

# NASOPALATINE DUCT CYST: OFTEN MISDIAGNOSED AND MISMANAGED

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## Abstract

Nasopalatine duct cyst is the most common non-odontogenic cyst in the oral cavity which is a development cyst that originates from epithelial remnants of nasopalatine duct. They are usually asymptomatic and found during routine clinical and radiographic examination. Most of the time the symptomatic lesions may get misdiagnosed that lead to inadvertent endodontic therapy of vital permanent maxillary incisors. So, clinician should be aware of the clinical and radiological features of the cyst. This article presents two cases of nasopalatine cyst that were earlier misdiagnosed and mismanaged. Both cases suggests CT scan is worth in making a tentative diagnosis which is confirmed by histological examination only. The treatment modality of nasopalatine dust cyst is enucleation of the cyst.

**Keywords:** Computed tomography, Endodontic therapy, Enucleation, Nasopalatine duct cyst, Nasopalatine foramen, Nonodontogenic cyst,

## Introduction

The nasal cavity communicates with the anterior region of the maxilla via a bony canal called the nasopalatine canal. The nasopalatine canal contains remnants of the nasopalatine vessels, nerves, and the nasopalatine duct, which is considered to be a primitive organ of smell. The canal opens at the midline in the anterior maxilla below the retroincisive papilla. During embryonic development, the duct narrows gradually to form one or two central clefts in the midline. Usually there are two nasopalatine foramen, right and left with independent neurovascular bundles, but there can be as many as six different foramina. The foramina may sometimes contain only vascular elements known as Scarpa's foramina.

Etiology of nasopalatine duct is unknown. However the epithelial remnants of the nasopalatine duct undergo proliferation and cystic degeneration to form the nasopalatine duct cyst.<sup>1,2</sup> The most likely explanation for the cystic transformation is the spontaneous proliferation theory.<sup>3,4</sup> Trauma and infections are considered triggering factors for cystic degeneration even though evidence based support is not documented.<sup>2,5-7</sup> The first description of nasopalatine duct cyst (NPDC) was given by Meyer in 1914.<sup>8</sup> Synonyms for NPDC are: anterior middle cyst, maxillary midline cyst, anterior middle palatine cyst, and incisor duct cyst.<sup>3,4</sup> It may occur anywhere along the course of the nasopalatine duct. If occurrence is near the nasopalatine foramen on soft tissues, it is called 'cyst of palatine papilla'.<sup>9</sup> Past literature considered NPDC as a fissural cyst, but WHO classified it as a developmental, epithelial, non-odontogenic cyst.

NPDC is the most common non-odontogenic cyst in the oral cavity.<sup>10</sup> It occurs in about 1% of the population and comprises 1% of all maxillary cysts.<sup>11</sup> The age distribution of the lesion is broad with common incidence between 4th and 6<sup>th</sup> decades of life with a mean age of 42.5 years.<sup>12</sup> The lesion has a male predilection with a ratio of 3:1.<sup>5,12</sup> Lack of representative studies give a vague idea on racial predilection but there is suggestion for higher occurrence in young African Caribbeans with clinical aggressive lesions.<sup>13</sup> NPDC is usually an asymptomatic lesion with 40% of cases being totally asymptomatic, found only on routine clinical and radiographical examination.<sup>14,15</sup>

Vasconcelos *et al*, in a retrospective study, reported 87% of cases as asymptomatic.<sup>8</sup> In symptomatic lesions clinical manifestations will range from pain, discharge from the anterior palate, labial swelling, pruritus, ulceration, or fistulation. The pain may be due to compression of adjacent neural structures or secondary infection.<sup>3,4</sup> NPDC usually do not cause nonvital teeth unless it becomes large enough to disseminate the infection to the periradicular area of incisors. The radiographic manifestation of the lesion is a well-delineated rounded or ovoid radiolucency that can be inverted pear or heart-shaped because of superimposition of the anterior nasal spine.<sup>1,2</sup> The radiolucency will predominantly be around the periapical region of maxillary central incisors leading to unwanted endodontic approaches of these vital teeth. An enlarged nasopalatine duct should also be considered before reaching the diagnosis in cases of NPDC. The tentative diagnosis is based on the clinical history, clinical examination including vitality tests, and radiographs, particularly a computed tomographic scan. The definitive diagnosis is based on histopathology of the lesion. This article describes two case reports of NPDC that are symptomatic with a history of misdiagnosis.

## Case Reports

### Case I

A 19 year old male patient reported to the Department of Conservative Dentistry and Endodontics with pain to the maxillary anterior teeth for the past two weeks. The patient reported a history of endodontic therapy to all maxillary incisors in the past 2 years. There was no history of any traumatic injury. Medical history was non-contributory. Clinical examination revealed a sinus tract on the labial aspect of 21. Both central incisors were tender to percussion. Periapical radiographs revealed radiolucencies at the periapical region of all four incisors with faulty obturations and loss of coronal restorations [Figure 1]

Retreatment of all four incisors was planned and completed with hand instruments and the lateral condensation technique. After six months, the patient reported with purulent discharge from the anterior palate. He also reported a salty taste from the discharge and a foul smell.



Figure 1: Pre-operative patient reported before treatment

A periapical radiograph taken with gutta percha in the sinus tract traced to the midpalatine suture area. An occlusal radiograph showed the gutta percha pointing to an oval radiolucency at the midpalatine area. [Figure 2A & 2B]

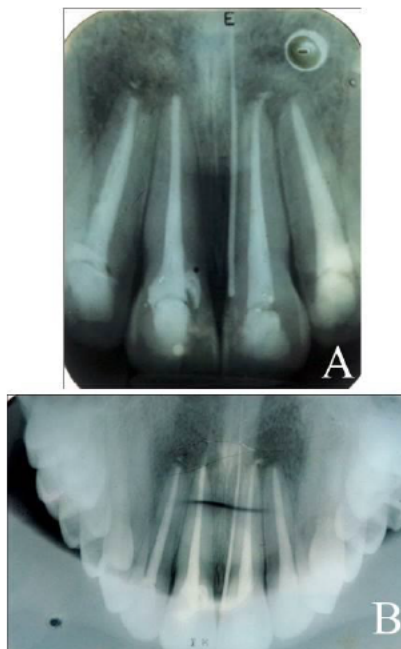


Figure 2: IOPA and Occlusal radiograph showing gutta percha pointing towards radiolucent area in midpalatine suture after 6 months of retreatment.

A CT scan showed an enlarged nasopalatine foramen and radiolucent lesion on the midpalatine suture area. [Figure 3]



Figure 3: CT scan images showing enlarged nasopalatine canal and foramen

There was also erosion of the bone at the periapical region of 21. Based on the clinical symptoms, findings, and CT scan report, we arrived at a tentative diagnosis of NPDC. Enucleation of the cyst was planned. The cyst was approached from the palatal aspect under local anesthesia. The enlarged nasopalatine foramen [Figure 4] was clearly seen after raising the mucoperiosteal flap.



Figure 4: Enlarged nasopalatine foramen after raising the palatal flap

The cyst with the epithelial lining [Figure 5] was enucleated and flap is sutured back.



Figure 5: Enucleated cystic lesion

At 6 months recall the sinus tract and periapical lesions of the incisors showed radiographic healing. [Figure 6]



Figure 6: IOPA after 6 months follow-up.

*Case II*

A thirty five year old female patient reported to the Department of Conservative Dentistry and Endodontics with pain and purulent drainage from a sinus tract



associated with the maxillary anterior teeth in the anterior palatine area. She reported a history of endodontic treatment of the central incisors twice in the past two years with no relief. Medical history was non-contributory. The two central incisors had mild tenderness to percussion. There was no mobility or periodontal pockets related to the central incisors. A periapical radiograph taken with gutta percha in the sinus tract traced toward a radiolucent area over 21. [Figure 7]

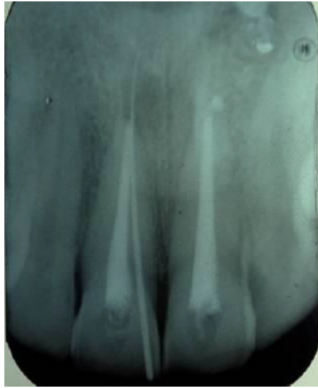


Figure 7: Pre-operative IOPA before periapical surgery

Based on these findings and previously root canal treated teeth, periapical surgery was planned. Root end resection was performed on both central incisors. After 2 months the patient reported with purulent drainage from the anterior palatal region. The periapical radiographs taken showed a heart shaped radiolucent area in between the central incisors. [Figure 8]



Figure 8: IOPA showing heart shaped radiolucency

A CT scan was done and showed an enlarged 7 mm nasopalatine foramen and a clear radiolucent oval area on the midpalatine region. [Figure 9]

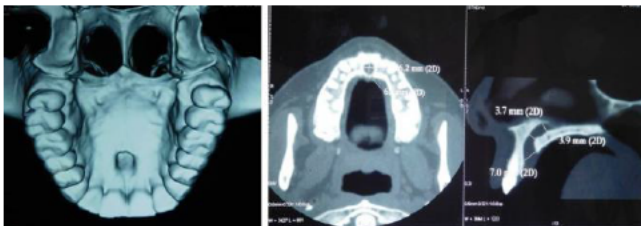


Figure 9: CT Scan images

A tentative diagnosis of nasopalatine duct cyst was made and enucleation of the cyst was planned. The muco-periosteal flap raised from the palatal aspect showed an enlarged incisive foramen. The cystic lesion with the epithelial lining is enucleated and the flap is sutured back. [Figure 10]

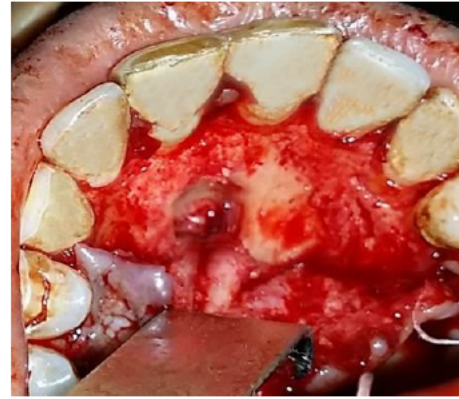


Figure 10: Enlarged Nasopalatine foramen with cystic lesion

**Discussion**

Nasopalatine duct cyst is a rare entity that is usually asymptomatic and encountered only during routine examination,<sup>2</sup> but the two cases reported here were symptomatic in nature. For the first case, we can make two hypotheses. One is that the patient previously had a misdiagnosed nasopalatine cyst by the practitioner and underwent endodontic therapy. The other is that improper management of the periapical lesions of the maxillary incisors could have chronically stimulated the formation of a nasopalatine duct cyst. In Case 2, there was clearly a misdiagnosis and the patient was subjected to inadvertent endodontic therapy and periapical surgery.

The etiology of NPDC is said to be unknown.<sup>3,4</sup> In Case 2, the etiology is unknown and the result of spontaneous proliferation theory. In Case 1, the persistent periapical pathology of the maxillary incisors might have disseminated to the nasopalatine duct which triggered the cystic degeneration, therefore supporting evidence of infection as a triggering factor.<sup>2,5,7</sup> The patients in this report were nineteen and thirty-five years of age which supports the broad range of occurrence of the nasopalatine duct cyst.

The NPDC may present with minor symptoms for a long time. The most frequent complaint will be a small well defined swelling posterior to the palatine papilla.<sup>2,5</sup> In some cases the lesion is aggressive leading to penetration of the labial plate with swelling below or to the one side of the maxillary labial frenum, or distortion of the nasal septum to cause a bulge in the nasal cavity. Sometimes burning sensation or numbness over the palatal mucosa is experienced due to pressure from the cyst on nasopalatine nerves.<sup>2,9</sup> In some cases, cystic fluid will drain to the oral cavity through a sinus tract and the patient will report a salty taste. These two cases had the same complaint but the

first patient reported the occasional salty taste in his mouth. The extension of this cyst posteriorly into the hard palate is referred to as a median palatal cyst. The term median anterior maxillary cyst is used when the cyst extends anteriorly between central incisors, expanding labial bone and diverging teeth.

The radiographic feature of the cyst is a well-defined circular or oval radiolucency. Sometimes the nasal spine gives a radioopaque shadow on the cyst giving a heart shaped or inverted pear shape,<sup>1,2</sup> more evident in Case 2. Root divergence of central incisors may occasionally be evident as well as root resorption. An enlarged or large incisive foramen is the most common differential diagnosis.<sup>16-19</sup> The incisive foramen may show great variation in size making it difficult for clinicians to differentiate between a large incisive foramen and a small asymptomatic cyst based on radiographs only. A diameter of greater than 6mm is considered to be cystic but clinical symptoms should also be taken into consideration. A rule of thumb is that if the radiolucency of the incisive canal is less than 6mm, it should not be considered cystic in the absence of other symptoms. In the two cases reported, the canals were more than 6mm in diameter and symptomatic.

Before arriving at a diagnosis, a differential diagnosis should be made to avoid inadvertent treatments in vital permanent maxillary central incisors. It is very common to see lesions of maxillary incisor lesions misdiagnosed as a periapical cyst or granuloma and treated via endodontic therapies, like these 2 cases mostly. Pulp vitality testing of teeth is helpful. In most cases of NPDC, the incisors will give a positive vitality test response unless the lesion extends to destroy the nerve supply of the teeth.<sup>19,20</sup> A percussion test should also be done to rule out periodontal defects. An asymptomatic nasopalatine cyst will have similar radiographic features as the radicular cyst or granuloma.<sup>20</sup> The presence or absence of lamina dura and enlargement of the periodontal ligament space is indicative of the inflammatory lesion. By shifting the horizontal angulation of the periapical radiograph, the radicular cyst may be centered over the incisors while NPDC shows an altered position. CT scan is most accurate in establishing a tentative diagnosis because fine detail of structures normally intact adjacent to the lesion can be viewed.<sup>21</sup> MRI is also reliable in the diagnosis of NPDC with specific axial T1-weighted images showing the presence of fluid, viscous and protein material within the cyst, and keratin abundance in the superficial level.<sup>22</sup> Other differential diagnoses include central giant cell granuloma, follicular cyst associated with mesiodens, primordial cyst, nasoalveolar cyst, osteitis with palatal fistulation and bucconasal and/or buccosinus communication.<sup>23</sup> The diagnosis is confirmed by histopathological examination. The NPDC is lined by different types of epithelium ranging from stratified squamous epithelium to pseudostratified columnar or a combination of the above.<sup>11,12</sup> 9.8% of cases have shown respiratory epithelium.

The treatment of choice of NPDC is enucleation of the cyst. If the vitality of the teeth is affected, endodontic therapy is also indicated. The two cases were treated successfully with enucleation of the cyst. In Case 1, retreatment of the incisors also helped to resolve the periapical pathology. Recurrence of the lesion is low with incidence varying from 0-11% in the literature.

### Conclusion

Nasopalatine duct cyst is of uncertain origin with nearly 40% of the lesions are totally asymptomatic and found only during routine examination. So clinician should be aware of clinical and radiological features of the cyst. It must be distinguished from other maxillary anterior radiolucencies. CT scan is worth in confirming tentative diagnosis of NPDC. Vitality and percussion tests should be done to avoid inadvertent endodontic treatment modalities of vital anterior teeth. Early surgical removal is the treatment modality with elimination of irritative factors to prevent recurrence.

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