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An enigmatic radiographic diagnosis of Nasopalatine duct cyst

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ABSTRACT

Aim – To present a case of nasopalatine duct cyst in a 21 year old female which was diagnosed radiographically and confirmed later by histopathological examination

Background - There are several developmental cysts derived from embryonic structures or faults in their remnants located in oro-facial region. Nasopalatine duct cyst (NPDC) is the most common of all the developmental, epithelial and non-odontogenic cysts of the maxilla. This cyst originates from epithelial remnants from the nasopalatine duct. Generally, the patients present without clinical signs and symptoms. Therefore, the tentative diagnosis "nasopalatine duct cyst" is often based on a coincidental radiological finding on a routine panoramic view or occlusal radiograph.

Conclusion –NPDC is the most common non-odontogenic, developmental cyst of oral cavity and should be differentiated from other radiolucencies of anterior maxilla

Clinical significance - Although NPDC is not rare, it is mostly misdiagnosed as periapical pathology. The definite diagnosis should be based on clinical, radiological and histopathologic findings.

KEYWORDS Cysts, Developmental, Naso palatine, Non-odontogenic

Introduction

Jaw cyst is a pathological intra-osseous cavity lined by a membrane and may contain liquid or semisolid material. Cysts in maxillary, mandibular and perioral regions widely change in distribution, characteristics, histogénesis, incidence, behavior and treatment. Authors have classified cysts in odontogenic, non-odontogenic cysts, and pseudocysts. Odontogenic cysts originate from odontogenic epithelium and arise in the tooth-bearing regions of the jaws. Non-odontogenic cysts probably derive from the proliferation of non-odontogenic epithelial remnants that are trapped along fusion lines during embryogenesis of the cephalic district. Pseudo-cysts differ from a real cyst because of the absence of the epithelial membrane.[1]

The nasopalatine duct cyst (NPDC) is an intraosseous developmental cyst of the midline of the anterior palate.[1]The nasopalatine duct communicates the nasal cavity with the anterior region of the upper maxilla. It is located on the midline and palatine to upper maxilla, above the retroincisor palatal papilla. During fetal development, the duct gradually narrows until one or two central clefts are finally formed on the midline of the upper maxilla. The nasopalatine neurovascular bundle is located within the duct, and emerges from its intrabony trajectory through the nasopalatine foramen. There can be as many as six different foramina, though there are usually only two, with independent neurovascular bundles right and left. The vascular and neuronal elements can emerge separately, thus foramina containing exclusively vascular elements are known as Scarpas's foramina. Often mistaken for an enlarged nasopalatine duct, NPDC are of uncertain origin. These were considered as fissural cysts in the past, these lesions have been classified by World Health Organization as developmental, epithelial, non-odontogenic cysts. The NPDC was first described in 1914 by Meyer.[2-4]

NPDCs are the most common non-odontogenic cysts of the oral cavity, representing up to 1% of all maxillary cysts.[3] Its other names includes anterior middle cyst, maxillary midline cyst, anterior middle palatine cyst or incisor duct cyst. It is more common in 4th and 6th decade of life[2-6]and three times more frequent in males than females. The lesion is unlikely to manifest subjective symptoms being commonly detected only during routine radiographic examination. Large cysts may present with a number of symptoms ranging from pain, anterior palatal/labial swelling, discharge, itching, ulceration, local infection, fistualization. Commonly present centrally or unilaterally with no side predilection. Radiographically, the cyst presents as a well delineated rounded, ovoid, inverted pear-shaped or heart shaped radiolucency in the midline of anterior maxilla. Superimposition of anterior nasal spine explains its heart shaped presentation. Diameter greater than 6 mm of incisive foramen is consistent with NPDC.[2-7] It present with an average diameter of 1.5-1.7 cm. A thorough differential diagnosis must be established to avoid unnecessary treatments like endodontic procedures in vital permanent maxillary incisors. Differential diagnosis includes an enlarged nasoplatine duct less than 6 mm in diameter, central giant cell granuloma, radicular cyst, follicular cyst associated with mesiodens, ostetis with palatal fistualization. A correct tentative diagnosis is based on positive vitality testing and negative percussion findings of permanent maxillary central incisors with no pulp or periodontal problems. In addition to panoramic, periapical and occlusal radiographs, computed tomography (CT) is also advised. Magnetic resonance imaging (MRI) may also prove useful in establishing the diagnosis. Specific axial T1-weighted imaging reflects the presence of fluid, viscous and protein material within cyst and abundant keratin at superficial level.[1-3,5,7,8]

Case report

A 21 year old female reported to the department of Oral Medicine and Radiology with a chief complaint of a discolored tooth since 1 year. History of patient revealed trauma to upper front tooth region of jaw 2 years ago due to falling from stairs followed by fracture of upper front tooth followed by gradu-

al discoloration of tooth past a year. Patient was completely asymptomatic with no history of previous or current pain and swelling. Patient's medical and family histories were non-contributory. On clinical examination, there was Ellis's class IV fracture with 11 with no swelling, erythema or discharge with tooth or surrounding mucosa (Figure 1). There was negative tenderness on vertical and horizontal percussion with no vestibular tenderness. On the basis of history and clinical examination a provisional diagnosis of Asymptomatic apical periodontitis with 11 was given and a differential diagnosis of chronic periapical abscess and periapical cyst was made. Pulp vitality test was negative and intraoral periapical and occlusal radiographs revealed a solitary, well defined heart shaped radiolucency, measuring approximately 1.2x1.5 cm in dimension with its epicenter between maxillary central incisors causing flaring of their roots and superimposed by anterior nasal spine with an illdefined radiolucency and loss of lamina dura at periapex of 11(Figure 2). Thus a radiographic diagnosis of Nasopalatine duct cyst along with chronic periapical abscess with 11 was made. Root canal treatment with 11 was performed (Figure 3) followed by enucleation of cyst was performed under local anesthesia (Figure 4) and specimen was sent for histopathological examination which confirmed the diagnosis of NPDC (Figure 5). Patient was recalled for subsequent follow ups for 1 month every week which revealed no recurrence of lesion.

Discussion

NPDC is a developmental cyst derived from proliferation of embryonic epithelial remnants of nasopalatine duct. Trauma, infection and mucous retention within associated salivary gland ducts have all been suggested as possible pathogenetic factors, however spontaneous cystic degeneration of residual ductal epithelium is the most likely etiology and some genetic determinants have been suggested. As far as pathogenesis is concerned, it was previously thought that the NPDCs originated from the trapping of epithelium during fusion of the embryological processes. This concept has been discarded,and currently NPDC is thought to develop from the epithelial remnants of the nasopalatine ducts present within the incisive canals. Patients may be asymptomatic, with the lesion being detected on routine radiographs, however, many will present with one or more symptoms.[3] Complaints are often found to be associated with an infection of a previously asymptomatic nasopalatine duct cyst and consist primarily of swelling, drainage of pus an pain with bulging of nasal floor, perforation of labial and palatal bony palate and tooth displacement with numbness of palatal mucosa due to pressure on nasopalatine nerves.[3,4]The vitality of nearby teeth is not affected, however, it is not uncommon to see evidence of endodontic therapy because the nasopalatine duct cyst has been previously clinically misdiagnosed as a periapical cyst or granuloma as with our present case. Due to similar signs and symptoms, the NPDC may be misdiagnosed as a periapical lesion. This is why many authors believe that its prevalence is actually higher than presented in the literature. Although a large NPDC might show the adjacent incisors roots to be within the cystic cavity, the lamina dura will be intact and the pulp usually vital whereas a radicular cyst is associated with a pulpless tooth and involve a portion of the root, usually with loss of continuity of the lamina dura. Practitioner incisors. Thus, in establishing a diagnosis of NPDC it is important to attempt to exclude the possibility of a periapical lesion by performing the pulp vitality tests of the incisor teeth.[3] Histologically, the nasopalatine duct cyst is lined by stratified squamous epithelium alone or in combination with pseudostratified columnar epithelium (with or without cilia and/or goblet cells), simple columnar epithelium, and simple cuboidal epithelium. The fibrous wall generally contains nerves, arteries and veins. Additionally, minor salivary gland tissue and small islands of cartilage may be found. Finally, if the cyst is infected, acute and chronic inflammatory cells will be seen throughout the specimen.[3,9,10]

The treatment of choice is surgical excision of the cyst, although some authors propose marsupialization of large NPDCs. The nasopalatine neurovascular bundle is a delicate and highly vascularised structure giving rise to profuse bleeding if inadvertently sectioned during surgery. Electrocoagulation is required in such cases. Paresthesia of the anterior palatal zone is a rare complication found in 10% of the cases, on removal of nerve endings of the nasopalatine nerve along with the membrane of the cyst.[3,8] Frequently the biopsy procedure results in adequate treatment. Recurrence is rare and has been reported in 11% of patients.[2] A very rare complication associated with long standing NPDC is squamous cell carcinomas originating in maxillary bone. This is due to the metaplasia seen in the epithelial wall of a cyst or of the epithelial remains that participated in odontogenesis. Therefore, there are cases in which NPDC gives rise to squamous cell carcinoma in the anterior zone of the upper maxilla. This explains the need for early removal of the NPDC, with the purpose of minimizing the risks and complications.[3]

Conclusion

The present case is of particular interest as NPDC in combination with a periapical pathology secondary to a traumatic pulpal pathology is rare and challenging to diagnose. A thorough clinical history and radiographic examination can lead to avoidance of misdiagnosis and unnecessary treatments and early management of cyst to avoid complications.

Legends

Figure 1 – Elli's class IV fracture with discoloration of 11
Figure 2 – Intraoral periapical and occlusal radiograph showing well defined heart shaped radiolucency having epicenter as 11 and 21 with flaring of maxillary incisors
Figure 3 – Post obturation intraoral radiograph of 11
Figure 4 – Enucleation of cyst
Figure 5 – H& E stained sections showing the nasopalatine duct cyst lined by stratified squamous epithelium with pseudostratified columnar epithelium and fibrous connective wall containing nerves and vascular bundles

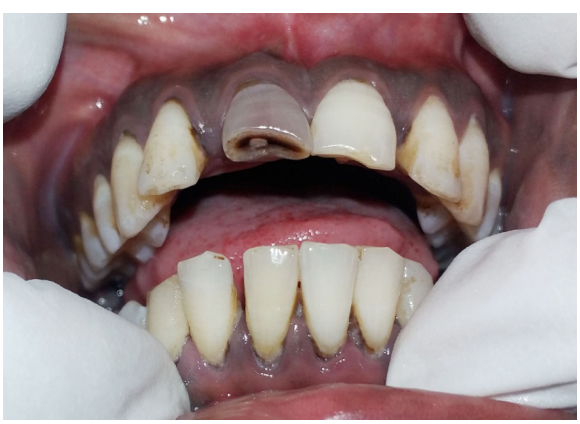

Figure 1

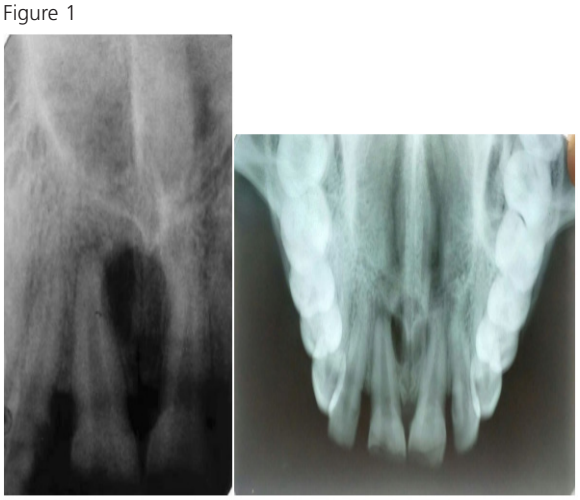

Figure 2

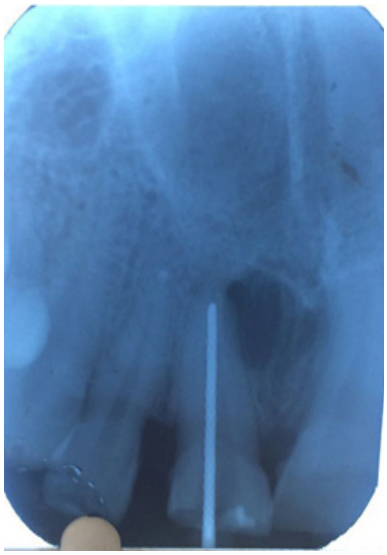

Figure 3

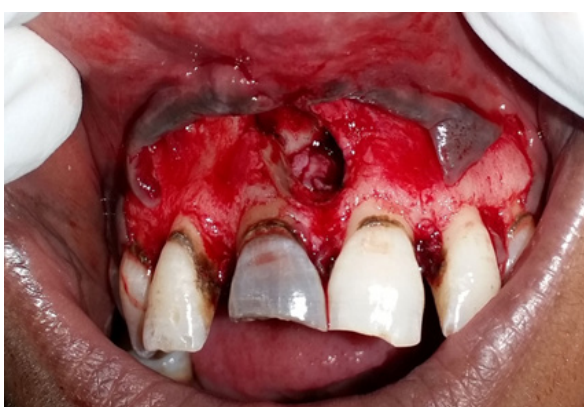

Figure 4

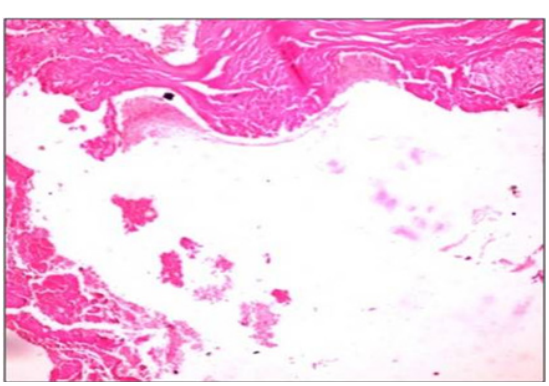

Figure 5

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