Case report

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Self-resolving congenital form of oral lymphoepithelial cyst: case report

Spontan düzelen konjenital oral lenfoepitelyal kist: Olgu sunumu

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Abstract

Oral lymphoepithelial cyst is a rare benign cystic lesion affecting the oral cavity; mainly the floor of the mouth. It was reported to affect patients with a wide age range extending from childhood to geriatrics. It is treated by conservative surgical excision with excellent prognosis. A case of 2-month old infant presented with a congenital asymptomatic white dome-shaped swelling in the floor of the mouth. The lesion was reported to resolve spontaneously with no intervention. To the best of our knowledge, this is the first report of a congenital oral lymphoepithelial cyst. We recommend the follow up of the cases of infants and children as resolution may occur spontaneously with no need for intervention.

Key words: *benign lymphoepithelial cyst, branchial cleft cyst, self-limiting*

Öz

Oral lenfoepitelyal kist, ağız zemini başta olmak üzere oral boşluğu etkileyen nadir bir benign kistik lezyondur. Çocukluktan geriatriye kadar geniş bir yaş aralığına sahip hastaları etkilediği bildirilmiştir. Konservatif cerrahi eksizyon ile mükemmel prognozla tedavi edilir. İki aylık bir bebek ağız zemininde doğuştan asemptomatik beyaz kubbe şeklinde bir şişlik ile başvurdu. Lezyonun hiçbir müdahale olmadan kendiliğinden gerilediği bildirildi. Bilgimize göre bu olgu, bildirilen ilk spontan gerileyen konjenital oral lenfoepitelyal kist olgusudur. Müdahaleye gerek kalmadan kendiliğinden düzelme olabileceği için bebek ve çocuk vakalarının takibini öneririz.

Anahtar kelimeler: *iyi huylu lenfoepitelyal kist, brankial yarık kisti, kendini sınırlayan*

Introduction

Oral lymphoepithelial cyst (LEC) is a rare benign lesion affecting the oral cavity. The floor of the mouth is the most commonly affected intraoral site. It affects a wide age range extending from 2 to 75 years of age. Rare cases

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of children have been reported.[1]

Case report

This report presents a case of a healthy 2-month-old female who was born with a whitish lump under her tongue. The mother reported that the lesion did not affect suckling or cause any pain to the infant.

Examination revealed a well defined, sessile, solitary, round, soft, white, non-tender, non-fluctuant swelling of 0.5 cm diameter, in the floor of the mouth near the orifice of the right submandibular salivary gland duct. (Fig. 1)

The differential diagnosis of the lesion included lymphoepithelial cyst and dermoid cyst. The dermoid cyst is characterized by its rubbery consistency and its location strictly in the midline. As both conditions did not fit the case, lymphoepithelial cyst was believed to be the most probable diagnosis.

An excisional biopsy was indicated as the treatment of choice. However, the risk for performing surgery under general anesthesia for the 2-month-old baby was outweighed by the benefits of performing periodic follow up for the asymptomatic lesion to monitor the progression of lesion size and symptoms till the case becomes operable. The parents provided their informed consent for this management plan. After two weeks of the first visit, the lesion spontaneously resolved during the patient's sleep. The mother reported that the patient woke up free of any oral lesions.

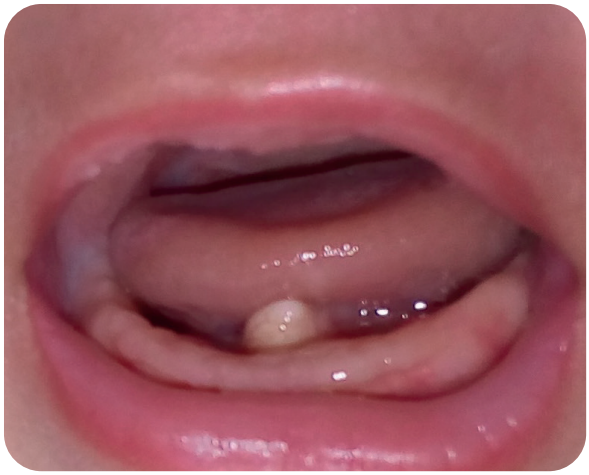

Fig. 1. Clinical presentation of the oral lymphoepithelial cyst

Discussion

To the best of our knowledge, this is the first report for a congenital form of oral LEC. The mean age of the affected patients has been highlighted in the literature as the fourth decade of life; however reports include cases ranging from 2 to 75 years old.[1] Some sporadic cases were reported for patients under 10 years of age.[1-3] However, only McDonnell[2] reported a case of a 5-year-old child who had the lesion "shortly after birth". It also constituted the only article to report spontaneous resolution of the lesion. The author assumed the lesion was exposed to minor trauma causing either rupture into the mouth or herniation through the thin overlying mucosa causing its resolution.[2]

As the name refers, oral lymphoepithelial cyst represents a cystic lesion with both epithelial and lymphocytic components.[4] It has been hypothesized to be caused by either the inclusion of epithelial cells in lymphoid aggregates followed by cystic growth[5], or being a pseudocyst caused by plugging of the crypt opening of lymphatic tissue by desquamated epithelial lining causing swelling.[6]

Reports show that it affects the floor of the mouth the most, followed by the lateral border of the tongue then the ventral surface and soft and hard palates.[1] The preference of the floor of the mouth was attributed to the hypothesis that the cyst originates from the excretory duct of the sublingual salivary gland or from ectopic minor salivary glands.[7]

Clinically, oral LEC is characterized by its presentation as a dome-shaped submucosal nodule with normal non-ulcerated covering mucosa. It has a yellow to white color and soft to firm cheese-like consistency.[1]

Diagnosis of oral LEC is based only on its clinical picture and behavior-namely its color and asymptomatic slowly growing nature- together with its histopathological picture. Imaging techniques are not used for diagnosis; as ultrasonography, computed tomography and magnetic resonance imaging were

reported to be non-conclusive.[1]

Accordingly, a decision should be made to stick to follow up or to perform conservative surgical excision or marsupialization under local aneasthesia. The management decision is based on the judgement of the lesion size and symptoms.[8] Intralesional injection of sclerosing agent was also a proposed line of treatment.[9]

Generally, the lesion has favorable prognosis of no recurrence.[1] However, if traumatized or irritated, the lesion either resolves -as in the hereby presented case- or becomes symptomatic secondary to proliferation of lymphoid tissue.[10]

Although it is always addressed as a rare lesion, the prevalence of oral LEC is thought to be underestimated due to scarcity of reports of such cases. This may be attributed to the small size of the lesions, asymptomatic nature and -according to this report- its occasional self-limiting nature.[10]

Informed consent: The author certifies that he has obtained all appropriate consent forms from the parents of the patient.

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