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CASE REPORT

Intraoral epidermoid cyst with extensive elastofibromatous changes: an unusual finding

Heitor Albergoni Silveira[1] · Luciana Yamamoto Almeida[2] · Matheus Henrique Lopes Dominguete[3] · Kamila Prado Pereira Graciano[3] · Andreia Bufalino[1] · Jorge Esquiche León[4]

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Abstract

Introduction Epidermoid cysts (ECs) are rare and occur in the head and neck regions with an incidence from 1.6 to 7% of all cysts. In the oral cavity, approximately 80 ECs have been reported, representing less than 0.01% of all cysts.

Case report We report a case of a 26-year-old man who developed a large EC in the midline floor of the mouth causing nodular swelling in the submental region and speech and swallowing difficulties. The lesion was surgically excised by intraoral approach and microscopically revealed an EC associated with extensive elastofibromatous changes in the cystic capsule.

Conclusion Oral EC with extensive elastofibromatous changes is a finding extremely rare. The meaning of this finding is unknown, but a traumatic origin or deposit disorder of elastic fibers is suggested. To the best of our knowledge, intraoral EC with elastofibromatous changes has not been reported to date.

Keywords Epidermoid cyst · Oral cyst · Floor of the mouth · Elastic tissue · Elastofibromatous changes

Introduction

Epidermoid cysts (ECs) are rare, classified as development cysts, arising from ectodermal tissue lined by stratified squamous epithelium similar to the skin without adnexal appendages. ECs occur in the head and neck region with an incidence from 1.6 to 7% of all cysts. In the oral cavity, ECs represent approximately 0.01% of all cysts [1, 2]. To date, approximately 80 oral ECs have been reported in the literature. They commonly occur in young adults and rarely in children [1]. Clinically, ECs present as an asymptomatic swelling, slow-growing, fluctuant masses, with the majority of cases arising on the floor of the mouth, followed by the tongue, buccal mucosa, and lower lip [2]. Histopathology shows a cystic wall lined by keratinized stratified squamous epithelium with prominent granular layer and lumen filled by keratin, in a lamellar pattern, with no skin appendages [2].

Elastic fibers constitute one of the main types of connective tissue fibers. Several acquired disorders containing accumulation or degeneration of dermal elastic fibers have been described, including actinic dermatitis, elastoderma, linear focal elastosis, focal dermal elastosis, acquired pseudoxanthoma elasticum, elastosis perforans serpiginosa, and Favre-Racouchot syndrome [3]. Moreover, actinic cheilitis, often affecting the lower lip of adult patients with excessive exposure to the sun, microscopically shows, among other findings, solar elastosis. In the oral cavity, several cases described as elastofibroma or elastofibromatous lesions have been reported. These are tumor-like lesions constituted by elastic and fibrous connective tissue deposition, which seem to represent reactive processes to friction irritation or trauma [4–7]. In addition, elastofibromatous changes have been reported affecting salivary gland neoplasms, oral epithelial dysplasia, and carcinoma in situ [4, 5, 8].

To the best of our knowledge, to date, no intraoral cyst (either odontogenic or nonodontogenic in origin) has been reported presenting elastofibromatous changes.

✉ Jorge Esquiche León
jleon@forp.usp.br

[1] Oral Medicine, Department of Diagnosis and Surgery, Araraquara Dental School, São Paulo State University (UNESP), Araraquara, São Paulo, Brazil

[2] Department of Pathology and Forensic Medicine, Ribeirão Preto Medical School (FMRP/USP), University of São Paulo, São Paulo, Brazil

[3] Universidade Vale do Rio Verde (UninCor), Três Corações, MG, Brazil

[4] Oral Pathology, Department of Stomatology, Public Oral Health and Forensic Dentistry, Ribeirão Preto Dental School, University of São Paulo (FORP/USP), Ribeirão Preto, SP, Brazil

Case report

A 26-year-old male was referred presenting an asymptomatic swelling in the floor of the mouth that had developed over the last 2 years, which caused speech and swallowing difficulties. His medical history was noncontributory. On extraoral examination was observed a "double chin" appearance. Intraoral examination showed a nodular lesion, asymptomatic, located in the midline floor of the mouth, covered by normal-appearing mucosa (Fig. 1). The tongue was elevated due to the presence of the lesion with difficulty to visualize the soft palate. The differential diagnosis included EC, ranula, lymphoepithelial cyst, and salivary gland tumor. Ultrasonography revealed a cystic mass in the sublingual region, measuring 7 × 7 cm in greater diameter, causing the displacement of the sublingual glands. A fine-needle aspiration revealed the presence of a keratin-like yellow material. An excisional biopsy was performed, and microscopy revealed a large EC associated with the presence of extensive elastofibromatous changes in the cystic capsule, corroborated by the strong positivity for Verhoeff-van Gieson stain and foci of florid granulomatous foreign body reaction in close contact to extracellular keratin deposits (Fig. 2). The elastofibromatous changes, such as observed in the current case, are extremely uncommon in EC. The postoperative period was uneventful, and there was no evidence of recurrence or alteration after 1year of follow-up.

Discussion

Among the developmental cysts, the most common are divided into three histopathological subtypes: epidermoid, dermoid, and teratoid types. ECs and dermoid cysts arise from ectodermal tissue lined by stratified squamous epithelium with (dermoid cysts) or without (ECs) skin appendages [1, 9]. In fact, dermoid cysts show epithelial lining enclosing skin appendages such as hair follicles, sebaceous glands, and sweat glands, whereas teratoid cyst contains tissue of nervous, gastrointestinal, and/or respiratory systems [2, 9].

Although the etiology of EC is unknown, some studies associate trauma and prior surgical treatment as important factors in its pathogenesis [1]. In the oral cavity, the floor of the mouth is the most common local, followed by the tongue, buccal mucosa, uvula, and lips [2].

Several intraoral nonodontogenic and odontogenic cysts are recognized, but interestingly, to date, there are no reports of elastofibromatous changes in these cystic lesions. Different from actinic cheilitis, a common lesion in oral pathology often presenting solar elastosis, curiously, some studies have reported elastofibromatous alterations in oral mucosal lesions [4–7, 10, 11].

A review of English-language literature from 2004 to 2018 using the MEDLINE database revealed 12 cases diagnosed as hyperelastosis ($n = 1$), elastofibroma ($n = 3$), and elastofibromatous changes ($n = 8$) of the oral cavity. Of them, 7 were males and 5 were females. The patients' mean age was 65 years (varying from 33 to 98 years). The site most commonly involved was the floor of the mouth (6 cases), followed by the palate (4 cases). Clinicopathological features are shown in detail in Table 1. Interestingly, a detailed review of these cases shows that there is in fact an overlap of the diagnostic criteria, suggesting a continuum of a broad clinicopathological spectrum. Reports of further cases, emphasizing the diagnostic criteria, are necessary to better define this group of oral lesions showing deposits of elastic and collagen fibers.

Elastofibroma and elastofibromatous lesions of the oral cavity are tumor-like lesions composed of elastic and

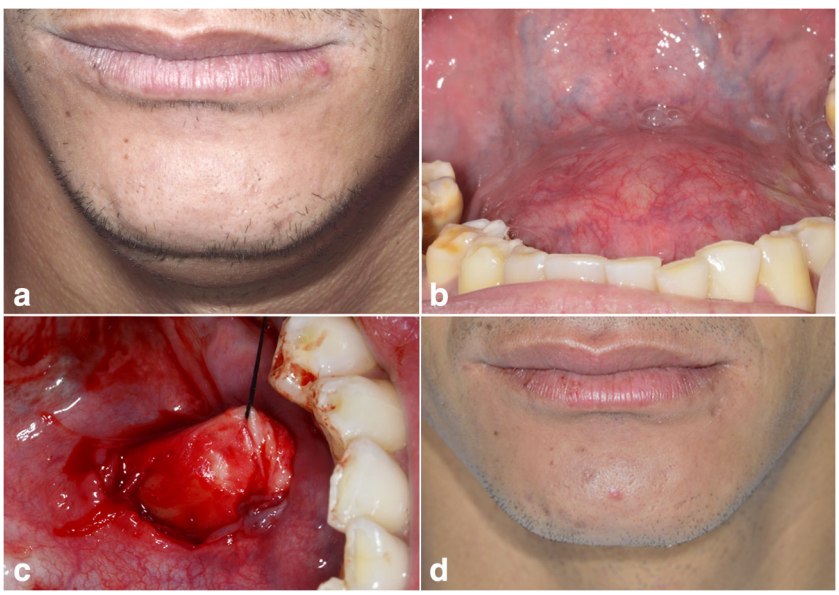

Fig. 1 Clinical features of the intraoral epidermoid cyst showing increased volume giving a "double chin" appearance (**a**). Intraoral view of the cystic lesion (**b**) and during the surgical approach (**c**). Extraoral view after 1 year of follow-up (**d**)

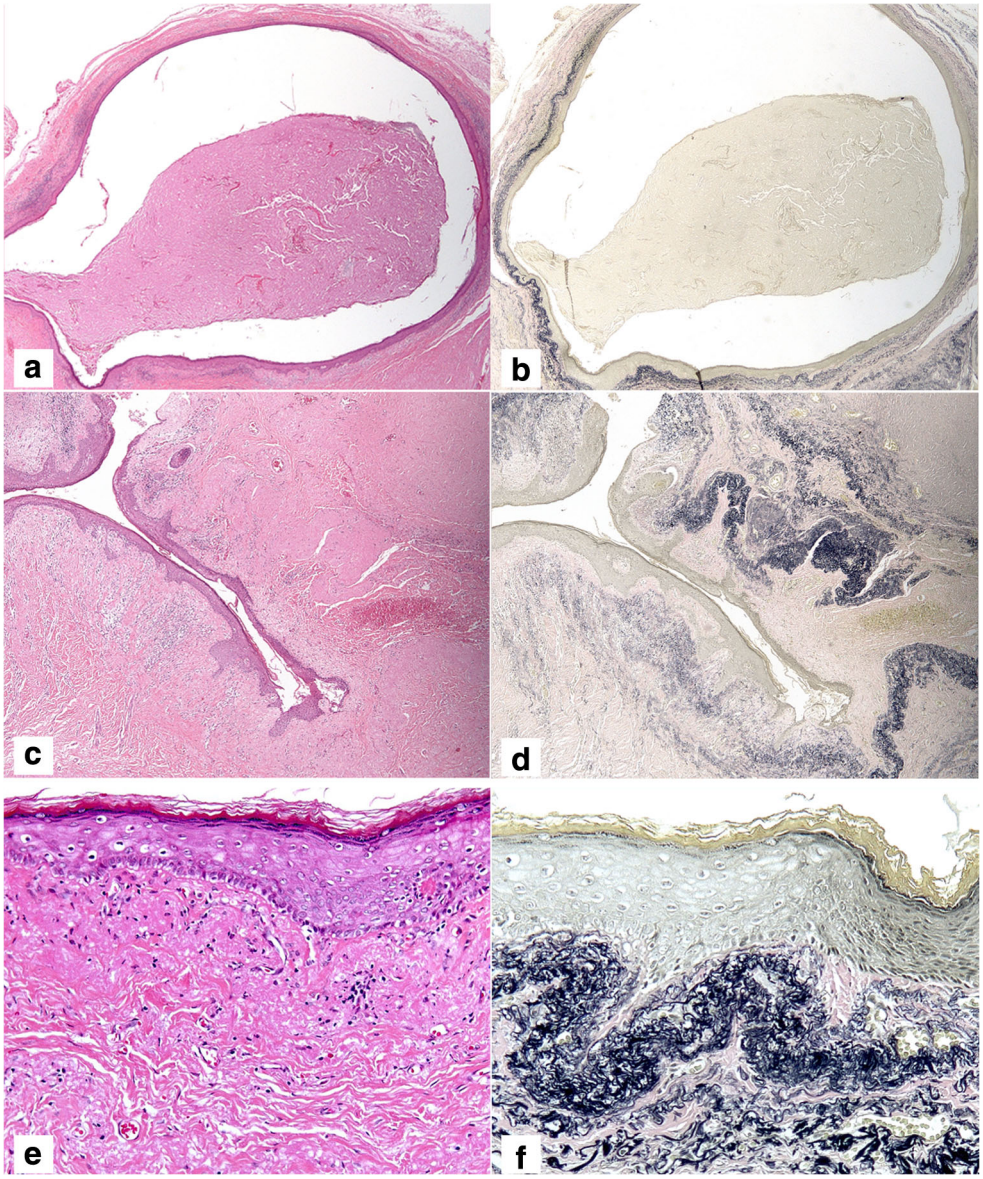

Fig. 2 Histopathological features of the intraoral epidermoid cyst, showing extensive elastofibromatous changes highlighted by elastic stain. Notice that serial consecutive slides were assessed (**a** and **c** H&E stain, × 2.5; **b** and **d** Verhoeff-van Gieson stain, × 2.5). In high-power view, observe histological features of the cystic capsule, typical epithelial lining, and morphological details of elastic fiber proliferation (**e** H&E stain, × 40; **f** Verhoeff-van Gieson stain, × 40)

fibrous connective tissues, which may represent a reactive process associated with friction irritation or trauma [4–6]. Interestingly, some cases reported previous history of squamous cell carcinoma and radiotherapy, with only one case showing association with orotracheal intubation [4–7, 10]. Clinically, elastofibromas are painless, localized, submucosal masses, and usually < 6 mm in diameter [6, 10], while those elastofibromatous lesions clinically mimic localized fibrous hyperplasia, fibroepithelial polyp, and leukoplakia [4–7, 10, 11]. By microscopy, elastofibroma exhibits similar proportions of collagen and elastic fibers, the latter showing a "petaloid" appearance, whereas elastofibromaotus lesion presents predominance of elastic than collagen fibers. Moreover, elastofibromatous alterations have been reported in tumor stroma of salivary gland neoplasms, as well as in subepithelial location of oral epithelial dysplasia and carcinoma in situ [5, 8]. However, after extensive review of the literature, we have not found elastofibromatous changes affecting cystic lesions, and the current case appears to be the first report involving an intraoral EC.

In the current case, to determine the presence of elastic fibers, the Verhoeff-van Gieson stain was performed, which showed strong positivity. However, due to the presence of poorly cellular and weakly eosinophilic areas, exhibiting a finely granular and slightly fibrillary appearance, we suggest distinguishing these alterations from amyloid deposition, as it can mimic elastofibromatous changes based on H&E stain only [12]. In the current case, the Congo red stain and polarizing microscopy was negative.

Table 1 Clinicopathological features of previously reported oral cases diagnosed as elastofibroma, hyperelastosis, or elastofibromatous changes

Author	Age	Sex	Location	Evolution	Clinical features or diagnosis	Previous history	Follow-up	Treatment	Histopathological diagnosis
Potter et al. 2004 [10]	56	F	Floor of the mouth	6 months	White, submucosal mass, asymptomatic	Trauma	2 years	Surgical excision	Elastofibroma
Manchandu et al. 2008 [6]	71	M	Floor of the mouth	NS	White, nodular mass	SCC of the tongue, surgery, radiotherapy	14 months	Surgical excision	Elastofibroma
Tosios et al. 2010 [4]	76	F	Floor of the mouth	NS	Small, flat, white area	NS	NS	Surgical excision	Elastofibromatous changes
Tosios et al. 2010 [4]	98	F	Alveolar mucosa, floor of the mouth	NS	Leukoplakia	SCC of the floor of the mouth	NS	Surgical excision	Elastofibromatous changes
Tosios et al. 2010 [4]	84	F	Floor of the mouth	NS	Ulcer	NS	NS	Surgical excision	Hyperelastosis
Nonaka et al. 2010 [7]	55	M	Soft palate*	6 months	Nodular mass	Orotracheal intubation 1 year earlier	8 months	Surgical excision	Elastofibromatous changes
Darling et al. 2011 [5]	33	M	Palate	NS	Fibroma	Trauma?	1 year	Surgical excision	Elastofibromatous changes
Darling et al. 2011 [5]	43	M	Palate	NS	Condyloma	Trauma?	9 years	Surgical excision	Elastofibromatous changes
Darling et al. 2011 [5]	50	M	Floor of the mouth	NS	Leukoplakia	Trauma?	3 years	Surgical excision	Elastofibromatous changes
Darling et al. 2011 [5]	76	F	Lower lip	NS	Recurrent SCC	SCC in lower lip, surgery, radiotherapy	10 years	Surgical excision	Elastofibromatous changes
Darling et al. 2011 [5]	75	M	Tongue	NS	Fibroma	Trauma?	6 months	Surgical excision	Elastofibromatous changes
Daley and Darling 2011 [11]	62	M	Hard palate	NS	Fibroma, fibroepithelial polyp	Trauma?	NS	Surgical excision	Elastofibroma
Current case	26	M	Floor of the mouth	2 years	EC, ranula, salivary gland tumor	None	1 year	Surgical excision	EC with extensive elastofibromatous changes

NS not stated, *SCC* squamous cell carcinoma, *EC* epidermoid cyst

*This case is located in the oropharynx

The pathogenesis of elastofibromatous lesions is not well known; however, it is suggested that these groups of lesions are of reactive and/or degenerative origin because of their association with ulcerative and/or inflammatory processes [13]. In the same pathway, evidences suggest that elastofibromas are degenerative pseudotumors, which result from the excessive formation of collagen and abnormal elastic fibers [5, 13]. The pathogenesis of the current case is uncertain, and these theories could not adequately explain the elastofibromatous changes, especially if we consider their developmental cyst nature. Surgical excision is the treatment of choice, and recurrence has not been reported [5], as it happened with the current case.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent Informed consent was obtained from all individual participants included in the study.

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