Nasolabial Cyst in Buccal Mucosa

Abstract
Nasolabial cysts are rare non-odontogenic, soft-tissue, developmental cyst reported till date in the sublabial area and anterior maxillary region. The cyst is a slowly enlarging asymptomatic swelling and non-painful. The cyst is believed to be associated with remnants of the nasolacrimal duct. In this report, we report a nasolabial cyst of a 48-year-old man in whom the cyst occurred in the buccal mucosa. To the best of our knowledge, this is the first case of nasolabial cyst occurring entirely in the buccal mucosa without involving vestibule. The etiopathogenesis of the cyst is reviewed in light of this case.

Keywords: Cheek, nasolabial cyst, nasolabial duct, soft-tissue cyst, swelling

Background
The Nasolabial cyst (NLC) or the Klestadt cyst/nasal vestibule cyst/nasal wing cyst/mucoid cyst of the nose clinically presents as a benign, slow-growing, often unilateral, painless, locally expansile entity, that often presents as a lateral soft tissue cystic lesion below the nasal ala and the nasolabial fold.[1‑4] It is often listed as a jaw cyst, though it is an exclusive soft-tissue cyst. It is an uncommon entity, first reported by Emil Zuckerkandl more than a century ago, makes only 0.6% of all jaw cysts, presenting more commonly in the 3rd to 5th decades of life, with a striking female preponderance, being 3-4 times more common in females, notably of East Asian descent. The other presenting complaint would directly correlate with the extent of involvement of the nasal cavity. Obliteration of the nasolabial fold and lifting of the nasal fold are common observations. NLCs are underneath the ala nasi muscles, associated with the distal portion of the nasolacrimal duct and the inferior nasal meatus. In systematic analysis, NLCs were reported to be localized most commonly (in descending order) to nasolabial, alar, lip, maxillary sinus and anterior nasal meatus. In clinical cases, NLCs were reported to be localized most commonly (in descending order) to nasolabial, alar, lip, maxillary sinus and anterior nasal meatus.

There is unanimous opinion on the etiopathogenesis based on NLCs morphologic, embryologic and clinicopathologic observations.[2-4] The NLCs are associated with being reflective of cystic changes in the solid cord remnants of cells that form nasolacrimal duct (NLD), while others hold it to being a hamartomatous developmental lesions associated with the lower, anterior region of NLD. The other theories indicate that NLCs are fissural cysts, which arise from trapped epithelium along the lines of the fusion of the maxillary, medial nasal and lateral nasal processes. This epithelium embryologically could be from remnants of the nasolacrimal ridge, rod, duct or endodermal cells. There have been similar immunohistochemical protein expression patterns between NLD and NLC.[2] Aikawa et al. proposed that chronic inflammation and scarring could lead to NLCs.[8] This case report aims to present a case of NLC in the hitherto unreported intraoral site.

Case Report
A 48-year-old man reported with a complaint of swelling in his right cheek for more than the past 1 year. The history revealed that it was insidious, non-tender and slow-growing over this period. Currently, it was interfering with his chewing and hence sought treatment. The past medical and dental histories were unremarkable.
On external examination, no abnormalities or asymmetry was noted [Figure 1a]. The lymph node examinations were normal. On intraoral examination, a well-defined spherical swelling of about 3 cm in diameter was noted along the right modiolus in the cheek along the line of occlusion, extending from right commissure of mouth till opposite to the premolars. The nasolabial folds or the buccal sulcus were not involved [Figure 1b]. The swelling was not fixed, appear to be in a submucosal plane, and was firm as well as non-tender on palpation. Orthopantomogram was not contributory. Blood and serum biochemistry were within normal limits.

Differential diagnosis of the condition included developmental cystic lesions (epidermoid or dermoid cysts), salivary glandular lesions (mucocele, pleomorphic adenoma, basal cell adenoma, canalicular adenoma and mucocoeplidermoid carcinoma), mesenchymal (lipoma, fibro lipoma, fibrosarcoma and liposarcoma) neural (neurilemmoma, solitary nerve sheath tumours and neurofibroma) or odontogenic tumours (keratocystic odontogenic tumour of the buccal mucosa and peripheral ameloblastoma) or, less probably, oral manifestation of infectious diseases such as cysticercosis or dirofilariasis.[9]

A working diagnosis of pleomorphic adenoma followed by an epidermoid cyst was made. The lesion was excised via an intraoral buccal incision under local anaesthesia following standard protocols. The cyst was dissected free and the area was closed in layers. Appropriate antibiotic and non-steroidal, anti-inflammatory agents were prescribed along with post-operative care. The healing was uneventful.

The post-operative specimen was a spherical, pink lesion, soft to rubbery-firm soft-tissue mass with a smooth surface [Figure 1c] It had variably cystic and fibrous areas filled by clear viscous fluid. Histopathological examination revealed that the lesion had a two-layered epithelial cystic lining with a cuboidal basal layer and a columnar luminal layer that contains a few mucinous goblet cells. At certain foci, pseudostratified and cuboidal epithelium were identified. The slides also revealed numerous muscle fibres of different orientation and also large blood vessels [Figure 2a–e]. A final diagnosis of NLC was made. The patient was followed up for six months with no recurrence and is under follow-up [Figure 3a and b].

**Discussion**

The present case presentation, barring the site, in its clinical and histological presentation was pathognomonic of the NLC. The presentation was consistent with the NLC as per previous reports.[4] The surgical management was in line with the prescribed protocol of excision in toto, ensuring no spillage.[2]

In the present case, though the list of clinical differential diagnosis was long, the histopathological diagnosis was relatively straightforward and pathognomonic of NLC.[9] The challenge was the site of occurrence. Previously, NLC has been described along the maxillary buccal-labial vestibular regions such as the nasolabial, alar, lip, maxillary sinus and anterior gingival region that lies in the course of the NLD-inferior nasal meatus anatomical region.[1-4] The hamartomas of the buccal mucosa are not unknown. Previous entities that are common to tooth-bearing areas such as keratocystic odontogenic tumour and adenoid cystic carcinoma have been reported in the buccal mucosa.[9-11] The present case is probably the first case to be outside this region- to exclusively occur in the buccal mucosa.
This off-site occurrence could be attempted to be explained on the basis of embryology. Embryologically, cheeks are formed around the 10th week of intrauterine life. The developing maxillary prominences continue growing till they merge laterally with the mandibular prominences to form the cheeks. This causes compression on the medial nasal prominences leading to fusion. Around the same time, the lateral nasal prominence forms the ala of the nose. It also fuses with the maxillary prominence, forming the NLD. This maxillary – mandibular prominences – nasal prominences fusion leads to the development of cheek and NLD. Pretreatment certain elements of NLD material be retained in the extreme edges of fusion, which during tissue remodelling during growth was pushed nearer to the modiolus. From these remnants, the NLC could have originated in the 5th decade of life. This occurrence appears to extremely rare as no case of NLC has been previously reported in other parts of the oral cavity except the nasolabial and associated region. The inflammation origin of NLC has been suggested, based on NLC case reports. As modiolus is also a more trauma prone area of the mouth, the role of trauma in the aetiology of NLC in the present case cannot be ruled out.

Conclusion

A case of nasolabial cyst entirely in the buccal mucosa is presented. This case also underlines the need to revisit such pathological entities to unravel the mystery of orofacial embryogenesis. This knowledge would help us to devise better management strategies for conditions such as orofacial clef.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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