



## NASOPALATINE DUCT CYST MASQUERADING AS DENTIGEROUS CYST – A RARE CASE REPORT

**Manisha D Yadav\***

Post Graduate Student. G.D. Pol. Foundation Y.M.T.D.C, Navi Mumbai, Maharashtra, India. Dept. of Oral Medicine and Radiology. \*Corresponding Author

**Deepa Das**

Professor and HOD. G.D. Pol. Foundation Y.M.T.D.C, Navi Mumbai, Maharashtra, India. Dept. of Oral Medicine and Radiology.

**Bhakti Patil Soman**

Reader. G.D. Pol. Foundation Y.M.T.D.C, Navi Mumbai, Maharashtra, India. Dept. of Oral Medicine and Radiology.

### ABSTRACT

The Nasopalatine duct cyst (NPDC) or the incisive canal cyst is the most common non-odontogenic cyst in the maxilla. It is thought to originate from epithelial remnants of Nasopalatine duct (NPD), which might be stimulated to proliferate by trauma, infection, mucous retention or by spontaneous cystic degeneration. The clinical and radiographic presentation of NPDC could be varied and can often pose a diagnostic challenge. It usually presents as a painless swelling in the area of the incisive papilla and on radiographs as a well-defined heart shaped radiolucency in the premaxillary region between the central incisors. NPDC was considered as a fissural cyst in the past, but now WHO has classified it as a developmental, epithelial, non-odontogenic cyst. A rare case of NPDC which was at first misdiagnosed as dentigerous cyst on conventional radiography due to the presence of an impacted tooth is presented here. The utility of advanced imaging for proper diagnosis of NPDC is highlighted through the present case report. This case emphasizes the necessity to study maxillary cysts extensively both clinically and with advanced radiography for accurate diagnosis.

**KEYWORDS :** Nasopalatine duct cyst, Incisive canal cyst, non-odontogenic cyst, mesiodens.

### INTRODUCTION:-

The nasopalatine canal/incisive canal is a narrow bony canal connecting the nasal and oral cavities.<sup>1</sup> Nasopalatine duct cyst (NPDC) occurs in the median of the palate within the incisive canal, located in the palatine process of the maxilla, posterior to the maxillary central incisors. The WHO (World Health Organization) has classified nasopalatine duct cyst as a developmental, epithelial, non-odontogenic cyst of the maxilla and it constitutes around 1.7-11.9% of all cysts of jaws.<sup>2,3,4</sup> It arises as a result of proliferation of embryonic epithelial remnants of the nasopalatine duct.<sup>2,3,5</sup>

Mesiodens, is a supernumerary tooth located between the two central incisors of the maxilla.<sup>6</sup> A mesiodens is usually conical in shape and its position can be normal, inclined or inverted.<sup>7</sup> It can induce many complications like alteration of tooth eruption, root resorption and cyst formation.<sup>7</sup> A rare case of NPDC which was radiographically misdiagnosed as a dentigerous cyst and associated with a mesiodens is reported here.

### Case Report:-

A 28-year-old housewife reported with the chief complaint of a swelling in the anterior region of upper jaw since 1 month and pain in the same region since 2-3 days. The swelling which was initially small and painless gradually increased in size. She also gave history of trauma to the same region 3 days before while playing with her child, after which she started experiencing pain.

On clinical examination an extraoral swelling was seen in the anterior region of upper jaw (mid-face) obliterating the nasolabial fold bilaterally (Figs:- 1 a & b).

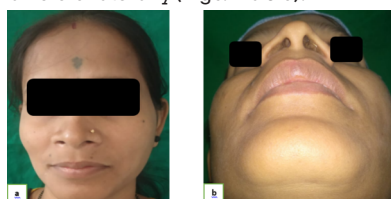


Figure 1(a & b) :- Extraoral swelling obliterating the nasolabial fold bilaterally.

On palpation the swelling was warm, tender and firm in consistency. Intraorally, a single well-defined dome shaped swelling was seen obliterating the upper labial vestibule extending from distal of 13 to mesial of 22. The mucosa over the swelling appeared to be slightly bluish (Fig:-2a). There was mesioproximal caries in relation to 11, 21, 22 and 11, 21 were tender on percussion. There was no palatal counterpart for the swelling nor any draining sinus (Fig:-2b).

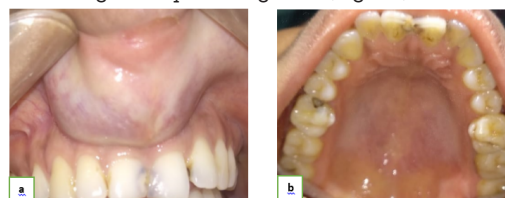


Figure 2(a & b):-Intraoral swelling labial to upper incisors with no palatal counterpart.

Based on clinical findings, we arrived at a provisional diagnosis of a benign odontogenic lesion in the anterior maxilla. 11, 21 & 22 were found to be vital on pulp testing. Intraoral radiographs (IOPA & Maxillary Occlusal) presented with a heart shaped radiolucency apical to and diverging the roots of 11&21. A tooth shaped radiopacity was also noted apical to 11 within the radiolucency (Figs:-4&5). Orthopantomogram (OPG) (Fig:-3) showed a well-defined radiolucency in the anterior region of maxilla, raising the floor of nasal fossa in relation to 11&21. The presence of an inverted and impacted mesiodens in relation to 11 was also observed (Figs:- 4,5).

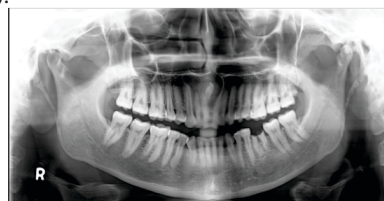
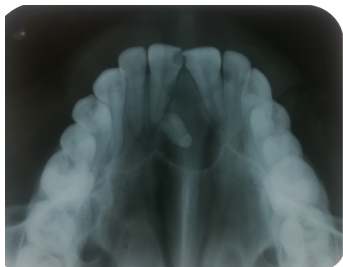


Figure 3:- Panoramic Radiograph showing a well-defined heart shaped radiolucency in the anterior maxilla with inverted and impacted mesiodens.

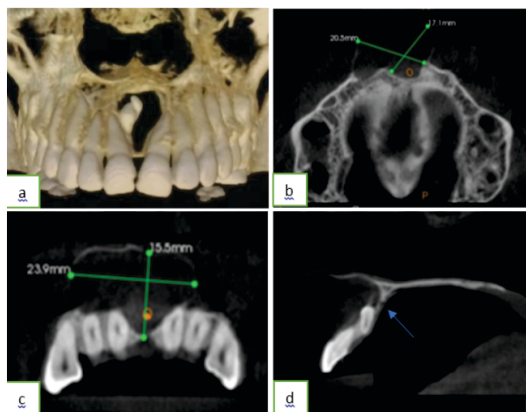


**Figure 4:-** Periapical radiograph showing a well-defined radiolucency apical to and diverging the roots of central incisors with an impacted and inverted mesiodens.



**Figure5:-** Maxillary cross-sectional occlusal radiograph showing a well-defined heart shaped radiolucency with an impacted and inverted mesiodens.

On CBCT scanning, in the axial plane a well-defined hypodense area was seen with a maximum dimension of 23.9mm mediolaterally. Expansion and perforation of the labial cortical plate in relation to 11,21,22(Fig: -6b) was observed. In the coronal section, a well-defined hypodense area was observed extending from the floor of nasal fossa superiorly and to the interdental alveolar crest in relation to 11,21&22 inferiorly(Fig:-6c). In the sagittal section, presence of a mesiodens palatally along with slight thinning of palatal cortical plate was noted (Fig:-6d).

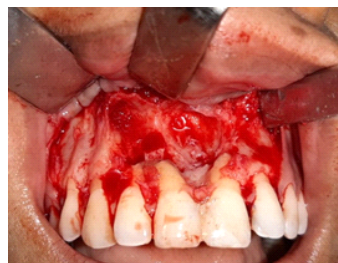


**Figure 6 (a, b, c, d):-**CBCT scan showing 3D reconstruction image and maximum dimension of the lesion in axial and coronal plane along with impacted and inverted mesiodense causing localized thinning of the palatal cortical plate in the sagittal section.

Based on the radiographic findings, a diagnosis of dentigerous cyst with impacted and inverted mesiodens was arrived at. Fine Needle Aspiration Cytology (FNAC) was positive with a chocolate colour fluid aspirate, which on histopathology was reported to be a chronic inflammatory lesion (Fig:-7). Root canal treatment was done with 11,21&22, after which surgical enucleation and removal of the impacted and inverted mesiodens was planned through a labial approach.

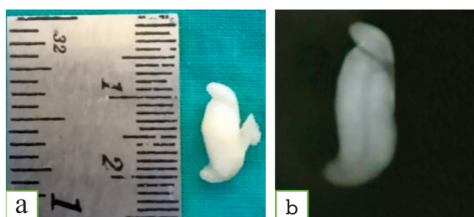


**Figure7:-** FNAC with chocolate colour aspirate.



**Figure8:-** Trapezoidal Flap is elevated using labial approach exposing the root surfaces of the upper incisors.

A trapezoidal surgical flap was raised from 13 to 23 and enucleation of cyst was done along with removal of the mesiodens followed by apicectomy of the root canal treated teeth 11,21&22 (Fig:-8). Then the cavity was curetted, irrigated and the specimen was sent for histopathological examination (Figs:-9a&b). The patient was recalled after 1 week and healing was found to be satisfactory (Fig: -10).

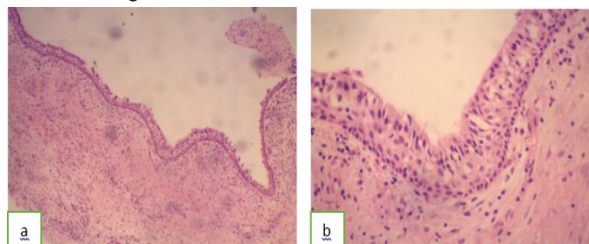


**Figure 9(a & b):-** Extracted mesiodens with radiograph.



**Figure 10:-** Post-operative 1 week follow up.

Histopathology of the excised specimen showed presence of a cystic lumen lined by a combination of thin non-keratinized stratified squamous cell epithelium and pseudo-stratified columnar epithelium. Connective tissue showed presence of chronic inflammatory infiltrate, bundles of collagen fibres and dilated blood vessels with one or two distinct neurovascular bundles (Fig:-11a&b). Based on the clinical, radiographic and histopathological features a final diagnosis of nasopalatine duct cyst with impacted, inverted mesiodens was made. A maxillary occlusal follow up radiograph was taken 2 months post operatively which showed adequate healing and bone formation(Fig:-12).



**Figure 11 (a & b):-** photomicrograph 10 X & 40 X

magnification (H & E stained section showing cystic lumen lined by combination of thin non-keratinized stratified squamous cell epithelium and pseudo stratified columnar epithelium. Dilated blood vessels with few neurovascular bundles seen in the connective tissue).



**Figure 12:-A 2-month postoperative maxillary occlusal radiograph showing adequate healing and bone formation.**

#### DISCUSSION:-

Nasopalatine duct cysts are the most common non-odontogenic cysts of the jaws accounting for 1% of all maxillary cysts.<sup>8,9</sup> The NPDC was first described by Meyer in 1914 who called them supernumerary nasal sinus.<sup>3,4,8,10-15</sup> They were previously thought to be fissural cysts and were known by synonyms like anterior middle cyst, maxillary midline cyst, anterior middle palatine cyst and incisor duct cyst.<sup>4,8,9,15</sup> These cysts are thought to originate either by proliferation of epithelial remnants of the nasopalatine duct, due to trauma, infection or mucous retention or by spontaneous cystic degeneration of epithelial remnants.<sup>3,10</sup>

NPDCs occur three times frequently in males than in females and are prevalent between 40 to 60 years of age.<sup>8,9</sup> The mean age of occurrence has been reported to be from 31 to 54 years.<sup>10</sup> The patient in the case reported here is a 28-year-old woman. According to the literature reported till now, NPDCs are mostly asymptomatic, unless secondarily infected, when they can present with symptoms of swelling, drainage of pus and pain.<sup>10</sup> In our case, the patient had presented with a painful swelling in the upper anterior jaw. NPDCs commonly cause a palatal swelling, but in our case, there was obliteration of labial vestibule only. It is quite possible that larger cysts tend to be more symptomatic, due to the pressure on adjacent structures.<sup>8,9</sup> There are reports claiming that, the more posterior the location of cyst is, the sooner symptoms are likely to appear.<sup>8,9</sup> Infected ones can present with pus discharge through the nasopalatine papilla. The maxillary incisors remain vital as we have seen in the present case. The differential diagnosis for NPDCs may include radicular cyst associated with maxillary central incisors, dentigerous cyst associated with mesiodens, primordial cyst, nasoalveolar cyst and so on. In the present case, a provisional diagnosis of benign odontogenic lesion in relation to anterior maxilla was made and a differential diagnosis of radicular cyst in relation to 11,21,22 was given.

The classical appearance of NPDC in the radiograph is an oval or heart shaped radiolucent lesion above the root apices of the maxillary incisors, at times, even overlapping their roots.<sup>9</sup> Roper – Hall has been the first person to recommend the use of radiographic size in diagnosing NPDCs. He suggested that since the average diameter of NPDCs was approximately 3mm, any radiolucent lesion less than 6mm in diameter should be considered within normal limits.<sup>10</sup> The mean radiographic diameter of NPDCs have been reported to be ranging from 6 to 15 mm.<sup>10</sup>

In our case, a radiographic diagnosis of dentigerous cyst was made as the radiolucency was circumferential to the mesiodens which was inverted and impacted. The diameter of the lesion was about 23mm on OPG (Fig-3). On CBCT, the dimensions were 23.9mm mediolaterally, 15.5mm

superoinferiorly & 17.1mm anteroposteriorly (Fig:-6b, c).

FNAC of NPDCs have been reported to give yellowish or brownish aspirate,<sup>5</sup> and we also obtained a chocolate coloured aspirate. Depending on the dimension, NPDCs are surgically managed by enucleation or marsupialization.<sup>5</sup> In the case presented, the cyst was enucleated by labial approach along with the removal of mesiodens.

On histopathological examination of NPDCs, more than one epithelial type is commonly found. Squamous epithelium in 75% to 82% of NPDCs, followed by ciliated columnar epithelium in 23% to 42% of the cases.<sup>10</sup> In the histopathology report of our case, there was a combination of the above two epithelia. The presence of one or two distinct neurovascular bundles was also observed (Fig:-11a&b). NPDCs have been reported to have a low recurrence rate, ranging from 0% to 11%.<sup>10</sup> Nevertheless, they have to be followed up so that any recurrence is promptly diagnosed and managed.

The case presented is unique as there was obliteration of the upper labial vestibule, while NPDCs usually present as palatal swellings. Another finding which adds to the rarity of this case is the presence of mesiodens within the cyst. Only two cases of NPDCs associated with supernumerary tooth have been reported till now.<sup>2,11</sup> The importance of reporting this case is to emphasize the fact that NPDCs can be easily misdiagnosed as dentigerous cyst or periapical cyst. Therefore, maxillary cysts should be extensively studied clinically, and radiographically with definitive diagnosis established histopathologically.

#### CONCLUSION:-

Nasopalatine duct cysts are of uncertain origin. The purpose of reporting a rare NPDC associated with an impacted and inverted mesiodens causing labial cortical plate expansion is to emphasize the dilemma in diagnosing such lesions.

It was clinically misdiagnosed as radicular cyst because of the presence of proximal caries along with history of trauma in relation to the associated teeth. Radiographically again, the lesion was misinterpreted as dentigerous cyst due to the presence of an impacted and inverted mesiodens. A definitive diagnosis was established by the histopathological examination of the surgical specimen. Hence, a triangular approach of clinical, radiographic as well as histopathological correlation is imperative for definitive diagnosis of a NPDC.

Thus, it is very important to consider NPDC in the differential diagnosis of radiolucent lesions in the maxillary anterior region.

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