Bilateral nasolabial cysts - case report and review of literature

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Abstract

Nasolabial cyst is a non-odontogenic, extraosseous, soft tissue cyst, commonly unilateral, located in the nasolabial fold. Bilateral nasolabial cysts are of rare occurrence. This case report describes the multimodality imaging appearance of bilateral nasolabial cysts with a review of literature.

Key words: Computed tomography; magnetic resonance imaging; nasolabial cyst; non-odontogenic; ultrasonography

Introduction

Nasolabial cysts are usually unilateral and have a strong female predilection. Bilaterality is reported in only two case reports, to our knowledge. This case is unique in that it is a male patient and is bilateral. We also describe the ultrasonography (USG), computed tomography (CT), and magnetic resonance imaging (MRI) features with surgical and histopathological confirmation.

Case Report

A 32-year-old male patient was referred to the radiology department for CT examination of the face with symptoms of swelling in the left side of nose for 7-8 months. There was an insidious increase in the size of the swelling over months. No pain, fever, or other constitutional symptoms were elicited. On clinical examination, the swelling was soft to firm, relatively mobile, and was seen in the groove between the cheek and the left nasal ala.

Non-contrast CT examination of the face revealed two well-defined hyperdense lesions on either side of the nasal

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ala at the nasolabial fold, obliterating the fold [Figure 1A]. The lesions measured 15 \times 11 mm and 37 \times 30 mm on the right and left sides, respectively, with an average attenuation value ranging from 33 to 52 HU. No calcification or fat density was seen. There was smooth scalloping of the anterior maxillary bone on either side of the anterior nasal spine [Figure 1B]. USG showed the lesions to be entirely cystic with dependent and mobile debris within [Figure 2]. No intramural nodule was seen. No significant vascularity was noted.

On MRI, both the lesions showed similar signal characteristics, appearing hyperintense on T1-weighted sequences with hyperintense dependent areas [Figure 3A]. On T2-weighted images, the lesions returned hyperintense signal with the dependent areas appearing hypointense [Figure 3B and C].

A final diagnosis of bilateral nasolabial cysts with debris representing proteinaceous secretions or hemorrhage within was considered.

Due to cosmetic reasons, sublabial surgical approach was used and complete enucleation of bilateral nasolabial

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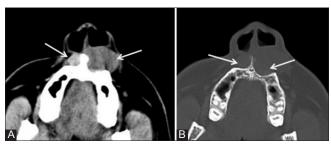


Figure 1 (A and B): (A) Plain axial CT shows two welldefined relatively iso-hyperdense lesions on both sides of nasal ala (arrows) (B) Axial CT at same section in bone window shows smooth scalloping (arrows) of the bilateral maxillary bones (left more than right)



Figure 3 (A-C): (A) Axial T1 weighted image shows two well defined cystic lesions with hyperintense levels (arrows) (B) Axial T2 weighted image at same level shows the leisons to be hyperintense similar to CSF signal with hypointense levels (arrow) (C) Coronal T2 weighted image shows bilateral nasolabial cysts

cysts was performed carefully removing the entire wall [Figure 4A-D].

The cut open cysts showed turbid contents appearing brownish in color with dark brown sediments within. Histopathology showed the cyst wall to be multi-layered and composed of pseudo-stratified columnar epithelium with goblet cells [Figure 5A and B].

Discussion

Nasolabial cyst, also called as the nasoalveolar cyst or Klestadt's cyst, is a relatively uncommon benign, non-odontogenic, extraosseous maxillary cyst. It is also called by other names such as nasal vestibular cyst, mucoid cyst of nose, nasal wing cyst, and subalar cyst. The cyst is classically related to the nasolabial fold or nasal alar soft tissue. Two popular theories that address its origin include the possible development from the nasolacrimal duct embryologic remnants and from the trapped epithelium

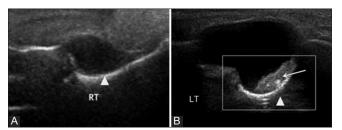


Figure 2 (A and B): (A and B) USG shows the leisons to be anechoiec with dependent echogenic debris (arrow). Note smooth scalloping of the bone (arrowhead)

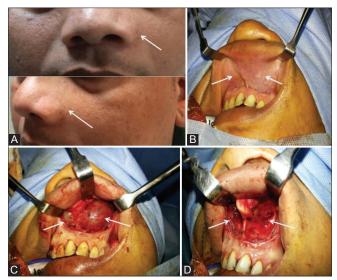


Figure 4 (A-C): (A) Clinical photographs of the patient showing smooth obliteration of the left nasolabial fold (arrows) (B) Per operative image showing sublabial bulge corresponding to the cysts (arrows) (C) After incision of the upper gingivobuccal sulcus, the cyst walls are exposed (arrows) (D) Post complete removal of the cysts

at the fusion of maxillary, medial nasal, and lateral nasal processes. The former theory is widely accepted due to the reason that the lining of the cyst is a pseudo-stratified epithelium similar to the lining of the nasolacrimal duct.

Though these cysts are referred to as developmental cysts, the mean age of occurrence is in the adult group, averaging 30-45 years, with female preponderance.^[1] The incidence of bilaterality is 10-11%.^[2,3]

Clinically, patients present with an asymptomatic slow-growing soft tissue swelling in the lateral aspect of the nasal ala, typically obliterating the nasolabial fold. The swelling is usually painless unless there is secondary hemorrhage or infection. Spontaneous rupture of the cyst into the nasal cavity resulting in transient discharge and complete resolution can occur. In some cases, recurrence is noted.

The Austro-Hungarian anatomist Emil Zuckerkandl reported the first case of nasolabial cyst.^[4] Klestadt did a detail study of these cysts and proposed the possible origin

of the same, and hence the name "Klestadt's cyst" was given in his honor.

Grossly, when resected in toto, the nasolabial cysts are soft to firm soft tissue mass with a smooth surface. Contents are variably cystic with clear fluid, hemorrhage, or purulent material if infected. The epithelium is bilayered or multilayered. The commonest type of epithelium encountered is pseudo-stratified columnar, followed by combined columnar, cuboidal and columnar, and stratified squamous epithelium. Intraepithelial goblet cells are a consistent feature. It is seen in up to 52% cases. [4] The cyst wall stroma is a hypocellular, collagen-rich fibrovascular tissue with or without chronic inflammatory cells.

Since the histology of the nasolabial cyst resembles that of the nasolacrimal duct lining, the nasolacrimal duct origin theory is more favored than the nasal process entrapped epithelium origin theory.

On ultrasound, these lesions are anechoic with or without internal debris representing hemorrhage, secretions, or calcium. On CT, the cyst usually shows hyperdense contents, but can appear hypodense also. One case report demonstrated calcium levels resembling "milk of calcium" as in renal cysts or in gall bladder. Underlying bone can show smooth bone scalloping in most of the cases due to chronic pressure effect. On MRI, they appear slightly hyperintense to CSF on T1-weighted images and brightly hyperintense on T2-weighted images. Hyperintense levels on T1-weighted images can be seen indicating calcium or hemorrhage.

Differentials of a cyst in the region of the nasolabial fold include odontogenic cysts such as dental or dentigerous cysts with cortical perforation. But they typically are related to the tooth and are osseous lesions. Fissural cysts such as incisive canal cysts and globulomaxillary cysts are also intraosseous and have typical locations. Incisive cyst or the nasopalatine cyst is an anterior midline maxillary lesion and is located either in the labial or the oral aspect. It is usually round in shape and can show a notch due to

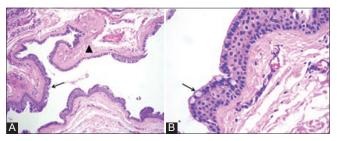


Figure 5 (A and B): (A) Cyst wall is multilayered with pseudostratified columnar epithelium lining (arrow). Note fibrovascular stroma (arrowhead) (B) Magnification image shows intervening goblet cells classical of nasolabial cyst (arrow)

indentation by the nasal septum or superimposition by the nasal spine. Globulomaxillary cysts are located off midline between the lateral incisor and canine teeth in the maxillary bone.

Dermoids are usually midline or located in the medial canthal region. They present usually in childhood. Location lateral to the nasal ala is unusual. Differentiation from epidermoid cyst can be possible with the color of the cyst, as the former appears yellowish due to the sebaceous content whereas the nasolabial cysts show slight bluish tinge. Other uncommon differentials include nasolacrimal mucocele, which occurs secondary to non-canalization of the nasolacrimal duct. It is commonly seen in infants and they present with epiphora and dacryocystitis. A unilocular lymphangioma may be difficult to differentiate from a nasolabial cyst since the imaging findings may be almost similar. One case report of schwannoma shows similar imaging features of nasolabial cyst with a 3D morphology of the soft tissue used for differentiating.^[7] Figure 6 illustrates typical locations of the commonly encountered perinasal cysts.

Pathological differentials include salivary gland retention cyst and oral heterotopic gastrointestinal cysts, which do not pose problem clinico-radiologically.

Surgery is primarily done for cosmetic reasons and for anticipated secondary complications like infection or hemorrhage. Malignant transformation in a nasolabial cyst, to our knowledge, is not reported in the literature. Open surgical or endoscopic technique is used. Su *et al.* described transnasal endoscopic marsupialization of the cyst. It is less invasive and easier to perform in large lesions.^[8] Open technique involves complete surgical enucleation of the cyst including the cyst wall using sublabial approach

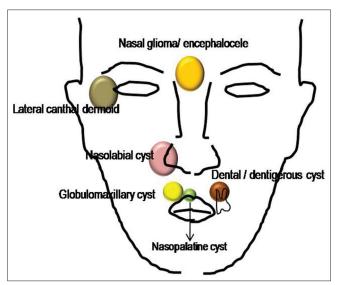


Figure 6: Illustration showing typical locations of various perinasal cysts

to avoid possible recurrences. Other treatment options including sclerotherapy or incision drainage usually lead to local recurrence and are performed in surgically unfit individuals.

Conclusion

In conclusion, nasolabial cysts are rarely encountered non-odontogenic extraosseous cysts typically located in the nasolabial fold. Unilaterality and female preponderance are commonly seen. Familiarity with the multimodality imaging appearance will allow confident diagnosis of this condition.

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Conflicts of interest

There are no conflicts of interest.

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