Recurrence of Congenital Dermoid Cyst of Paramedian Location

Recidiva de Cisto Dermoide Congênito de Localização Paramediana

Estevam Rubens Utumi*, Camila Eduarda Zambon**, Irineu Gregnanin Pedron, Gustavo Grothe Machado****, André Caroli Rocha*****.***

* Master in Dental Science with specialization in Integrated Clinic. Maxillofacial Surgeon Brazilian Air Force, Air Force Hospital in Sao Paulo.
** Master in Medical Sciences at the Hospital das Clinicas, FMUSP. Specialist in Maxillofacial Surgery.
*** Master of Dental Science with specialization in Integrated Clinic. Periodontist clinical staff of the Brazilian Air Force, Air Force Hospital in Sao Paulo.
**** Master in Medical Sciences at the Hospital das Clinicas, FMUSP. Chief of Maxillofacial Surgery of Trauma Hospital of Medical College, USP.
***** Ph.D. in Oral Diagnosis, Faculty of Dentistry, University of São Paulo. Service of Traumatology and Maxillofacial Surgery, Hospital das Clinicas, FMUSP.

Institution: Hospital of the Faculty of Medicine, University of São Paulo.
São Paulo / SP - Brazil.
Mail Address: Esteban Rubens Utum - Rua Pelotas, 284 - Apt. 21 - Vila Mariana - São Paulo / SP - Brazil - Zip code: 04012-000 - Telephone: (+55 11) 5549-8241 - E-mail: estevamutumi@uol.com.br
Article received on April 29, 2009. Article accepted on May 24, 2009.

SUMMARY

Introduction: The dermoid cyst is a cyst of unusual development in the face and more often involves the mouth floor. Most injuries occur in young adults with a slight predilection for males. Lesions in neonatal and children are extremely rare. It manifests as swelling floating asymptomatic and slow-growing and progressive, reaching dimensions. His capsule may contain one or more skin appendages such as sebaceous glands, hair or nails. The treatment is surgical, by enucleation, and its recurrence uncommon. The clinical appearance, histopathology, differential diagnosis and treatment of a case of congenital dermoid cyst, and its recurrence, are discussed by the authors.
Keywords: dermoid cyst, mouth floor, recurrence.

RESUMO

Introdução: O cisto dermoide é um cisto de desenvolvimento incomum na face e envolve mais frequentemente o assoalho bucal. A maioria das lesões ocorre em adultos jovens, com ligeira predileção pelo gênero masculino. As lesões neonatais e em crianças são extremamente raras. Manifesta-se como tumefação flutuante, assintomática e de crescimento lento e progressivo, atingindo dimensões variadas. Sua cápsula pode conter um ou mais anexos cutâneos, como glândulas sebáceas, pêlos ou unhas. O tratamento é cirúrgico, através de enucleação, sendo sua recidiva incomum. O aspecto clínico, histopatológico, diagnóstico diferencial e tratamento de um caso de cisto dermoide congênito, e de sua recidiva, são discutidos pelos autores. Palavra chave: cisto dermoide; soalho de boca; recorrência.
Palavras-chave: cisto dermoide, soalho bucal, recidiva.

Introduction

The dermoid cyst is a relatively rare developmental change in head and neck. When the mouth, the most common location is the mouth floor. There is a slight predilection for males and the majority of injuries occur between the second and third decades of life. Congenital lesions and those originating in children are extremely rare (1-7).

Commonly presents as a lump floating in the anterior floor of mouth, with slow, gradual and painless, and may vary Tamanaha (3,4).

The treatment of dermoid cysts of the mouth floor is surgical, by enucleation. So far, few cases of malignant transformation (8) and no recurrence reported. The authors report a case of dermoid cyst with sublingual paramedian location, and its recurrence after enucleation in a newborn.

Case Study

FCS patient 10 days old, caucasian, female was referred by her pediatrician to our clinic for evaluation of swelling in mouth floor that was perceived immediately after birth. There were difficulties in sucking and swallowing during breastfeeding.

On physical examination, she was in good condition and no material changes in medical history and hereditary antecedents. The intra-oral examination, there was nodular submucosal region paramedian left floating on palpation, measuring about 1cm in diameter. The mucosa recobrinte had normal color and texture (Figure 1). With presumptive diagnosis was ranula, it was chosen for monitoring. After three months, no involution, indicating a progressive increase of the nodule, with maintenance of the characteristics described above. At this point, asked whether a CT scan with contrast, which showed a cystic image, crossing the middle line, situated between the genio-hyoid muscles and dislocated left at its most superior. The capsule was thickened and content of low attenuation compared to adjacent soft tissues of the mouth floor (Figure 2). The presumptive diagnosis was dermoid cyst.

Excisional biopsy was performed under general anesthesia and tracheal intubation. It began with an intraoral incision in the longitudinal floor of the mouth, on the injury, followed by blunt with dilatation of adjacent tissues (Figure 3A). At the end of the dissection, there was capsule rupture and leakage of cystic contents pasty whitish (Figure 3B). The removal was completed, not viewing the shop macroscopic residual lesion surgery.

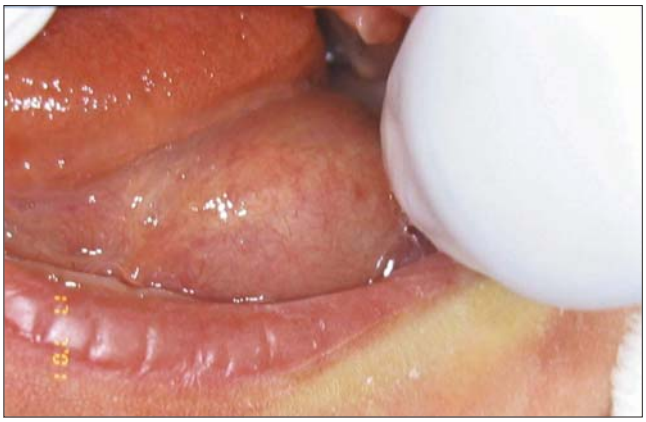

Figure 1. Oral cavity and visualization of the lesion.

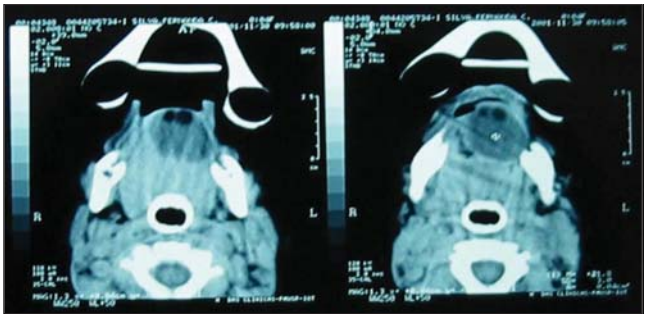

Figure 2. Computed tomography.

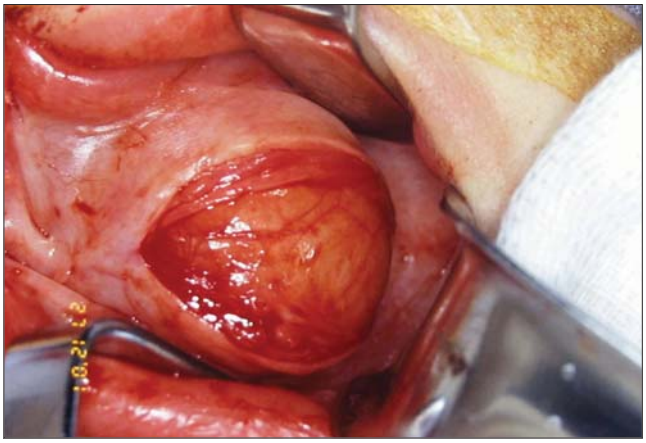

Figure 3a. Exerece injury.

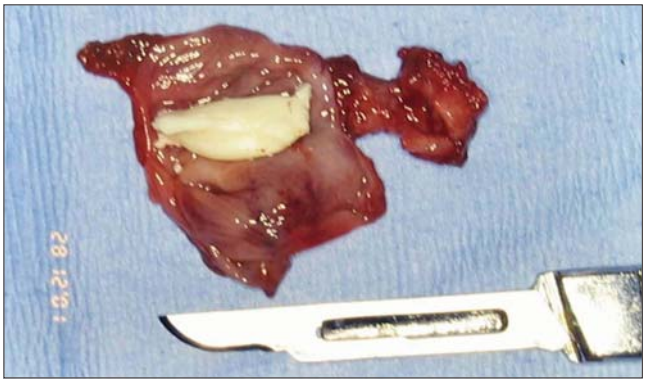

Figure 3b. Mouth injuries.

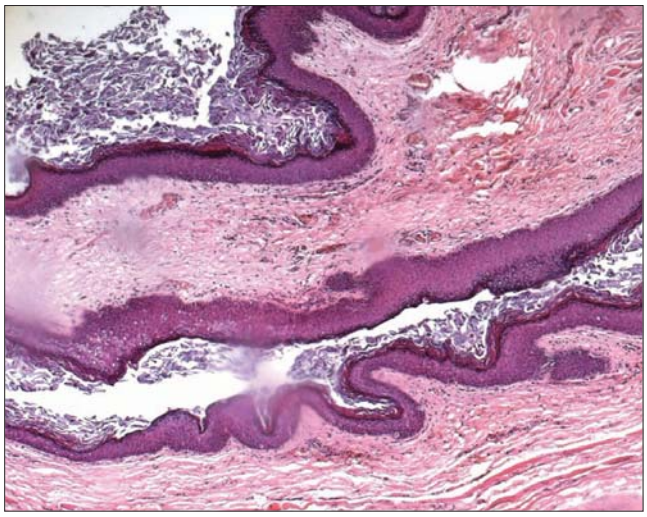

Figure 4a. Cut histopathology.

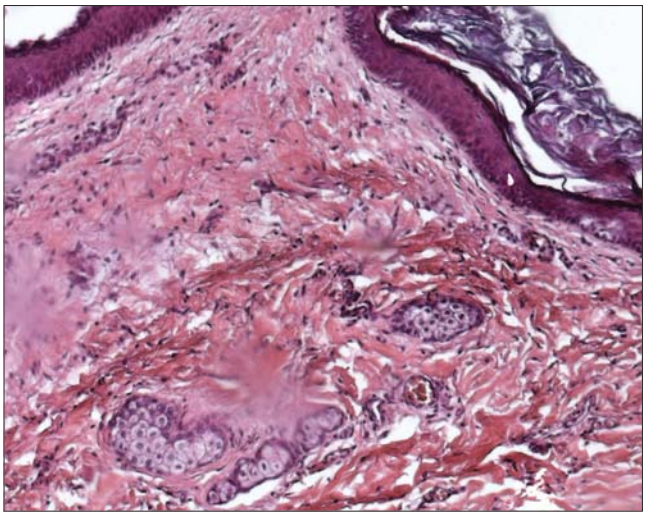

Figure 4b. Cut histopathology.

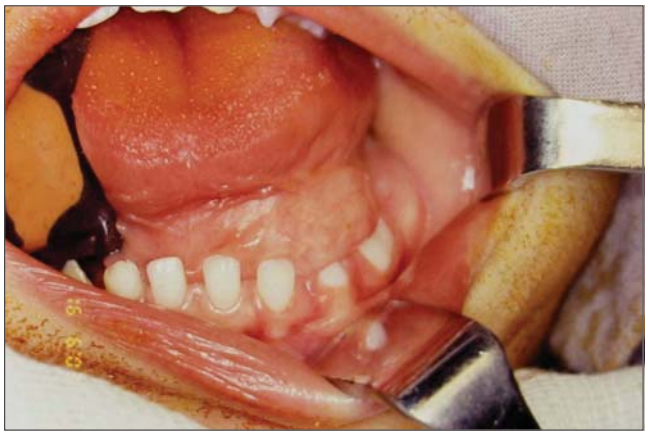

Figure 5. Postoperative appearance

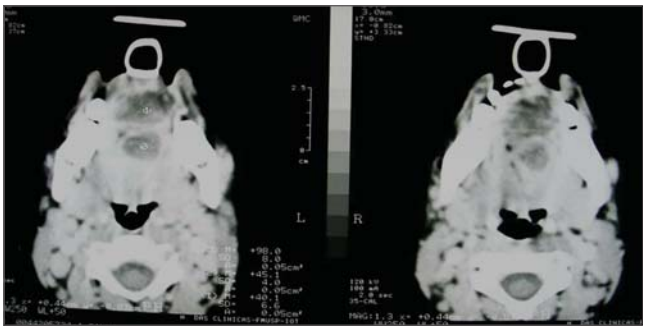

Figure 6. CT scan.

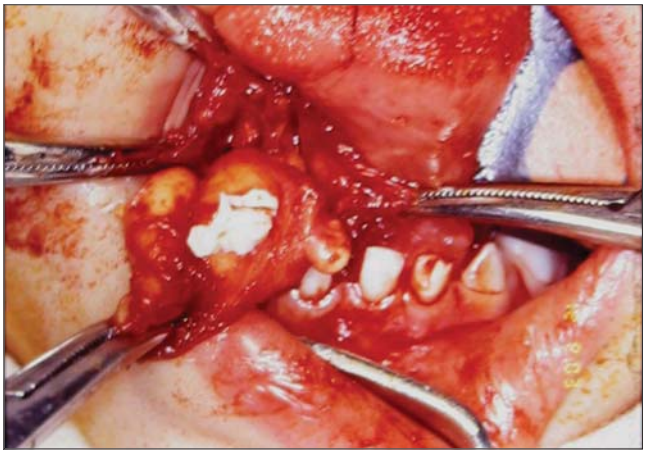

Figure 7. Resection of a second injury.

Histopathology revealed cystic capsule lined by keratinizing stratified squamous epithelium, and skin glands attached. Slight mononuclear cell infiltration was seen in sparse area (Figure 4A and B).

The postoperative course was uneventful and, though 1-year follow-up found to be volumetric increase again in the same region (Figure 5). Another CT scan was performed, showing two well-defined cystic areas in the mouth floor, ceilings and thick with content hipoantenuante in relation to the surrounding soft tissue. (Figure 6). A new surgery under general anesthesia was performed for enucleation of the cyst (Figure 7). The incision was vertical in the midline of ventral tongue and mouth floor. The lesion was removed and featured content pasty, dense and whitish. The histopathologic diagnosis of dermoid cyst was again similar to the previous result. The patient is well without recurrence after four years of follow-up of the last intervention.

Discussion

The first case of sublingual dermoid cyst was described in Jordan in 1778 (5.7). Common locations include the orbit, back of the nose and throat (1). These injuries are rare in the mouth, but when they occur, are

usually found in anterior and middle portion of the mouth floor (2,7,8,9). paramedian view the location for this case confirmed a diagnosis of ranula bias that was only corrected with the observation of the evolving pattern of injury that did not support the initial diagnosis.

Few cases have been reported in children or elderly, and extremely rare in the neonatal period (1,3,4,10). There is no sex predilection, although a study showing more affected men (6). The most likely etiology of dermoid cyst of floor of the mouth is the entrapment of ectodermal tissue in the midline, at the time of the merger of the first (mandibular) and second (hyoid) arches (3.11). In this case, the lesion was congenital, complicating diagnosis.

Classification of the dermoid cysts is based on both anatomic locations as in histopathology. Meyer, in 1955, separated the dermoid cysts into three types: (1) epidermoid cyst - with limited wall by stratified squamous epithelium without associated structures, (2) dermoid cyst - limited by stratified squamous epithelium with dermal annexes such as sebaceous glands and sweat and hair follicles in the underlying connective tissue and (3) teratoma - with cavity bounded by epithelium derived from mesoderm and endoderm, such as muscle, intestinal mucosa, respiratory mucosa, fiber, bone, blood vessels, in addition to dermal annexes typical dermoid cyst (3 , 6,13,14). In 1969, Katz and Passy classified anatomically dermoid and epidermoid cysts of the mouth floor as: (1) Sublingual - located in the midline under the tongue, between the muscles and geniohióideo mylohyoid, and, when it reaches large proportions, the tongue is moved to the oropharynx (2) geniohióideo - located in the midline region submentual between the skin and muscles geniohióideo, leading to an appearance of double chin and (3) lateral cyst - located in the submandibular region and usually increases in size downward to the hyoid bone and compresses the upper floor of the mouth, forcing his tongue to the opposite side (12). The histology of the lesion does not influence the prognosis of the case, however the location, whether above or below the muscle geniohióideo, may hinder the removal by intraoral approach, besides increasing the risk of recurrence if an incomplete removal is performed (3). Thus, Bloom et al. (2002) (1) recommended dual access, intra-and extra-oral, in cases of dermoid cysts side. Moreover, in newborns, the sublingual location may impair the feeding and breathing, producing systemic alterations secondary to initial pathology (5,11). Thus, an anatomical classification should be used to aid surgical planning and prognosis (4). In this case, even with the impression of surgical complete removal of the initial injury, the rupture of the capsule seems to have predisposed the permanence of residual lesion at some level. At 1 year of evolution, this remnant has evolved to the formation of new clinically evident cystic area. Either way, the congenital occurrence, the new growth and the standard CT scan, suggest that this is an injury to more aggressive behavior.

The differential diagnosis should be considered, infection or cellulitis submandibular and sublingual ranula, unilateral or bilateral blockage of Wharton's duct, cystic hygroma, branchial cyst, thyroglossal duct cysts, fat mass in the submental area, neoplasia sublingual or minor salivary gland , lipoma, fibroma, hemangioma or lymphangioma and malignant tumor (6,7,9,15,16). Supplementary tests include ultrasonography, computed tomography and magnetic resonance imaging. These allow visualization and precise localization of the lesion in relation to adjacent anatomical structures, and help in choosing the most appropriate surgical technique (3). This feature provided a visualization of the deeper part of the lesion, which was located in the midline between the geni-hyoid muscles, contributing to the diagnosis of dermoid cyst.

The intra-oral access is used for most cases. However, lesions located below the mylohyoid muscle should be addressed by extra-oral access (4.13). Although this is an injury to an excellent prognosis, the postoperative follow-up of at least three years should be performed in all cases. The evolution of this case reinforces that need.

Final Comments

The dermoid cyst is an uncommon injury occurring in the mouth. Its manifestations and congenital atypical location (paramedian) led to greater diagnostic and therapeutic difficulty.Details tomographic and clinical outcome defined the most appropriate time for surgery. The postoperative follow-up should always be performed.

Bibliographical References

1. Bloom D, Carvalho D, Edmonds J. Neonatal dermoid cyst of the floor of the mouth extending to the midline neck. Arch Otolaryngol Head Neck Surg. 2002, 128:68-7.

2. Bodner L, Woldenberg Y, Sion-Vardy N. Dermoid cyst of the maxilla. Int J Oral Maxillofac Surg. 2004, 34: 453-55.

3. Eplley BL, Bell MJ, Sclaroff. Simultaneous occurrence of dermoid and heterotopic intestinal cysts in the floor of the mouth of a newborn. J Oral Maxillodac Surg. 1985, 43(11): 880-3.

4. Gibson WS, Fenton NA. Congenital sublingual dermoid cyst. Arch Otolaryngol. 1982, 108(11):745- 748.

5. Howell CJT. The sublingual dermoid cyst. Report of five

cases and review of the literature. Oral Surg Oral Med Oral Pathol. 1980, 59:578-580.

6. Longo F et al. Midline (dermoid) cysts of the floor of the mouth: report of 16 cases and review of surgical techniques. Plast Reconst Surg. 2003, 112(6):1560-65.

7. Oygur et al. Oral Congenital dermoid cyst in the floor of the mouth of newborn. Oral Surg. 1992, 74(4-6):627-630.

8. Devine JC, Jones DC. Carcinomatous transformation of a sublingual dermoid cyt. A case report. Int J Oral Maxillofac Surg. 2000, 29:126-7.

9. Howell CJT. The sublingual dermoid cyst. Report of five cases and review of the literature. Oral Surg Oral Med Oral Pathol. 1980, 59:578-580.

10. Nagar H, Baratz M. Congenital sublingual teratoid cyst. Case report. Int J Oral Maxillofac Surg. 1993, 22:44-45.

11. Ferran F, Abifadel M. Kyste dermoid du plancher buccal. Ann Chir Plast Esthét. 1990, 35(1):69-72.

12. Lowry RE, Tempero RM, Davis LF. Epidermoid cyst of the floor of the mouth. J Oral Surg. 1979, 37:271-73.

13. Ceccheti MM. et al. Intraoral enucleation of dermoid cyst- a case report. Rev Pós Grad. 2003, 10(1):88-93.

14. Seah T, Sufyan W. Case report of a dermoid cyst at the floor of the mouth. Ann Acad Med Singapore. 2004, 33(4):77-79.

15. Janjua TA, Goranvalingappa R. Quiz case 1 Submandibular dermoid cyst Arch Otolaryngol Head Neck 1990, 25(11):1270-1272.

16. Verrina G, Carta M. Considerazioni sulle cisti dermoidi del pavimento della bocca. Minerva Stomatol. 1989, 38(1-6):683-686.