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Case Report

Clinical presentation and management of nasolabial cyst-a case report

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ABSTRACT

Nasolabial cysts are rare, non-odontogenic, soft-tissue cysts that develop between the upper lip and nasal vestibule with an overall incidence of 0.7% out of all maxillofacial cysts. This lesion grows slowly and measures 1 to 5 cm in diameter. Its pathogenesis is uncertain. A cyst that erodes the surface of the maxilla is named nasoalveolar cyst. The predominant presentation of a nasolabial cyst is a painless localized swelling with varying degrees of nasal obstruction, facial asymmetry. Several treatment modalities have been described in the management of the nasolabial cyst. In this paper, we present a case of a nasolabial cyst in a 45-year- female patient that slowly increased in size over a period of 2 years, with associated mild pain and nasal obstruction, and increased rapidly in size since 2 months causing gross facial asymmetry. It had caused a mass effect upon the maxilla, resulting in scalloping. The cyst was excised entirely with no evidence of recurrence at the 9 months follow up. The nasolabial cyst is a rare soft-tissue non-odontogenic cyst. Complete surgical excision using an open approach performed with no recurrence but she developed inferior orbital neuralgia 1 month after surgery which was managed successfully by conservative management with steroids and gabapentin.

Keywords: Nasolabial cyst, Maxillofacial cyst, Non-odontogenic cyst, Enucleation, Soft tissue cyst

INTRODUCTION

Nasolabial cysts are rare benign soft tissue non-odontogenic, extraosseous lesion located in the paramedian region between the nasal vestibule and upper lip.[1] nasolabial cyst has been known by other names such as nasovestibular cyst, nasoglobular cyst and nasoalveolar cyst, nasal vestibular cyst, mucoid cyst of the nose, and Klestadt's cyst, among a variety of other names.[2-4]

Zuckerkandl was the first to describe the cyst in 1882.[1,5] In 1953, Klestadt studied about nasolabial cysts in depth after which lesion became named Klestadt's cyst.[5] Thoma suggested the term nasoalveolar cyst, but it was Rao in 1951 that used the term nasolabial cyst and is now used in the international literature.[6,7]

The incidence of nasolabial cysts is 0.7% of all maxillofacial cysts. It is classified as a developmental, nonodontogenic, extraosseous cyst, and is usually located in the area of the nasolabial sulcus, just below the ala nasi, accounts for approximately 7% of maxillary cysts, 2.5% of non-odontogenic cysts and is unilateral in 90% of cases and 10% bilateral.[1-11] The majority of cysts are situated in the left nasal cavity (48–50%).[3,6,9,10] The size measures 1 to 5 cm in diameter.[12] The majority of cases are diagnosed in African American and Asian subjects in the fourth to fifth decades of life nasolabial cysts predominantly affect women (75% of cases).[2,6,13,14]

The WHO classification defines 3 types of maxillary cysts: 1) epithelial odontogenic cysts, 2) epithelial non-odontogenic cysts; 3) inflammatory radicular and paradental cysts.

Nasolabial cyst, nasopalatine duct and lateral fissure cyst are classified among maxillary epithelial non-odontogenic cysts.

It is not uncommon to misdiagnose nasolabial cysts and not treat them appropriately because of their rarity.

The pathogenesis is uncertain with two main etiological theories have been proposed. In 1920 Bruggemann proposed the most acceptable theory, which suggests that the nasolabial cyst arises from the remnants of the epithelium in the anterior lower part of the nasolacrimal duct.[10,11] This theory.is supported by the fact that the nasolacrimal duct is lined with pseudostratified columnar epithelium, which is found in the cavities of nasolabial cysts.

Another theory suggests its origin as developmental, although it does not manifest until adulthood. Klestadt postulated an embryologic origin for these cysts and considered that these lesions must originate from embryonic epithelium, entrapped in the developmental fissures between the lateral nasal and maxillary processes.[5] Since then, many authors have classified this entity based on Klestadt's embryologic theory as a fissural cyst.[8] However, this theory has been refuted.

Nasolabial cyst is generally asymptomatic and painless and is an incidental finding in 1% of cases. The typical presentation of a nasolabial cyst is a painless localized swelling with varying degrees of nasal obstruction between the upper lip and nasal aperture caused by a smooth and fluctuant, well defined space occupying lesion, elevation of the nasal ala and obliteration of the nasolabial fold.[11] According to recent reviews, nasolabial cyst is rarely diagnosed in Western countries but may be more frequent in others regions, e.g. Eastern Asia.[3,11,15] Patients generally report to clinician for the cosmetic reason or problem of nasal blockage.[13] The mean time to presentation ranges between 30 and 40 months.[3,10]

The most common sign is enlargement causing facial asymmetry due to displacement of the upper lip, with elevation of the ala nasi and effacement of the nasolabial sulcus.[10] Sometimes in cases of acute inflammation, local tenderness, nasal obstruction, and concomitant infection-which can lead to abrupt enlargement of lesion.[16]

The location and presentation of these cysts make them diagnosis nearly clinical exclusively.[16] The diagnosis tests include nasal endoscopy, CT and MRI.[3,6,10,11] Both CT and MRI are valuable in revealing the origin of the cysts and avoids unnecessary needle aspiration or dental surgery.[17,18]

The differential diagnosis includes periapical inflammatory lesions, nasal abscess, nasopalatine duct cyst, dermoid or epidermoid cyst, and salivary gland neoplasm. Pulp vitality testing of the adjacent teeth is useful, as a positive result will rule out periapical inflammatory lesions.[19]

Surgery is equally diagnostic and curative by allowing histological examination.[16] In this paper, we present a case of nasolabial cyst in a 45 year old female presented with soft tissue cystic swelling on the left fascio-maxillary region that slowly increased in size through a period of 2 years, associated with mild pain and nasal obstruction and increased rapidly in size since 2 months causing gross facial asymmetry. CT scan confirmed the clinical diagnosis and managed successfully by enucleation with no recurrence in follow-up period of 9 months.

CASE REPORT

A 45-year-old female presented with a left-sided mass on the face and nose since 2 years, rapidly increasing in size since 2 months, and associated with dental pain since 1 month as in Figure 1-with gross facial deformity and elevation of the ala nasi on the left with partial obstruction of the left nasal cavity. There was no history of epistaxis or nasal discharge. There was no significant present or past history. General and systemic examinations were normal.

Figure 1: Pre-operative photograph revealing gross facial asymmetry.

On local examination there was gross facial asymmetry with elevation of the left ala nasi and bulging of the left nasolabial sulcus and revealed a huge soft cystic swelling in the anterolateral part of the left nasal cavity, with swelling of the left nasolabial fold and left upper lip, side of face in the maxillary region encroaching over the lt ala of nose with complete obstruction of the lt nostril. Intraorally the cystic bulging is seen in upper GBS. On palpation the swelling was soft cystic, non-tender, well circumscribed margins, approx 4 to3 cm in size. All routine blood investigations were normal. CT scan showed a rounded, water-density mass (Figure 2) in the lt nasolabial region with a mean diameter of 2.8×2.8×4 cm with thick fluid consistency. No solid component and no teeth parts are seen.

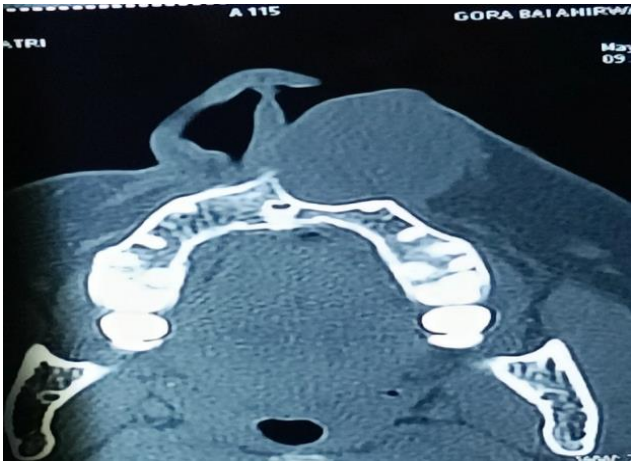

Figure 2: Revealing homogenous cystic lesion with scalloping of the maxilla.

Considering the cystic nature of the lesion, decision was taken for the surgical excision. Under GA, sublabial incision was given, superior and inferior flaps were raised, the cyst capsule was dissected from flap, infraorbital nerve was dissected from the cystic wall. The cyst wall was adherent with alar soft tissue, from where it was excised completely. Post -op uneventful. The patient was discharged on the next day.

Histopathological analysis revealed a cystic lesion, mostly lined with a thin layer of stratified squamous epithelium, with a fibrous capsule and chronic inflammatory changes of varying intensity, consistent with a nasolabial cyst.

After 1 month the patient came with complained of severe radiating pain in the upper GB sulcus especially while eating, brushing. Topical depomedrol injection was given along with systemic deflacort in tapering dose for 9 days along with gabapentin for 7 days. Patient responded well with conservative treatment, and she remained asymptomatic at 9-month follow-up.

DISCUSSION

Nasolabial cysts represent about 0.7% of all cysts in maxillofacial region, and 2.5% of non-odontogenic cysts.[3,7,8]

Three hundred eleven cases were extracted from 79 English language publications between 1961 to2014 (i.e., more than 6 published cases/year) by Sheikh et al.[10]

El-Hamd has reported, however, that some patients reported development of this lesion over three to five years, not having sought medical assistance due to its slow, asymptomatic growth and lack of pain or discomfort and complications, when they occur, generally cause nasal obstruction and esthetic problems.[12] Cohen and Hertzanu described the sudden growth to exuberance of a cyst within 2½ months after one year of slow development, in one of the nasolabial cyst cases and patients only seek therapy when there is deformity, nasal block or infection caused by this lesion.[20,21]

Other significant findings include greater incidence in females (4:1) and possible greater prevalence among blacks.[8]

Clinically the most marked signs, such as effacement of the nasolabial sulcus and elevation of the ala nasi were observed in this case and has been reported as a definitive sign of a nasolabial cyst by Graamans et al.[2,14,16,18] Some patients with a nasolabial cyst may be asymptomatic, but most have at least one of the three cardinal symptoms: partial or complete nasal obstruction, well-circumscribed swelling (both seen in this patient) or localized pain.[3,14]

Arising from this location, the growth of the cysts can be possible in three directions: to the nasolabial fold, the mouth vestibule, and the nasal vestibule.[22]

The signs of the cysts are rather specific comprising of a well localized fluctuating swelling with a cystic consistency in the nasolabial sulcus, maxillary labial fold and the floor of the nasal vestibule.[11,17]

In nearly 30% of patients, the initial presentation is an infection. In one series done by Kuriloff, half of the patients developed an infection.[22] Once infected, the cyst becomes painful and could rupture spontaneously to drain into the oral cavity or nose.[12] The diagnosis of nasolabial cysts is essentially clinical. Bidigital palpation reveals a fluctuating swelling between the floor of the nasal vestibule and the gingivolabial sulcus, which helps to confirm the diagnosis.

The differential diagnosis of nasolabial cysts may include other lesions affecting the anterior maxillary region, such as odontogenic cysts and periapical abscesses and granulomas. Infected nasolabial cysts may be mistaken for furuncle of the nasal vestibule floor. Dermoid and epidermoid cysts should also be considered in the differential, although they are associated with yellow discoloration of the overlying mucosa, whereas in nasolabial cysts, the mucosa conserves its normal pink hue or appears blue-tinged. Furthermore, dermoid and epidermoid cysts are usually diagnosed in childhood, while nasolabial cysts are more common in adult patients.[1]

Physical examination must be completed by radiological examination. Nasolabial cysts do not present any finding on plain radiographs except when it causes significant maxillary bone erosion. Panoramic dental radiography associated with an occlusion view, with/ without opacification of the cyst, is no longer performed.[10]

Computed tomography and MRI can reveal the cystic nature of these lesions in greater detail and extent and relation of lesion to surrounding structures as well as

bone involvement, only one case of carcinoma progressing from a nasolabial cyst has been described in literature.[3,6,10,11]

CT scans usually reveal a homogeneous, cystic lesion, with no contrast uptake, anterior to the piriform aperture. Larger lesions may be associated with bone remodeling of the underlying maxilla.[23] In the case reported herein, CT revealed a well circumscribed cystic lesion in the lateral nasal region. It had caused a mass effect upon the maxilla, resulting in scalloping.

Most studies have reported enucleation as the treatment of choice for nasolabial cysts.[1,22] As they are entirely composed of soft tissues, nasolabial cysts are unresponsive to marsupialization.[12] Alternative treatment modalities have been suggested, including aspiration, cauterization, injection of sclerosing agents, and incision and drainage; however, these methods are associated with high recurrence rates.[24,25] Treatment is surgical resection, performed under local or general anesthesia. Surgery is both diagnostic by allowing histological examination and curative. It consists of en bloc enucleation of the cyst. The most commonly used approach is a vestibular incision underneath the upper lip, without tearing the nasal mucosa or entering the maxillary sinus.[10] The recurrence rate with this surgical technique is less than 1%.

Therefore, in view of the ease of surgical resection and its curative potential, we believe alternative methods should not be employed unless mandated by circumstance.

As in our case the swelling was comparatively large in size and infra-orbital nerve was separated from the cyst wall, the patient complained of moderate severe neuralgic pain in upper GB sulcus, after 1 month following surgery, which responded well to the conservative treatment with steroid and Gabapentin. As nasolabial cysts are usually in close proximity with the floor of the nasal cavity, perforation of the nasal mucosa during surgical excision is not unusual.[22] When very small perforations are caused, they can be left untreated; however, larger ones must be sutured with absorbable sutures so as to prevent development of oronasal fistula.[3,8,16]

Histopathological examination is necessary to confirm the diagnosis of nasolabial cyst.[10] Brown-Kelly first described the histopathology of this lesion in 1898.[26] Histopathological examination reveals that the cyst consists of respiratory epithelium (pseudostratified ciliated cylindrical or stratified ciliated cylindrical epithelium with goblet cells), although squamous metaplasia may occur in infected cysts.[1,13,17,27] In a scanning electron microscopy study of the inner surface of nasolabial cysts, non-ciliated columnar epithelium with basal cells and goblet cells, no cytonuclear atypia is found.[14] The cyst is filled with mucoid fluid produced by goblet cells, with no cholesterol crystals is.[8]

In patient, histopathology revealed a cystic lesion with signs of chronic inflammation, a fibrous capsule, a glossy and smooth inner surface, and yellow-tinged seromucous contents, confirming the diagnosis of nasolabial cyst.[2]

CONCLUSION

Despite the low frequency of nasolabial cysts, it is essential to recognize the key features of these lesions so as to be able to distinguish them from lesions of odontogenic origin and thus enable safe and proper treatment planning. Such key features may be difficult to detect because patients may be asymptomatic, but most exhibit well-circumscribed swelling, localized pain, and partial or complete nasal obstruction on the affected side, elevation of ala nasi and obliteration of the nasolabial sulcus.

The nasolabial cyst can be diagnosed clinically supported by imaging and confirmed by Histopathological examination. After the diagnosis of nasolabial cyst is established, the optimal treatment modality consists of complete excision of the lesion, showing good prognosis and rare cases of recurrence and complications. In the present case the patient had pre-op dental pain and post-op she had developed neuralgic pain which was managed successfully by conservative treatment.

In our opinion, the difficulty in diagnosing this lesion is their rarity as awareness is needed for this rare lesion which can be managed easily surgically, significant morbidity of facial asymmetry can be managed.

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