Diagnosis and Management of Nasopalatine Duct Cyst: A Report of Two Cases and Literature Review

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Diagnosis and Management of Nasopalatine Duct Cyst

A Report of Two Cases and Literature Review

Feiyi Sun, D.D.S.; Harry Dym, D.D.S.

ABSTRACT

The nasopalatine duct cyst (NPDC) is one of the most common non-odontogenic, non-neoplastic developmental cysts. It arises from the epithelial remnants of the nasopalatine duct within the incisive canal and crosses the midline of the anterior maxilla. Most cases are asymptomatic and are discovered either by chance on radiographs or present as a soft-tissue swelling, with pain or drainage in the palate. The present study describes the diagnosis and surgical management of two NPDC cases seen at the Department of Oral and Maxillofacial Surgery at the Brooklyn Hospital Center.

The incisive canal is a funnel-shaped, narrow, bony structure that connects the nasal and oral cavities. The canal starts superiorly at each side of the nasal septum, extends inferiorly and ends palatally to the two maxillary central incisors underneath the incisive papilla. The incisive canal commonly takes form as either a “Y” or “V” shape, given its bilateral termination superiority at the base of the nasal septum, known as the foramina of Stenson, and its single endpoint at the incisive foramen. However, anatomic variances of the incisive canal have been reported, as the canal can exist as one single canal, two parallel canals or a “Y”-shaped canal with two or more nasal openings. The dimension of the canal is generally greater in males, and the length of the canal decreases with increasing age. The mean value of the length of the canal is approximately 11 mm.

Neurovascular structures traveling through the incisive canal include the nasopalatine nerve, which provides the sensory input to the mucosa of the hard palate from the maxillary central incisors up to the maxillary canines bilaterally. This neurovascular bundle also gives vascular supplies to the same area via the anastomosis between the posterior septal branch of the sphenopalatine artery and the greater palatine artery.

The traditional view on the formation of the incisive canal during embryogenesis is based upon a popular theory that the canal is the central point of fusion between the primary and secondary palates. It represents a rare form of a cleft palate. However, Radianski et al. reported that the incisive canal develops solely from the primary palate within the premaxilla, and the associated neurovascular structures are derived from the mesenchymal tissue.

One should not confuse the incisive canal with the nasopalatine duct, as the duct is one of the major components inside the incisive canal aside from the neurovascular bundle. In most mammals, the nasopalatine duct serves to facilitate the passage of pheromones between the oral and nasal cavities. The duct terminates near the vomeronasal organ in the nasal cavity to transmit...
neural signal to the central nervous system. In humans, the nasopalatine duct can be blocked following birth. The duct, however, can remain patent at times to maintain communication between the oral and nasal cavities. The vestigial form of the nasopalatine duct can lead to the development of a NPDC in the anterior maxilla.[7,8,9]

NPDC was first described by Meyer in 1914 and was believed to arise from epithelial remnants of the nasopalatine duct. The lesion shares other names, such as anterior midline cyst, maxillary midline cyst, anterior middle palatine cyst and incisive duct cyst. According to the classification of the World Health Organization (WHO), NPDC is regarded as a developmental, epithelial, non-neoplastic and non-odontogenic cyst occurring in about 1% of the population.[10] It is one of the most common non-odontogenic cysts that occur in the anterior maxilla.[7,8,9]

This study reports two cases of NPDC that were diagnosed and treated at the Department of Oral and Maxillofacial Surgery at the Brooklyn Hospital Center.

**Case Reports**

**Patient One**

The patient, a 34-year-old Hispanic female with no significant past medical history, presented to the dental clinic for a routine checkup. Clinically, the patient was asymptomatic, with vital anterior maxillary teeth. There was no facial or palatal vestibule swelling or depression at the anterior maxilla. Radiographically, it may appear as a well-demarcated, heart-shaped, unilocular radiolucency apical to the roots of maxillary central and lateral incisors.

Clinically, patients are often asymptomatic but may sometimes present with a soft-tissue elevation at the anterior palate.[11] It is usually discovered during a routine dental examination and can be confused with periapical lesions of dental origin. Patients presenting with NPDC can be incorrectly treated with endodontic therapy of the maxillary anterior teeth. Misdiagnosis of NPDC is not uncommon due to its anatomic location near the apices of the maxillary anterior teeth. Therefore, it is important to recognize NPDC and avoid giving the wrong treatment to patients who may have healthy maxillary anterior teeth.

The patient was taken to the OR for enucleation and curet-tage of the cyst at the anterior maxilla. Access to the cystic lesion was gained from the palatal side due to the palatal perforation. A surgical handpiece was used to uncover the cyst to facilitate the removal of the cystic lining. A curette was used to completely enucleate the cyst. One cc of corticocancellous bone was placed into the cystic cavity and covered with platelet-rich fibrin membranes, which were fabricated using the patient’s own blood. A
primary closure was achieved at the surgical site using dissolvable 3-0 vicryl sutures.

On the follow-up appointment one week after the procedure, the patient denied any sensory changes, swelling or bleeding. The maxillary anterior teeth remained vital.

**Patient Two**
The patient is a 37-year-old Caucasian male with no significant past medical history referred by his previous general dentist for evaluation of a cyst at the anterior maxilla and possible root canal therapy of teeth #8 and #9. He reported a fall and trauma to the anterior maxilla around two years ago. The patient was clinically asymptomatic, with vital teeth #8 and #9, as well as the rest of the anterior maxillary teeth. There was no facial or palatal soft-tissue swelling or depression.

On the panoramic X-ray, it was difficult to appreciate a well-defined, radiolucent, unilocular lesion at the anterior maxilla. A CT maxillofacial without contrast was ordered to further evaluate the extent of the cyst, and a well-circumscribed ovoid radiolucent lesion within the anterior maxilla with a thin sclerotic border measuring approximately 1.6 cm x 1.4 cm x 1.3 cm was appreciated (Figure 3).

The patient was subsequently taken to the OR for enucleation and curettage of the cyst at the anterior maxilla. The access to the cystic cavity was made from the facial side due to the thinning of the buccal cortex (Figure 4). After careful removal of the cystic lining and a thorough curettage inside the cystic cavity, 1 cc of corticocancellous bone was placed, along with PRF membranes, on top. The surgical site was closed primarily using dissolvable 3-0 vicryl sutures.

The patient did not report any postoperative complaints on the follow-up appointment. However, teeth #8 and #9 were found non-vital following the surgery. A root canal therapy of the two teeth was recommended.

**Histopathological Examination**
Microscopic examination of Patient One revealed a benign cystic lining composed of flatten cuboidal (Figure 5a), whereas that of Patient Two revealed a benign cystic lining composed of squamous epithelium (Figure 5b). Both findings were consistent with nasopalatine duct cyst.

**Discussion**
The NPDC is one of the most common developmental, non-neoplastic and non-odontogenic cysts of the oral cavity, occurring in around 1% of the population. During fetal development, when bones of the anterior maxilla start to fuse and form the incisive canal, the nasopalatine duct, which communicates between the nasal cavity and anterior palate, starts to degenerate as well. In addition to the neurovascular bundle, which includes the nasopalatine nerve and vascular anastomosis from the posterior septal branch of the sphenopalatine artery and the greater palatine artery, the incisive canal also carries the degenerated epithelial remnants of the nasopalatine duct.

The stimulus for cyst formation from the remnants is unclear. Some believe that it may derive from spontaneous proliferation of embryonic tissue remains, while others believe causative factors lie in dentoalveolar trauma, local infection, ill-fitting denture, and genetic or racial predilection. Mucous glands within the epithelial lining have also been speculated for cyst formation due
to mucin secretion.\textsuperscript{[6,10,11]} In our study, Patient Two reported a history of trauma to the anterior maxilla due to a fall.

In an analysis of 334 cases of NPDCs, Swanson et al. reported a mean age of 42.5 years, with no significant difference between the ages of males or females, and blacks or whites at time of diagnosis.\textsuperscript{[12]} Most literature indicated that NPDCs have a slight male predilection while remaining equal in different races. Socioeconomic status can play an important role when reporting cases of NPDCs, as people who visit dentists regularly are more likely to be diagnosed and managed.\textsuperscript{[12]}

NPDCs are usually clinically asymptomatic. Patients with the cyst might experience soft-tissue elevation and fluctuance at anterior palate. Associated pain, swelling and drainage may be reported. Paresthesia of the anterior palate can occur as a result of pressure on the nasopalatine nerve. Mobility and displacement of teeth are rare.\textsuperscript{[10]}

Vitality and percussion tests of the anterior maxillary teeth should be performed to avoid unnecessary endodontic treatment. Radiographically, an ovoid-shaped, well-defined, unilocular radiolucency with a sclerotic boarder can be appreciated. Sometimes, a heart-shaped radiolucency can be seen as a result of superimposition of the anterior nasal spine.

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A prominent incisive canal can often mimic the radiographic findings of a NPDC. Most literature supported that a NPDC should be considered when the radiolucent area is more than 6 mm of diameter at the midline of the anterior maxilla.\textsuperscript{[10,11]} The clinical and radiographic findings of the two patients in this study were very consistent with the literature. Both patients were asymptomatic clinically with insignificant findings from the soft tissue and dentition. The radiographic findings for both patients revealed a unilocular, well-demarcated radiolucent lesion that is greater than 6 mm at the anterior maxilla.

A histopathological study of the cyst confirms the diagnosis of NPDC. The type of epithelial lining from the nasopalatine duct can be variable, depending on its relative proximity to the nasal and oral cavity. The superior part of the duct usually contains ciliary cylindrical cells that are consistent with nasal epithelium. The lining type
can switch to cuboidal epithelium as the duct travels inferiorly and eventually to squamous epithelium as it reaches the oral cavity.\textsuperscript{[10,13]}

Abrams et al. reported that 82% of the 61 NPDCs were of squamous epithelium, followed by pseudostratified columnar and cuboidal.\textsuperscript{[14]} However, it is also common that the epithelial lining has a combination of either stratified and pseudostratified squamous, transitional, columnar, cuboidal or ciliated columnar epithelium.\textsuperscript{[15]}

In this study, the cystic lining from Patient One was cuboidal epithelium, while that from Patient Two was squamous epithelium.

Differential diagnosis must be established before definitive treatment. Odontogenic cysts, such as radicular cyst, lateral periodontal cyst and odontogenic keratocyst (OKC), odontogenic tumor, such as ameloblastoma, and non-odontogenic tumors, such as central giant cell granuloma and central hemangioma, should be included as the differentials when tentatively diagnosing NPDCs based on clinical and radiographic presentations.\textsuperscript{[10,13]} There have been reports of misdiagnosing OKC crossing the midline at the anterior maxilla as NPDC.\textsuperscript{[16]} It is, therefore, critical for the clinician to include other lesions of the midline maxillary region in the differential diagnosis to help guide proper and timely management of the patients.\textsuperscript{[10]}

The treatment of NPDCs involves enucleation and curettage of the cyst, although a few reports in the literature support marsupialization of large cysts. The recurrence rate of NPDC after proper surgical management is extremely low.\textsuperscript{[10,11]} The neurovascular bundle within the incisive canal can cause profuse bleeding during surgery. Paresthesia of the anterior palate is also possible due to nasopalatine nerve injury during the procedure. There was no profuse bleeding or reported paresthesia for the two patients in this study. However, Patient Two’s maxillary central incisors were tested non-vital in subsequent follow-ups. Platelet-rich fibrins were applied in both cases to accelerate wound healing and reduce postoperative pain and infection.\textsuperscript{[17]}

Conclusion

NPDCs occur in 1% of the population, with a slight male predilection and no racial predilection. It is often seen in patients between 30 and 60 years of age. The clinical presentation of NPDCs can be asymptomatic or include pain, swelling and drainage from the anterior palate. A well-circumscribed, unilocular, ovoid- or heart-shaped radiolucent lesion crossing the midline of the anterior maxilla is appreciated on radiographs. Histopathological findings include squamous, cuboidal, columnar or respiratory cells, or a combination of these.

A correct diagnosis must be established to avoid improper treatment. NPDCs should be surgically managed with enucleation of curettage in a timely fashion to prevent complications, such as malignant transformation of the epithelial lining. \(\text{\textcopyright}\)

Queries about this article can be sent to Dr. Sun at faye.sun.1227@gmail.com.

REFERENCES


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