Nasopalatine Duct Cyst: A Common Lesion with Uncommon Presentation in a Young Girl

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ABSTRACT
Nasopaline cysts are the nonodontogenic developmental cysts. These are most frequently occurring asymptomatic cysts in the oral cavity and are commonly observed as a swelling in the anterior maxillary region. Nasopalatine duct cyst presents in the fourth to sixth decades of life with a male preponderance. The main aim of this case study is to review the prevalence, epidemiology, and clinical presentation, to describe the radiographic and pathological findings, and to discuss surgical management of this entity in an 11-year-old female patient.

Keywords: Asymptomatic, Nasopalatine, Nonodontogenic.

CASE REPORT

INTRODUCTION
The nasopalatine duct passes through the incisive canal (nasopalatine canal) which is a passage that connects the palate to the floor of the nasal cavity. During fetal development, the ducts narrow until one or two clefts are formed on the midline of the anterior maxilla. Generally, there is a degeneration of the epithelial lining of the canal. But sometimes the embryogenic remnants persist and proliferate and give rise to a cyst. According to the WHO classification of cysts 2019, nasopalatine duct cyst is a developmental, epithelial, nonodontogenic epithelial cyst of the maxilla along with nasolabial cysts. It constitutes <1% to 33% (average 12%) of all the cysts of the jaw. Its frequency is very high being 1% of the total population. Although they can occur at any age, the most commonly affected age-groups are 40 to 60 years with an average being 42.5 years. It shows a significantly higher frequency in males than females. It is usually an asymptomatic lesion, but on many occasions, the cases present with swelling and pain. The vitality of the teeth is not usually affected, but the cyst could be misdiagnosed as a periapical cyst or a granuloma. After surgical treatment, recurrence is uncommon, having been reported in 0 to 11% of patients.

CASE HISTORY
An 11-year-old female reported to the hospital with the complaint of painless swelling of the face and the palate. The swelling appeared one and half months ago. Facial asymmetry was seen with swelling on the right side of the face. There was no history of trauma. She had no significant past medical history and her laboratory records were under normal limits.

Extraoral examination findings showed swelling on the right upper lip extending to the buccal area. The swelling was tender on palpation. Facial deformity was observed (Fig. 1).

On intraoral examination, the palatal swelling was well-defined, 4 x 4 cm at the center extending to the gingivobuccal sulcus. The right maxillary central incisor was mesially displaced. The maxillary incisors showed grade 1 mobility and their vitality was preserved (Fig. 2).

A provisional clinical diagnosis of benign cystic lesions was made. Differential diagnoses of nasopalatine cyst, nasolabial cyst, benign cystic lesion of minor salivary gland origin, central giant cell granuloma, and root cyst were considered.

On computed tomography (CT) scan record showed a lytic expansile mass of 3.5 x 4.2 x 4 cm involving the hard palate, the midline of the upper alveolar ridge with cortical thinning, and dehiscence at the upper alveolar ridge. There was no evidence of unerupted teeth. The mass was protruding in the oral cavity and the bilateral nasal cavities. The remainder of the maxillofacial bones appeared intact. Paranasal sinuses appeared clear to the extent visualized. There was deviation of the nasal septum to the right side (Fig. 3). Fine needle aspiration cytology carried through intraoral approach yielded hemorrhagic cyst fluid. No epithelial or malignant cells were observed.

Taking into consideration the aspiration cytology and radiological findings, a diagnosis of benign cystic lesion was considered, a surgical enucleation was planned intraoperatively, and about 30 cc cystic fluid was aspirated. Then under general anesthesia, a claviculcer incision from the left canine to the right first molar was given. The mucoperiosteal flap was reflected and the cystic cavity was accessed. The lining of the cystic cavity was removed totally. Hemostasis was achieved and chemical
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Discussion

The nasopalatine duct cyst was first discovered by Meyer in 1914.\(^9\) Being one of the most common developmental nonodontogenic cysts, it originates from the remains of the nasopalatine duct epithelium.\(^1,2,9\) The nasopalatine duct, in mammals, is a transmission pathway for pheromones and chemosignals entering the oral cavity to the vomeronasal organ called the Jacobson’s organ.\(^2,10\) The stimulus for cyst formation from the epithelial remnants of the nasopalatine canal is uncertain, although trauma and bacterial infection are thought to play a role.\(^1,11\) It usually presents as an asymptomatic lesion, but some cases show symptoms like mild

cauterization was done with Carnoy’s solution. The closure was with 3.0 silk. A retentive palatal plate was created by taking an impression prior to the surgery for coverage of the exposed palatal bone (Fig. 4). Histopathological examination revealed cyst lined by either stratified squamous epithelium or pseudostratified columnar epithelium which was ulcerated at places. The fibrocollagenous cyst wall showed leukocytic infiltration. With these findings, diagnosis of nasopalatine cyst was established (Fig. 5). Postoperatively the healing was satisfactory. Retentive palatal plate was used to achieve hemostasis and to eliminate the dead space after surgery to prevent refilling of the cavity after enucleation (Fig. 6).

Figs 1A and B: Extraoral swelling in the right maxillary region

Fig. 2: Intraoral swelling seen at the palatal region

Figs 3A to D: The CT scan report shows an expansile cyst involving the hard palate and the nasal cavities. (A) Frontal view, (B) Lateral view, (C), and (D) Axial sections of the palatal area
swelling with well-defined border. It makes up 10% of the total jaw cysts. In spite of being a developmental cyst, it is rarely seen in the first decade of life.\textsuperscript{11}

We are presenting a case of a unilateral nasopalatine duct cyst in a 11-year-old female which is uncommon. The literature shows that this cyst is more commonly seen in men than women with a ratio being 2.5:1 and is most commonly observed in the fourth and sixth decades of life.\textsuperscript{1,2,11} The literature shows that it can be present bilaterally (60%), unilaterally (20%), or centrally (20%).\textsuperscript{12} A study of 334 nasopalatine duct cysts shows the mean age to be 42.5 with the male to female ratio being 1.2:1.\textsuperscript{6} Vas concelos et al. found the mean age to be 37.4 years and the male to female ratio being 3.4:1.\textsuperscript{5} A more recent study of 57 published cases has found the mean age to be $34.1 \pm 17.6$ years and the male to female ratio being 2:1.\textsuperscript{13}

Pain is not a frequent complaint but it can be due to pressure on the nasopalatine nerves or if the cyst becomes secondarily infected.\textsuperscript{11} Our case gives us a history of painless palatal swelling extending to the gingivobuccal sulcus.

Drainage occurs in 25% of the cases and may be mucoid, in which case the patients describe a salty taste, or it may be purulent and the patients may complain of a foul taste. Some reports show fluid aspirated to be straw colored with a mixture of blood.\textsuperscript{2,11} Histologically, the nasopalatine duct cyst is lined by stratified squamous epithelium alone or in combination with pseudostratified columnar epithelium, simple columnar epithelium, and simple cuboidal epithelium.\textsuperscript{4,11} In our case, on aspiration of the cyst, deep red clear (hemorrhagic) fluid was obtained which on pathological examination gave the diagnosis of nasopalatine duct cyst. The literature shows that the cyst usually has a diameter of 1.5 to 2 cm.\textsuperscript{2,13} Very few cases are reported to be more than 3 cm in dimension.\textsuperscript{4} In our case, the cyst was large measuring $3.5 \times 4.2 \times 4$ cm in the largest dimension. Radiographic appearance of the cyst is reported to have round, ovoid, or heart-shaped radiolucency due to the superimposition of the nasal spine.\textsuperscript{3,6,14} Her CT scan reported a
3.5 × 4.2 × 4 cm expansile involving the palatal process of maxilla and the nasal bone which helped confirm the existence of the nasopalatine cyst. These cysts usually involve the hard palate and may have nasal involvement. Nasopalatine cyst is located in the midline with sclerotic margins that show smooth expansion and displacement of teeth apices while radicular cysts have teeth apices within the cyst rather than being displaced. CT scan imaging aids in differentiating nasopalatine cysts from radicular cysts.

Ficarra et al. proposed complete enucleation and fixation of the surrounding bone by Carnoy’s solution as a treatment modality for nasopalatine cyst. Surgical enucleation was planned for the removal of the cyst. Complete cystic lining removal was performed under general anesthesia and Carnoy’s solution was used for chemical cauterization. Carnoy’s solution is a fixative composed of 60% ethanol, 30% chloroform, 10% glacial acetic acid, and 1 gram of ferric chloride. She was discharged in the next 5 days with continued Ryles tube feeding.

**Conclusion**

Nasopalatine cysts are the most common odontogenic cysts. These cysts are generally 2 to 3 cm, observed in males in fourth and sixth decades of their life. Our case is unique for three reasons, (1) young age, (2) female sex, and (3) large size. Hence, we conclude that the diagnosis of nasopalatine cyst must be kept in mind even in large palatine swelling in young girls.

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