



Nasopalatine Duct Cyst: A Case Report and Review

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Running title: Non-odontogenic cyst

Clinical significance: The dentist must be alert and not overlook this common non-odontogenic lesion on the hard palate.

ABSTRACT

Nasopalatine duct cyst (NPC) is one of the most usually arising non-odontogenic cysts in the maxillofacial region. The cyst is also known as nasopalatine duct cyst. It is a developmental, epithelial, fissural cyst and is exceptional in that; it arises in only at a single site: in the midline of the anterior maxilla. The cyst is usually asymptomatic and is often identified during the routine radiographic examination.

INTRODUCTION

Nasopalatine duct cyst is also known as the:

- ✓ Median anterior cyst
- ✓ Anterior median palatine cyst
- ✓ Incisor duct cyst
- ✓ Midline maxillary cyst
- ✓ Incisive canal cyst

Meyer first reported the nasopalatine duct cyst (NPC) in 1914. NPC, also named as incisive canal cyst, is a development lesion that arises from embryologic remnants present in the nasopalatine duct. It is one of the most common non-odontogenic cysts of the oral cavity, occurring in about 1% of the population. Predominantly the cases occur in the 4th and 6th decades of life and males are affected more often than females, with a ratio of 3:1. Although cysts may arise at any point along the nasopalatine duct, most of them originate in the upper part (nasal part) of the opening that extends from the nasal spine to an area between the roots of the two maxillary central incisors, and some lesions arise entirely within the soft tissue of the incisive papilla. These are often designated as cysts of the palatine papilla. It may be asymptomatic and discovered on routine radiographic examination or may manifest as a slow growing swelling in the anterior region of the midline of the palate. Pain may occur if the cyst becomes secondarily infected. Radiographically, NPC usually presents as a clear heart shaped radiolucent area. They are symmetrical, but some are displaced to one side. The cyst may be distinguished from a normal but widened NP canal and periapical granuloma or radicular cyst associated with the roots of anterior teeth. A correct diagnosis can only be made after proper clinical, radiographic, and histopathologic examination is conducted. Malignant transformation has rarely been reported. Enucleation is the ideal treatment with low recurrence rates.¹

Etiology: Many etiologic factors have been stated:

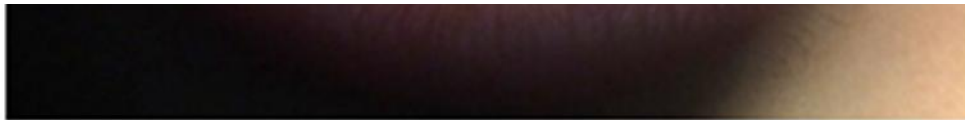
- ✓ Local trauma during mastication or
- ✓ Ill-fitting dentures,
- ✓ Bacterial infection, and
- ✓ Spontaneous proliferation.
- ✓ Blockage of glandular ducts as well as
- ✓ Racial or genetic factors

are other factors mentioned in the literature. During inadvertent surgery, there are chances of bleeding from the nasopalatine vessels which are entrapped within the cystic cavity. The nasopalatine neurovascular bundle is a highly vascular and delicate structure.²

Case Report



Figure 1: Swelling 2x2 cm in size between the roots of the 11 and 21.



of a swelling in the upper front teeth region for about a year. Swelling was reported to be small initially and then gradually increased to its present size. There were no significant signs and symptoms associated with the lesion. On clinical examination, all the subjective signs and symptoms were within normal parameters. Intraoral examination revealed a 2 x 2 cm sized swelling in the anterior teeth region, soft in consistency, and its overlying mucosa was normal. (**Figure1**). Radiographic examination revealed a diffuse radiolucency between the apex of 11 and 21-tooth region measuring around 1.5 x 2 cm in size (**Figure 2**).

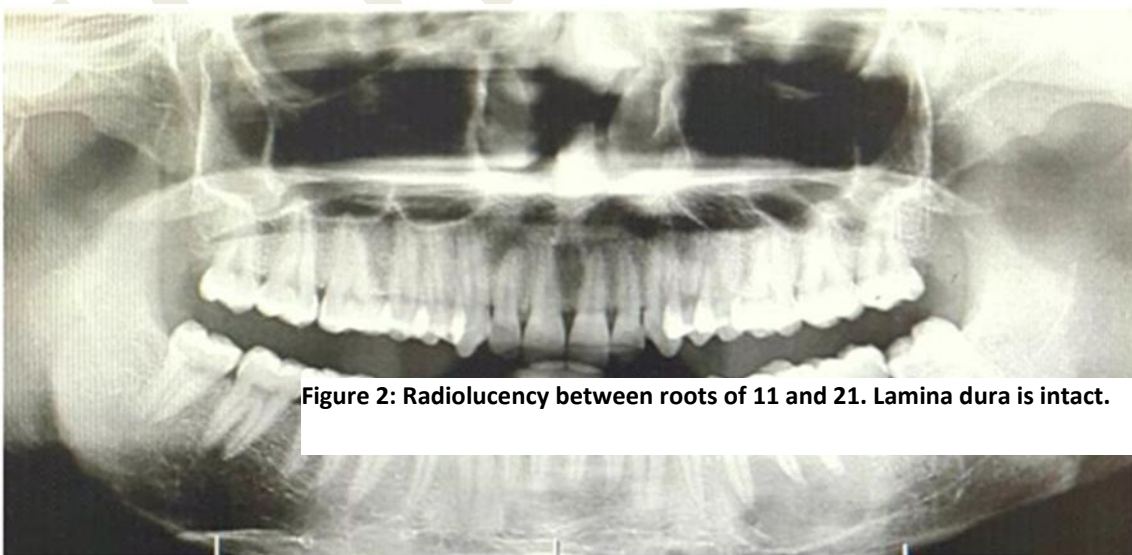


Figure 2: Radiolucency between roots of 11 and 21. Lamina dura is intact.

After observing the clinical and radiographical findings, a provisional diagnosis of nasolabial cyst/ nasopalatine cyst was made. Following strict aseptic precautions and under local anesthesia a flap was raised, and the enucleation of the cyst was done. On microscopic

A young female patient, about eighteen years of age, reported to a private clinic in Bangalore Urban, Karnataka, INDIA with a chief complaint

examination, the hematoxylin and eosin stained soft tissue section, showed a fibrovascular capsular wall surrounding a cystic lining. **(Figure 4)** Surgical specimen was sent for histopathological investigation. **(Figure 3)**



Figure 3: Enucleation site of the lesion seen in relation to 11, 21 region

Figure 4: H and E stained section shows the cystic lining made up of cuboidal cells.

FIGURE 4

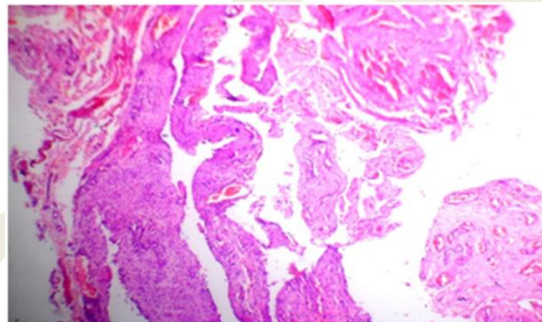
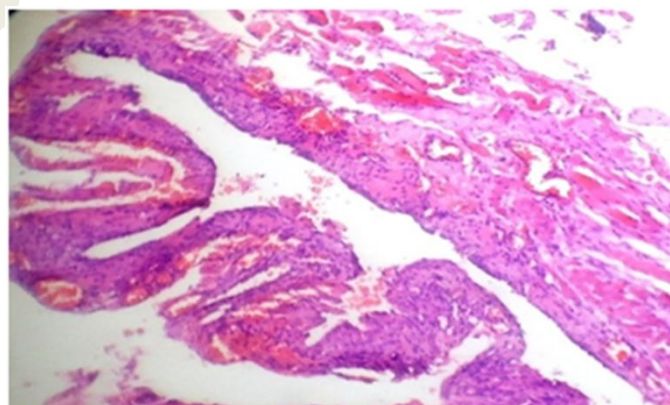


Figure 5: H and E stained section shows the epithelial lining enclosing a lumen and a fibrovascular capsular wall.

FIGURE 5

On high power magnification, the showed a single pseudostratified epithelium, which exhibited cilia in



cyst lining layer of columnar also some areas.

(Figure5) Observation of the clinical, radiographical and histopathological findings helped in

arriving at the final diagnosis of the nasolabial cyst was made. The patient was kept under observation with periodic follow-up. The healing was uneventful.

Discussion

The nasopalatine cyst is an embryonic, nonodontogenic, cyst that has a controversial origin, which is explained by various theories of pathogenesis:

Incidence: The cyst predominantly occurs in about 1% of the population. Moreover, it comprises of 1.7-11.9% of all jaw cysts.³

Etiopathogenesis: Two theories of origin:

- The cyst is said to arise from the degeneration of nasopalatine ducts; ⁴a which is the connection between the anterior maxilla and the nasal cavity in the developing fetus, ⁵which normally regresses in fetal life. ⁶Continuation of the fetal development, causes the connection between the two structures to narrow down as the bones of the anterior palate fuse. This results in the formation of the nasopalatine canals that carry the nerves and vessels, and the epithelial rests from the degenerated nasopalatine ducts. In case they persist, they degenerate. ⁵
- The second theory, though out of favor, states that the cyst arises from the epithelial remnant trappings during the embryologic fusion between nasal cavity and anterior maxilla.³

Etiology: Factors like local trauma during mastication, bacterial infection, ill-fitting dentures, and spontaneous proliferation have been suggested as etiological factors for the NPC. Racial or genetic factors, as well as the blockage of glandular ducts, have been proposed as other possible causes that have been mentioned in the literature.

Clinical Features:

Age: 4th to 6th decades of life.⁴

Sex: Males more commonly affected than females.⁴

Site: Always seen between the two maxillary central incisors. ⁴Rarely, the cyst may develop within incisive papilla, and in that instance, it is termed as the cyst of incisive papilla, or cyst of palatine papilla

Presentation:

Signs: Shows as swelling in the anterior palatine region. Discharge of pus from the swelling may be seen which may have a salty taste.

Symptoms: Early symptoms may occur in the caudal portion of the cyst. Growth is very rarely seen extraorally even if the cyst is inflamed (46% of cases) because the growth or expansion is intraorally (palatine). In advanced cases, the cyst may manifest with pain and itching. Pain due to compression of adjacent structures has been reported in 17% of cases when it becomes infected. ²In occasional cases, the patients have complained of a burning sensation that may radiate into the orbit and the bridge of the nose.²

The lesions are mostly asymptomatic, and the lesion could be detected on routine radiographs.⁵ The vitality of teeth is normal, but occasionally the tooth would have undergone an apicectomy because of a previously misdiagnosed periapical cyst or granuloma.²

Other symptoms include:

- ✓ A salty taste in the mouth.
- ✓ The swelling may be bluish in color depending on the proximity of the fluid to the surface.
- ✓ Numbness.
- ✓ Central incisor root divergence
- ✓ Central incisor crown overlapping

Radiographic features: The cyst presents as a solitary ovoid or round, well-defined unilocular radiolucency, between the maxillary central incisors both on the occlusal radiograph and on the Computer tomography scan. In cases, where superimposed by the nasal spine it may appear “heart-shaped.”⁴ The cysts are located apical to the roots of the maxillary incisors, but they rarely cause root resorption. Some cases may show root resorption and tooth displacement.⁴ Cysts range in size, with an average diameter of approximately 1.5 centimeters. The incisive foramen is smaller than 6 mm in diameter, therefore, making the identification of a small nasopalatine duct cyst difficult.⁵

Magnetic Resonance Imaging

There is usually homogeneous high-signal intensity on both on T1 and T2, wherein T2 it is high intensity and intermediate on T1 wherever there are contents (keratin and viscous fluids); though most cysts of maxillofacial regions exhibit intermediate signal intensity on T1 weighted images.⁴ MRI is more specific than CT.⁷

Histopathological features: The nasopalatine duct cyst is lined by stratified squamous epithelium alone or in combination with a pseudostratified columnar epithelium (with or without cilia and goblet cells), simple cuboidal epithelium, and simple columnar epithelium.⁵ Approximately, 30% cases contain respiratory epithelium.⁴

The fibrous wall of the cyst is made up of nerves, arteries, and veins. Minor salivary gland tissue and small islands of cartilage may also be found. An infected cyst would show acute and chronic inflammatory cells throughout the specimen.⁵

Differential diagnosis

- Schwannoma in the incisive canal region
- An enlarged incisive fossa
 - The incisive foramen usually is not expected to exceed 6 millimeters.
 - A radiolucency in the maxillary anterior region with ill-defined borders is regarded as a large incisive fossa
 - It can be distinguished from a nasopalatine duct cyst, clinically by aspiration.³
- Central giant cell granuloma
- Ameloblastoma
- Keratocystic Odontogenic Tumor
- Glandular odontogenic cyst:

- It is an intraosseous developmental odontogenic cyst that may show mucous or ciliated cells in cystic lining
- Should NOT contain the contents of incisive foramen (peripheral nerve, cartilaginous rests, muscular, vascular channels)
- Nasolabial (Naso-alveolar) cyst:
 - It is a soft tissue (non-intraosseous) cyst with similar histologic features as nasopalatine cyst
 - Occurs in soft tissues of upper lip lateral to midline
 - shows this lesion
 - Contents of incisive foramen such as the peripheral nerve, cartilaginous rests, muscular, vascular channels must be absent
 -
- Periapical (radicular) cyst:
 - It is one of the most common inflammatory odontogenic cysts
 - Stratified squamous epithelium of variable thickness, often with scattered ciliated cells
 - lines the cyst
 - Rests of Malassez
 - gives rise to the cyst
 - The lamina dura shows loss of continuity of the lamina dura and the pulp is usually non-vital
 -
- Surgical ciliated cyst:
 - It normally arises as a post-operative "complication" with cystic expansion of respiratory epithelium within maxilla, which may have mucus or ciliated cells within cystic lining
 - Usually located in posterior maxilla, and lacks contents of incisive foramen (peripheral nerve, cartilaginous rests, muscular, vascular channels).³

Treatment and prognosis

Nasopalatine duct cyst is completely enucleated, generally by palatal approach⁵ and it is usually curative.¹ Mostly surgery is an adequate treatment. Recurrence is rare.⁵ Histological confirmation is recommended.¹ Marsupialisation may be sufficient in the case of small lesions but in cases of large lesions marsupialization, followed by cystectomy and autogenous bone grafting would be the ideal treatment modality.⁸

Prognosis:

- A malignant transformation of this cyst has been reported though it is extremely rare,
- There may be a relapse rate which may vary but usually, ranges from 0-11%
- Higher relapse rate is frequently associated with hyperkeratosis, histologically (close to 30%).²

CONCLUSION

Nasopalatine duct cysts are the commonest non-odontogenic cysts of the oral cavity seen in the entire population. It must be differentiated from other radiolucencies of the maxillary anterior region. To rule out the presence of a radicular cyst, the vitality testing of teeth adjacent to or of the teeth involved with a cyst-like lesion is obligatory, and the final diagnosis can be given only after histological analysis has been made. Hence it is mandatory that practitioners are aware of this lesion arising on the palate.

Footnotes:

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A patient consent form is available with the editors.

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