CASE REPORT

An unusual large nasolabial cyst: A case report and review of literature

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Abstract

The nasolabial cyst (NC) is a soft tissue lesion that is very uncommon developmental, non-odontogenic fissural cyst that is developed in the inferior region of the nasal ala. Its pathogenesis is uncertain, but according to most authors, they instigate from entrapped epithelium inside line of union of lateral nasal, globular and maxillary processes, attributable to unusual changes in entrapped epithelium all along the line of fusion throughout the development process. These cysts are generally unilateral sometimes occur bilateral and grow slowly at a size ranging between 1.5 and 3 cm. Clinically, they may present as a floating tumefaction within the nasolabial sulcus region associated with nasal alar elevation along with involved side upper lip projection. The diagnosis is usually based on the esthetic concern rather than pain. The ideal treatment option is complete surgical excision. This paper presents a rare case of an unusually large NC reported in 65-year-old female patient along with the review of literature and our previous operated NC cases.

Keywords
Developmental cyst, nasolabial cyst, non-odontogenic cyst

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Introduction

Nasolabial cyst (NC) is developmental, soft tissue lesion that is non-odontogenic. It accounts for 0.7% of all jaw cysts and 2.5% of the non-odontogenic cysts. It was first reported by Zuckerkandl in 1882. These lesions were first described in greater detail by Brown–Kelly in 1989 subsequently in 1892 the first case was reported by McBride. Further NC was termed after the name of Klestadt who explored NC in depth.

The exact origin of NC is rather controversial. It is considered to be developed during 4th and 8th weeks of intrauterine life during the development of base of nose and nasal alae from maxillary process; hard palate development by the midline fusion of each maxillary lateral palatine process with the base of the septum, simultaneously initiating formation of the nasal fossa. Any atypical variation at these fusion points initiates the origin to a fissure cyst. Although the most current and widely recognized theory is that it develops at the inferior and anterior portion of the nasolacrimal duct, the stimulus for its initiation is uncertain. In general, females are affected predominantly accounting for 75% of NC cases and commonly observed at the fourth and fifth decades of life. These lesions are frequently unilateral in 90% of cases and are bilateral in remaining 10% of the cases.

The distinctive clinical presentations may include local pain, nasal obstruction, and may possibly lead to tumefaction with considerable inflammation along with infection; a floating collection in the nasolabial sulcus region with an extraosseous presentation, alar nose elevation, projection of upper lip and extending till the ventral inferior part of the pyriform margin.

The differential diagnosis of NC includes odontogenic cysts, periapical abscesses, and granulomas in maxillary anterior teeth area; dermoid and epidermoid cysts. For proper diagnosis, it is important to assess the pulp vitality of the regional teeth since the teeth are vital in cases of NC and non-vital in cases of odontogenic lesions. The overlying mucosa of dermoid and epidermoid cysts may be associated with yellow discoloration, whereas the NC mucosa retains its normal pink hue.

In periapical radiographs, NC may present as a radiolucent area in the apical region of the maxillary incisors, whereas standard occlusal views illustrate posterior displacement of the radiopaque line in relation to the bony margins of the anterior nasal aperture. For a distinct, analysis of the lesion computed tomography (CT) is the imaging modality of choice.

Brown–Kelly in 1898 described the histopathology of this lesion and accordingly the cyst is composed of respiratory epithelium (pseudostratified or stratified ciliated cylindrical...
epithelium), while in cases of infected cysts squamous metaplasia may be seen. Fluid enclosed within cysts is formed by goblet cells.\(^{10}\) The diagnosis of NC is basically clinical. The present case report endeavors to illustrate an unusual sized NC occurring in an elderly female patient along with similar previously operated NCs including two rare cases of bilateral presentations and literature review of NCs.

**Case Report**

A 65-year-old elderly female patient reported to the department with the chief complaint of pain and swelling in upper front labial left tooth region for 1 month. A history of presenting illness revealed a slow growing swelling on inner aspect of lip raising the upper lip on the left side after which patient consulted the local dentist and symptoms relieved on medication but did not get complete treatment; again it aggravated since 1 month.

The patient presented with a stable medical history. There was no history of trauma, fever, or similar swelling elsewhere in the body with any positive history of nasal discharge.

The extraoral examination [Figure 1] revealed a diffuse swelling, present on middle third of face causing obliteration of both nasolabial folds and philtrum. Elevation of the ala of the nose on the left side is seen.

Intraoral examination revealed a solitary well-defined, oval-shaped swelling with no discoloration present on upper labial mucosa wrt 21, 22 and 23 region measuring around 4 cm × 4 cm with obliteration of vestibular region; On palpation, it was soft, fluctuant and tender. Hard tissue examination did not reveal any significant findings and there was no history of any recent or past dental intervention. The electric pulp vitality tests were done irt 21, 22 and 23 which showed an early response to all teeth. The oral hygiene status was good.

On aspiration, blood tinged creamish discharge was present. Intraoral periapical and panoramic radiographs revealed widening of periodontal ligament spaces around the root of 21, 22 and 23 without any obvious radiolucency. The CT scan [Figure 2] of the area involved revealed a swelling of about 4 cm × 4 cm on the left nasolabial region with extension into the floor of the left nasal cavity and probable attachment to the nasal mucosa. The gross examination of the cystic lesion shows external smooth surface mass with rubbery consistency. Based on history and clinical findings, provisional diagnosis of infected developmental cystic lesion was made and the differential diagnosis of benign minor salivary gland pathology, nasal abscess, nasopalatine duct cyst, periapical inflammatory lesions, and dermoid cyst was considered. A clinical diagnosis of unilateral NC was made, and the cyst was excised completely under general anesthesia. The lesion was approached through an intraoral labial vestibular incision extending from 11 to 24 [Figure 3]. The whole lesion was enucleated after aspiration of the cystic contents. The cystic attachment to the nasal mucosa was meticulously dissected with repair of nasal mucosa under direct vision. Later incisions were approximated primarily without any uneventful postoperative recovery. The cut section...
of the biopsy tissue presents cystic and fibrous areas without any papillary projections or focal hemorrhagic areas.

The excised tissue was then fixed in 10% buffered formalin and sent for histopathological examination. About 5 μm thick sections were cut and stained with hematoxylin and eosin.

The histopathology [Figures 4-6] revealed a cystic lesion lined by pseudostratified squamous epithelium and cuboidal epithelium in few areas with goblet cells. There was no sign of keratinization in the tissue. The presence of cilia was observed in few areas. The fibrous capsule was made up of seromucous contents along with signs of chronic inflammation confirming the diagnosis of NC. The patient was advised complete surgical resection of the lesion.

Table 1 gives a summary of the clinical conditions of the present case and also few similar cases operated previously by the author.[11]

Discussion

The NC is a rare developmental, non-odontogenic cyst that most commonly involves the nasal furrow region. Allard explains that in 1882 the first description of this lesion was recorded by Zuckerkandl. It is also named as Klestadt’s cyst, nasal vestibular cyst, nasoalveolar cyst, nasal wing cyst, and mucoïd cyst of the nose.[12] Its etiology is assumed to be based on three theories. In first theory, it was assumed as a retention cyst that arises from the inflamed mucus glands.[13] The second theory was postulated by Klestadt as an embryologic origin theory for these cysts and explained that these cysts originate from embryonic epithelium that gets entrapped in the developmental fissures between the lateral nasal and maxillary processes.[4] Based on this theory, many authors have classified this entity as a fissural cyst. The third theory was postulated by Bruggemann and is the most accepted which states that NCs arises from the remnants of the lower anterior part of the nasolacrimal duct.[2]

Many authors believe that its prevalence is actually higher than that depicted in the literature and however due to the misdiagnosis its indexes remains low.[14] The NC commonly seen in patients with a wide age distribution, but more frequent between the fourth and fifth decades, and females are more affected than males (male:female ratio - ¼:6.5).[15] NCs are usually unilateral, with no specific prevalence of side involvement but also presenting with bilateral involvement approximately around 11% of the cases.[2,16] It occurs more commonly among Afro-Americans. Even though it has a developmental origin, it is generally not obvious until adulthood. It usually occurs in the left-hand side and most cases reported in literature also report more of left side involvement. In 1969, Bhaskar in his study on 3750 maxillary cysts (0.19%) reported seven cases of NCs in 231 fissural cysts (3%) that presented on the left-hand side.[17] Similarly, our case is not typical in relation to age and gender as these are more frequently seen in women in fourth and fifth decades.

The clinical features of the NC include fluctuant swelling in the maxillary labial fold and floor of the nasal vestibule along with

![Figure 4: High power view of cystic lining and wall. The lining is cuboidal epithelium and fibrous connective tissue wall (H and E, ×40)](image)

![Figure 5: High power view of cystic lining and wall. The lining is pseudostratified columnar epithelium with hemorrhagic fibrous connective tissue wall (H and E, ×40)](image)

![Figure 6: High power view of cystic lining consisting of extravasated blood vessels, nerve bundles and chronic inflammatory infiltrate (H and E, ×40)](image)
Table 1: Demographic details of all NCs operated

<table>
<thead>
<tr>
<th>Case</th>
<th>Age (years)/sex</th>
<th>Involved site</th>
<th>Size (cm)</th>
<th>Symptoms presented</th>
<th>Medical history</th>
</tr>
</thead>
<tbody>
<tr>
<td>Present case</td>
<td>65/Female</td>
<td>Unilateral</td>
<td>Left side 4x4</td>
<td>Swelling, alar flare, nasolabial fold obliteration</td>
<td>None</td>
</tr>
<tr>
<td>Previous case 1</td>
<td>62/Male</td>
<td>Bilateral</td>
<td>Right side 2x2, Left side 4x3</td>
<td>Swelling, alar flare, nasolabial fold obliteration</td>
<td>None</td>
</tr>
<tr>
<td>Previous case 2</td>
<td>72/Female</td>
<td>Bilateral</td>
<td>Right side 4x2, Left side 2x2</td>
<td>Swelling, alar flare, nasolabial fold obliteration</td>
<td>None</td>
</tr>
<tr>
<td>Previous case 3</td>
<td>43/Female</td>
<td>Unilateral</td>
<td>Right side 3x2.5</td>
<td>Swelling, alar flare, nasolabial fold obliteration</td>
<td>None</td>
</tr>
<tr>
<td>Previous case 4</td>
<td>54/Female</td>
<td>Unilateral</td>
<td>Right side 5x3</td>
<td>Swelling, alar flare, nasolabial fold obliteration</td>
<td>Hypertension, diabetes mellitus, depression</td>
</tr>
</tbody>
</table>

NC: Nasolabial cyst

oblitration of the nasolabial fold and elevation of the nasal alae. Usually, the NC is asymptomatic and painless except in cases of superimposed secondary infection. It is undisruptive and presents as a very slow-growing swelling that leads to difficulty in nasal breathing in some patients. These slow-growing cysts rarely involve the maxillary alveolus, but one case has been reported with maxillary alveolar erosion and displacement of incisor teeth. Associated teeth are positive to pulp vitality tests and suggest of non-odontogenic causes. Sometimes these cysts may rupture spontaneously and drain their contents into nasal cavity or the nose. In situations relating to cosmetic and nasal obstruction reasons, the patient may seek very late medical advice. In our case, the swelling was slow growing since 2-3 months and the patient sought medical advice when the swelling became very obvious causing asymmetry of the face. The common complaints in these patients are mainly of increased volume in the maxillary labial sulcus, alar nose flaring, and diminished nasolabial sulcus. On intraoral palpation, the swelling appears soft and fluctuant. Clinical differentiating diagnosis should include the developmental, odontogenic, and neoplastic cysts. The odontogenic cysts excluded are (a) primordial, central and lateral dentigerous cysts, (b) apical and lateral periodontal cysts, and (c) residual cysts. The incidence of occurrence of NCs in Indian population is not recognized, and our collection of five cases perhaps is the single largest compilation in the Indian literature.

In the present case, the patient reported with no dental intervention and no intraoral aggravating factors like poor oral hygiene. Even though NC being a soft-tissue cyst can sometimes cause underlying maxillary bone erosion as detected on radiographic examination. These similar signs and symptoms in lesions such as odontogenic cyst or tumor, dental or periodontal abscess and choanal polyp can lead to misdiagnosis. The conventional radiography shows insignificant changes and is usually not detected as these are soft tissue lesions except in cases where erosive changes of maxilla are seen.

Various authors have described different radiological features. Seward states two possible radiographic observations in these lesions: One is increased radiolucency adjoining to the apical area of the incisors, and the second is deformity of the radiopaque line in relation to the lower border of the pyriform orifice. Similarly, Nixdorf et al. substantiate the signs described by Seward using occlusal radiography, whereas contradicting opinions exist among other authors who assert there are no radiographic signs in NC. There was no bony involvement observed in our case.

In CT, magnetic resonance imaging (MRI) scans the cystic nature of these lesions are revealed to a greater detail along with the relationship of the nasal alae with the maxillary bone, as well as bone involvement. In our case, the cyst is seen as an unusually large lesion measuring approximately 4 cm × 4 cm. The surgical line of treatment for these cysts is complete surgical resection. According to the literature documented till date, recurrence is very rare.

The surgical line of treatment includes injecting sclerotic substances, marsupialization, and surgical removal. Surgical excision remains the treatment of choice as large soft-tissue lesion does not respond to marsupialization. During surgical removal perforation of the nasal mucosa may be expected since this cyst develops in close approximation to the floor of the nose. Instead very small perforations occurred can be left untreated and the larger ones must be sutured.

**Conclusion**

On the basis of literature review and the present case report, it can be concluded that, even though NCs are rare but can cause nasal obstruction and cosmetic deformity, plain film radiographs presents little or no information of cystic features and morphology.

Dental practitioners should be competent to recognize and distinguish the essential diagnostic features of these cystic lesions to separate them from developmental, odontogenic and neoplastic cystic lesions and thus facilitate safe and suitable treatment planning.

**References**
