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Case Report

Nasolabial cyst in a patient with cleft lip and recurrent dacryocystitis

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ABSTRACT

Nasolabial cysts are uncommon non-odontogenic cysts that appear mainly in females of the fourth and fifth decade of life. This paper presents a 45-year-old female with a nasolabial cyst, who also had ipsilateral cleft lip and a history of recurrent dacryocystitis.

1. Introduction

Nasolabial cysts are rare non-odontogenic cysts with undefined etiology and female preponderance. They appear as swelling at the nasolabial fold region, they grow slowly and can lead to facial deformity. They cause elevation of the nasal ala and the upper lip. Intraorally they obliterate the gingivolabial sulcus. Treatment of choice is excision. The purpose of this paper is to present a case report of a 45-year-old female with nasolabial fold swelling, who had undergone cleft lip repair surgery and external dacryocystorhinostomy.

2. Case report

In November 2016, a 45-year-old woman self-referred to the outpatient department of the Oral and Maxillofacial Surgery Clinic, due to painless swelling of the left nasolabial fold for a month (Fig. 1). She reported no toothache or pain during mastication. She had undergone two surgical interventions for cleft lip repair during infancy and adolescence. In 2014 she had undergone an external dacryocystorhinostomy procedure with insertion of silicone tube stents, due to recurrent dacryocystitis. She was a smoker and received no other medication.

Intraoral examination revealed a fluctuant, firm, painful mass only during palpation, which bulged in the left labiogingival sulcus (Fig. 2).Tenderness to percussion or mobility of adjacent teeth was absent. A panoramic radiograph was performed, with non-specific findings (Fig. 3). Computed tomography revealed postoperative findings of surgical intervention at the area of the left nasolacrimal duct with disorder of the architecture. Under the subcutaneous tissue, next to the left nasal ala, a cystic cavity (2.5 cm × 1.5 cm) was recognized with peripheral contrast enhancement. Depression of the adjacent bone was also noticed (Figs. 4 and 5).

The cyst was removed under general orotracheal intubation, via an intraoral approach. After injection of lidocaine 2% with 1/100,000 adrenaline solution and bimanual palpation of the lesion, an incision was carried out at the upper gingivolabial sulcus. The mucosa was carefully dissected, until the cyst was exposed. The lesion was enucleated, without rupture. The adjacent pyriform aperture was intact, while the underlying bone was depressed, without defect. The cystic wall was removed along with the periosteum, due to adhesions.

Histopathologic examination revealed a cystic cavity that consisted of cuboidal and pseudostratified columnar ciliated epithelium of respiratory type with some mucoid cells. The cystic wall was made up of dense and loose connective tissue. Mild diffuse chronic inflammatory cells infiltration was also observed (Fig. 6A, B). These findings were consistent with a diagnosis of a nasolabial cyst.

3. Discusssion

Nasolabial cysts are rare, non-odontogenic cysts, that grow slowly and appear from 12 to 75 years old, with a mean age of 40–60 years old. Apart from the aneurysmal bone cyst, the nasolabial cyst is the only one in the oral region with a female predominance (female/male ratio 3/1) [1]. To the best of the authors knowledge this strong female bias has not been clarified yet. Chandrasekharan et al. claim that there is a possible hormonic factor, because these are common in perimenopausal women [2].

Some authors claim that there is no preference for the right or the

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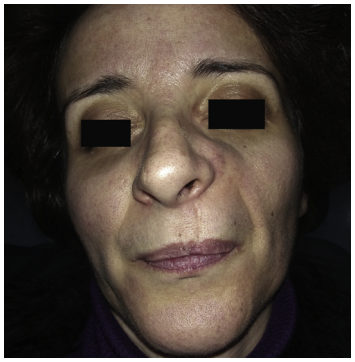

Fig. 1. The patient at referral to the outpatient clinic. Swelling of the left nasolabial fold and conspicuous scar in the upper lip from cleft lip repair.

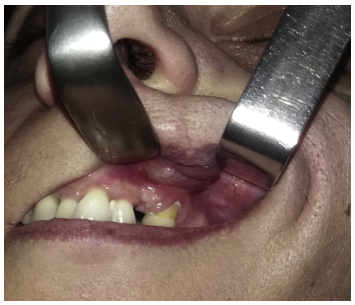

Fig. 2. Intraoral examination revealed a fluctuant, firm, painful mass, which bulged in the left labiogingival sulcus.

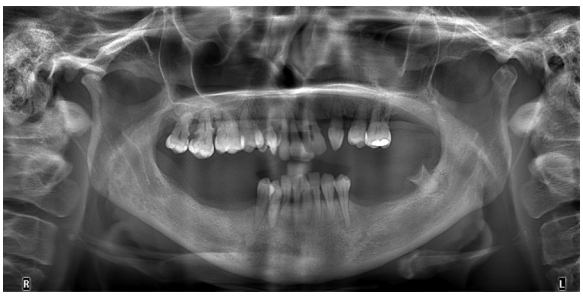

Fig. 3. Panoramic radiograph of the patient revealed non specific findings.

left side [3]. However on a systematic review of 311 cases a slight prevalence of the left side was noticed. The etiology for this difference remains unclear [4]. In 10% of these cases they are bilateral. Their prevalence is 0.7% among the cysts of the maxilla or the mandible, though this percentage is thought to be higher due to misdiagnosis [4,5].

The formation triggering of these cysts is not clear. Their genesis is thought to be triggered by trauma or infection. These events stimulate inert epithelial cells to form into a cyst [6]. Zuckerkandl reported the first case in 1882. Rao proposed the term nasolabial cyst and described 9 cases, 3 of which had a history of surgical intervention in the area involved [7]. Kyrmizakis et al. correlate the presence of bilateral nasolabial cysts with chronic bilateral dacryocystitis and previous dacryocystorhinostomy [8]. Ozdogan et al reported a 33-year-old man, with bilateral nasolabial cysts, who had undergone rhinoplasty and alar base reduction in the past [9]. To the best of our knowledge two more cases with a history of cleft lip or palate surgery and concomitant nasolabial cysts have been reported so far. The first is of a 20-year-old Japanese female with a cleft lip and palate and the second is a 13-year-old Japanese female with a cleft lip and alveolus. Both authors suggest that the migration of nasal epithelium during previous surgical interventions in the nasolabial area may have contributed to cyst formation [10,11].

Etiopathogenesis of nasolabial cysts is not clearly defined. At first the cyst was thought to originate due to obstruction of glandular ducts. Later, Klestadt suggested that the cyst derives from entrapped embryonic epithelium between the medial nasal, the lateral nasal and the maxillary processes [12,13]. Bruggemann in 1920 suggested that the cyst is formed by the epithelial remnants of the nasolacrimal duct [14].The nasolacrimal duct origin theory is considered to be more favorable, due to the same epithelial lining of the duct and the cyst [15]. Nasolabial cysts are located near the inferior part of the nasolacrimal duct and its opening into the inferior nasal meatus [16].

The cyst is usually asymptomatic, but it may cause pain, when infected [17]. Spontaneous rupture of the cyst may lead to drainage into the nasal or oral cavity [1]. A nasolabial cyst doesn't result in teeth displacement. Nevertheless, Cohen and Hertzanu report a large cyst (greatest diameter 7 cm), which had erosed the underlying bone and was firmly attached to the central and lateral incisors [18].

No radiographic changes are expected, since they appear in soft tissues [19]. Computed tomography and magnetic resonance imaging have been suggested as diagnostic tools. CT is preferable, due to lower cost. Ultrasonography has also been reported [20]. Cure et al. report that a nasolabial cyst appears in MRI (magnetic resonance imaging) as a mass below the nasal aperture, which develops extraosseously and occasionally resorbs the underlying bone [21]. Kato et al. conclude that there are different signal intensities, mainly on T1-weighted sequence, due to different viscosities of intracystic fluid [22]. Tanimoto et al. report that diagnosis should be based on the location and appearance of the cyst, rather than its content [23].

Differential diagnosis includes odontogenic cysts, such as radicular cysts or odontogenic keratocysts, periapical abscesses, soft tissue or salivary gland tumors, dermoid or epidermoid cysts and nasopalatine duct cysts. A periapical abscess or an inflammatory odontogenic cyst correlates with a non-vital tooth and appears with the clinical signs of inflammation. A nasopalatine cyst duct is a non odontogenic, developmental, intraosseous lesion in the midline of the hard palate, while the nasolabial cyst concerns the soft tissues of the nasolabial fold. An odontogenic keratocyst is also an intraosseous lesion, although when there is labial cortex perforation, it could mimic nasolabial cyst in clinical presentation. Epidermoid and dermoid cysts are diagnosed mainly during childhood, while nasolabial cysts appear on the fourth or fifth decade of life. Finally, soft tissue or salivary gland tumors are usually more solid and lack enhancement on computed tomography [6,24,25].

Histopathological analysis of nasolabial cyst reveals lining of pseudostratified columnar, squamous stratified or simple cuboidal epithelium [5]. The fibrous wall can be infiltrated by inflammatory cells and focal squamous metaplasia or apocrine changes may also be encountered [26].

Several therapies have been proposed. The gold standard remains an intraoral surgical excision, via a sublabial approach. Su et al. described a transnasal approach that allows endoscopic marsupialization [27]. Marsupialization has also been proposed for large cysts, although not seldom this is followed by recurrence. Furthermore, sclerotherapy and aspiration have been described. The cyst should be ideally removed intact, although no recurrence has been reported after rupture. Perforation of the nasal mucosa might also happen during surgery [28]. Rao reports that the underlying bone, when the cyst is enucleated looks like polished fovea, due to the continuous pressure of the slow-growing cyst [7].

Nasolabial cysts are rare entities with controversial pathogenesis and diagnostic challenge. In regard to our patient, a history of both cleft lip repair surgery and dacryocystorhinostomy was present. We assume that multiple surgical interventions in this area have contributed to the genesis of a nasolabial cyst.

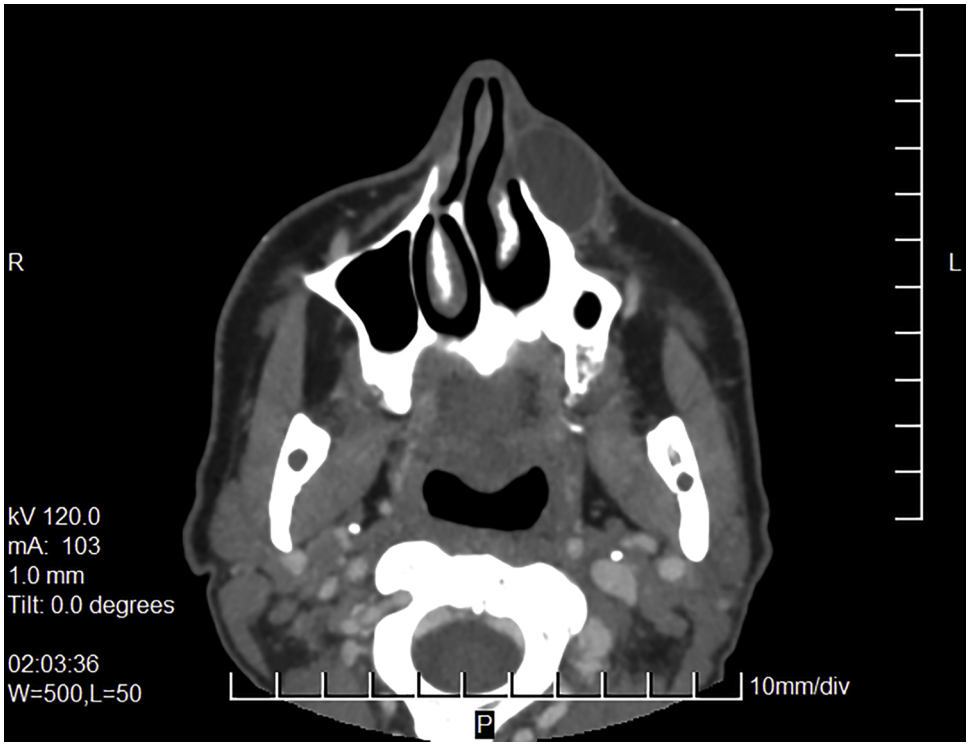

Fig. 4. Computed tomography, axial view. A cystic cavity (2.5 cm × 1.5 cm) with peripheral contrast enhancement is recognized under the subcutaneous tissue, next to the left nasal ala.

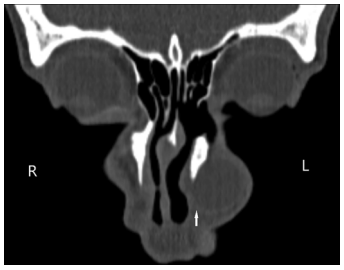

Fig. 5. Computed tomography, coronal view. The white arrow indicates the lesion.

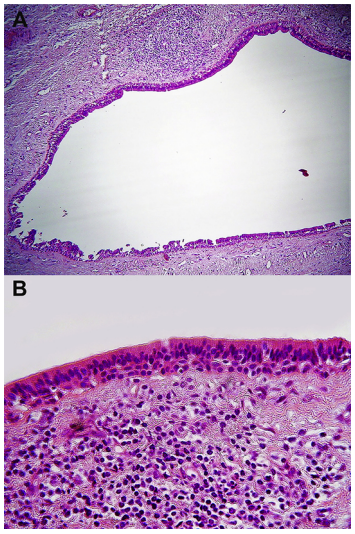

Fig. 6. Histopathologic examination. Hematoxylin and Eosin staining. A) 100× magnification, B) 400× magnification. A cystic cavity that consisted of cuboidal and pseudostratified columnar ciliated epithelium of respiratory type with some mucoid cells. The cystic wall was made up of dense and loose connective tissue. Mild diffuse chronic inflammatory infiltrate was also observed.

Ethical approval

Written consent from the patient has been obtained.

Conflict of interest

None declared.

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References

[1] Shear M, Speight PM. Cysts of the oral and maxillofacial regions. 4th edition Oxford: Blackwell Munksgaard; 2007. p. 120–2.
[2] Chandrasekharan R, Varghese AM, Mathew J, Ashish G. A rare case of bilateral nasolabial cysts in a postpartum lady. Indian J Dent Res 2014;25:225–7.
[3] Schuman DM. Nasolabial cysts: mechanisms of development. Ear Nose Throat J 1981;60:389–94.
[4] Sheikh A, Chin O, Fang C, Liu J, Baredes S, Eloy J. Nasolabial cysts : a systematic review of 311 cases. Laryngoscope 2016;126:60–6.
[5] Choi JH, Cho JH, Kang HJ, Chae SW, Lee SH, Hwang SJ, et al. Nasolabial cyst: a retrospective analysis of 18 cases. Ear Nose Throat J 2002;81:94–6.
[6] Nixdorf DR, Peters E, Lung KE. Clinical presentation and differential diagnosis of nasolabial cyst. J Can Dent Assoc 2003;69:146–9.
[7] Rao R. Naso-labial cyst. J Laryngol Otol 1955;69:352–4.
[8] Kyrmizakis D, Lachanas V, Benakis A, Velegrakis G, Aslanides I. Bilateral nasolabial cysts associated with recurrent dacryocystitis. J Laryngol Otol 2005;119:412–4.
[9] Ozdogan F, Ozel H, Esen E, Yuce T, Baser S, Yavuz C. An unexpected rhinoplasty complication: bilateral nasolabial cyst. Aesthetic Plast Surg 2015;39:889–91.
[10] Aikawa T, Iida S, Fukuda Y, Nakano Y, Ota Y, Takao K, et al. Nasolabial cyst in a patient with cleft lip and palate. Int J Oral Maxillofac Surg 2008;37:874–6.
[11] Suenaga H, Hosokawa R, Saijo H, Hoshi K, Takato T. Nasolabial cyst in a patient with cleft lip and alveolus: a case report. J Oral Maxillofac Surg Med Pathol

2015;27:839–42.
[12] Klestadt W. Nasal cysts and the facial cleft cyst theory. Ann Otol Rhinol Laryngol 1953;62:84–92.
[13] Wesley R, Scannel T, Nathan L. Nasolabial cyst: presentation of a case with a review of the literature. J Oral Maxillofacial Surg 1984;42:188–92.
[14] Kuriloff D. The nasolabial cyst-nasal hamartoma. Otolaryngol Head Neck Surg 1987;96:268–72.
[15] Patil A, Singh A, Nandikoor S, Meganathan P. Bilateral nasolabial cysts - case report and review of literature. Indian J Radiol Imaging 2016;26:241–4.
[16] Toribio Y, Roehrl M. The nasolabial cyst: a nonodontogenic oral cyst related to nasolacrimal duct epithelium. Arch Pathol Lab Med 2011;135:1499–503.
[17] Vasconcelos R, Alencar Souza P, Mesquita R. Retrospective analysis of 15 cases of nasolabial cyst. Quintessence Int 1999;30:629–32.
[18] Cohen M, Hertzanu Y. Huge growth potential of the nasolabial cyst. Oral Surg 1985;59:441–5.
[19] Parwani R, Parwani S, Wanjari S. Diagnosis and management of bilateral nasolabial cysts. J Oral Maxillofac Pathol 2013;17:443–6.
[20] Acar AH, Yolcu U, Asutay F. Is ultrasonography useful in the diagnosis of nasolabial cyst? Case Rep Dent 2014:1–3.
[21] Cure J, Osguthorpe J, Van Tassel P. MR of nasolabial cysts. AJNR Am J Neuroradiol 1996;17:585–8.
[22] Kato H, Kanematsu M, Kusunoki Y, Shibata T, Murakami H, Mizuta K, et al. Nasoalveolar cyst : imaging findings in three cases. Clin Imaging 2007;31:206–9.
[23] Tanimoto K, Kakimoto N, Nishiyama H, Murakami S, Kishino M. MRI of nasoalveolar cyst : case report. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2005;99:221–4.
[24] Narain S. Nasolabial cyst: clinical presentation and differential diagnosis. J Maxillofac Oral Surg 2015;14:7–10.
[25] Tiago R, Maia M, Nascimento G, Correa J, Salgado D. Nasolabial cyst: diagnostic and therapeutical aspects. Braz J Otorhinolaryngol 2008;74:39–43.
[26] Toribio Y, Roehrl M. The nasolabial cyst: a nonodontogenic oral cyst related to nasolacrimal duct epithelium. Arch Pathol Lab Med 2011;135:1499–503.
[27] Su C, Chien C, Hwang C. A new transnasal approach to endoscopic marsupialization of the nasolabial cyst. Laryngoscope 1999;109:1116–8.
[28] Kajla P, Lata J, Agrawal R. Nasolabial cyst: review of literature and a case report. J Maxillofac Oral Surg 2014;13:227–30.