid), sublingual space (below the lingual mucosa and above the mylohyoid), or lingual midline (potentially virtual junctional space in the tongue musculature). Sometimes, they can even form an image similar to an hourglass (2,5).

Treatment is surgical, taking into account the location and size of the tumor, with intraoral access for lesions above the muscular plane of the mylohyoid or submental if below the mylohyoid, and even in combination, if necessary, due to the considerable volume of the tumor (6-8). In this report we present six cases of pediatric patients clinically diagnosed with dermoid cyst of the floor of mouth, resolved by surgery, with no evident recurrence after one year of follow-up.

Pediatric patients characteristics

Six cases of pediatric patients with a clinical diagnosis of mouth floor cysts during a period of 8 years (2015-2023), seen in the Specialty of Head, Neck, and Maxillofacial Surgery, were presented. The patients were between 3 to 12 years old, with an average of 7.5 ± 3.02 years, being 66.7 % (n=4) female (see Table 1). All patients had an asymptomatic cervical tumor, and a CT scan was performed to determine the volume, dimensions, location, and density of the tumor lesion. All six patients underwent surgery under general inhalation anesthesia with orotracheal intubation. Additionally, cephalothin at 30 mg/kg body weight was used as a prophylactic antibiotic after anesthetic induction and before the surgical incision, either intraoral or cervical. The patients remained hospitalized for one day for monitoring of hematomas compromising the airway, with a laminar drain for 24 hours in the case of the intraoral incision and for 48 hours for the cervical incision, according to the location of the tumor (see Table 1). All patients presented similar clinical characteristics and a similar diagnosis and evolution. All were followed up for one year without recurrence.

Table 1. Characteristics of the six interventional pediatric patients

Case	Age	Sex	Disease time	Location	Size (tomography)	Incision
1	12 years	Masculine	8 years	Above and below the mylohyoid	41x27x40mm	Submental
2	7 years	Femenine	4 years	Above and below the mylohyoid	33x28x29mm	Submental
3	6 years	Masculine	4 years	Below the mylohyoid	38x30x25mm	Submental
4	3 years	Femenine	2 years	Above the mylohyoid	25x21x27mm	Mouth floor
5	8 years	Femenine	5 years	Below the mylohyoid	40x37x29mm	Submental
6	9 years	Femenine	6 years	Above and below the mylohyoid	37x30x38mm	Submental

Case report N° 1

We present the description of one of the cases, a 12-year-old male patient who had the tumor for approximately eight years. The patient had a tumor at the level of the submental region, of slow growth, and without limiting feeding and speech. The patient did not present pain or acute symptoms, so he never needed pharmacological treatment. The consultation was due to the increase in volume of the soft tissues in the submental region, with no visible or palpable tumor in the oral cavity. On physical examination, a mobile semi-solid mass with defined borders was palpable, with a slightly firm consistency, approximately 3 cm in diameter, without compromising the color or texture of the skin. There was the absence of mobilization of the tumor mass with tongue protrusion and swallowing. There was no other alteration or discomfort. Due to the physical inspection findings, a contrast-enhanced CT scan was requested. A cystic image measuring 41 x 27 x

40mm, with defined borders and homogeneous content, was described at the level of the base of the tongue in front of the hyoid bone. According to the evaluation of tomography images, it was concluded that the tumor lesion was compatible with a dermoid cyst located on the floor of the mouth (Figure 1).

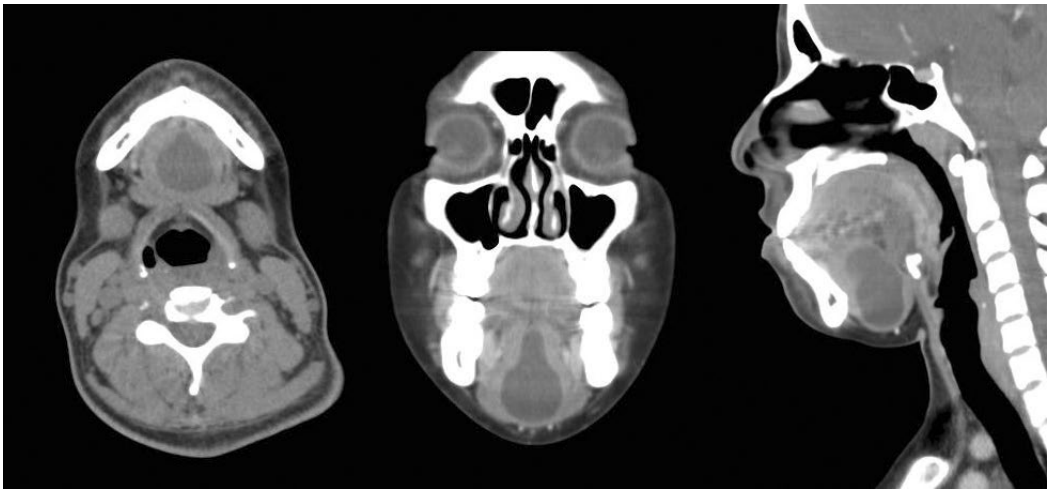

Figure 1. Patient's tomographic images (Case N° 1)

Axial (left), coronal (center), and sagittal (right) views. In the midline, there is a tumor with defined borders that protrudes towards the submental region

The patient was operated with orotracheal intubation and in the cervical hyperextension position. A transverse incision of about 20 mm was made at the level of the most prominent portion of the submental mass. With a blunt dissection, it was disclosed along the midline of the mylohyoid, and with a pressure maneuver at the base of the tongue by the assistant, the protrusion became evident and more accessible for dissection (Figure 2). With sufficiently long retractors, the lingual tissue and the uppermost portion of the cyst were reached. Without further surgical bleeding, the tumor was removed. Then, hemostasis was verified, a laminar drain was placed for 48 hours, and the tissues were closed in planes according to the muscular and fascial structures. The cyst's whitish, lumpy content was macroscopically observed, which was compatible with a dermoid cyst. The patient was discharged, and the drain was removed after 48 hours without evidence of active bleeding or seroma. The stitches were removed seven days later, and the patient returned to his activities without restrictions.

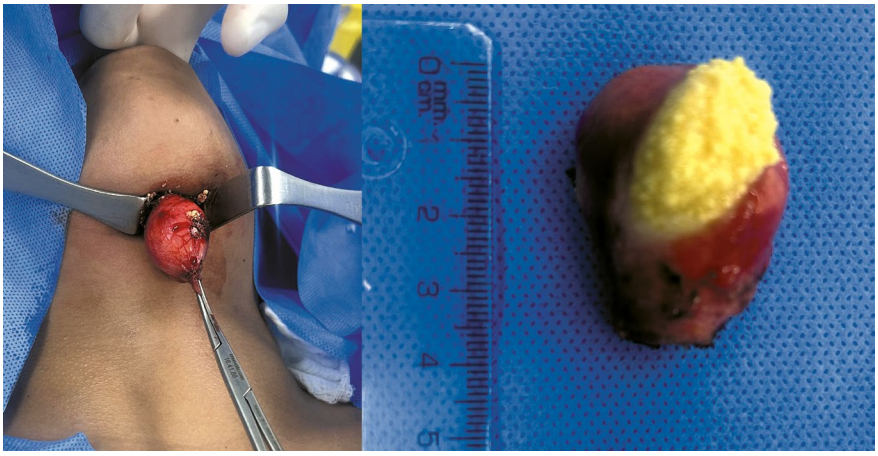

Figure 2. Surgical excision of dermoid cyst (Case N° 1)

Surgical access via the submental approach, the specimen is partially opened to visualize its granulomatous content

DISCUSSION

Tumors at the level of the floor of the mouth in pediatric age can have diverse etiologies and are even congenital. However, they are not detected at an early age due to the absence of problems during feeding or speech. Although they are diagnosed in the first two decades of life, for their clinical detection, they must already have a volume that makes them visible or even palpable (1,2). Our report presents a relatively wide age range, ranging from 3 to 12 years of age, with the majority being female. Kyriakidou *et al.* (9) describe a bimodal age distribution, with a higher frequency in adolescence and a lower frequency during the first years of life; they also found no predilection of presentation according to sex.

Surgical excision is usually the treatment of choice, either orally if the tumor is above the mylohyoid muscle or cervically if it is below the muscular plane (10). The differential diagnosis includes neoplasms, infections, and developmental processes. These include ranulas, submandibular gland blockage, sublingual and minor salivary gland neoplasia, hygroma, acute infection, neurofibroma, hemangioma, and lymphangioma (11,12). Since its presentation has no clinical particularities, it should be supported by diagnostic imaging. It is even recommended that the diagnosis include histological reports to know the cell line involved (13).

Conclusions

We report six cases of oral floor cysts in pediatric patients that were successfully treated with surgical excision. The one-year follow-up of all patients showed no recurrence.

Authors' contribution

Conceptualization: JFOA; data collection, management, and curation: JFOA, ESC; data analysis: JFOA, ESC, KCCC; visualization: JFOA, ESC, KCCC; drafting of the original version: JFOA, ESC, KCCC; drafting and revision of the final version: JFOA, ESC, KCCC; funding: JFOA.

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Ethical aspects

The authors declare that they have obtained consent from the patients' parents or guardians to use their patient information. The present study complied with ethical standards set out by the Declaration of Helsinki.

Conflicts of interest

The authors have no conflicts of interest associated with the material presented in the manuscript.

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