

Case Report of a Non Odontogenic Cyst Being Misdiagnosed

Neha Jain^{1*}, Ashutoshdutt Pathak², Sabhrant Singh², Sushil Kishnani³, Sheweta Narwani⁴ and Ruchi Jain⁵

¹MDS, Reader, Department of Oral and Maxillofacial Surgery, People Dental Academy, Bhopal, India

²MDS, Reader, Department of Oral and Maxillofacial Surgery, People College of Dental Science and Research Center, Bhopal, India

³MDS, Reader, Department of Conservative dentistry and Endodontics, People College of Dental Science and Research Center, Bhopal, India

⁴MDS, Reader, Department of Prosthodontics, People Dental Academy, Bhopal, India

⁵MDS, Reader, Department of Prosthodontics, Bhabha College of Dental science and research Center, Bhopal, India

***Corresponding Author:** Neha Jain, MDS, Reader, Department of Oral and Maxillofacial Surgery, People Dental Academy, Bhopal, India.

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Abstract

Non odontogenic cysts are developmental cyst originating from epithelial remnants which gets entrapped along the line of fusion between two developing bones during embryogenesis. Nasopalatine duct cyst (NPDC)/Incisive canal cyst is most frequently occurring non odontogenic cyst of anterior maxilla. The remnants of the epithelial cells of the nasopalatine duct may get stimulated spontaneously or by infection or trauma resulting in development of cyst. The cyst usually remains asymptomatic but can get infected for which the patient seeks doctor advice. A well - defined heart -shaped radiolucency between the maxillary central incisors is one of the pathognomonic radiographic features of NPDC. NPDC is frequently misdiagnosed as radicular cyst specifically if preceded by trauma. Clinical, radiographic and histopathology makes the final diagnosis. Surgical enucleation of the cyst is the treatment preferred.

We are reporting two cases of nasopalatine duct cyst along with its review of literature.

Keywords: *Nasopalatine Duct Cyst; Incisive Canal Cyst; Non Odontogenic Cyst*

Introduction

Meyer in 1914 was first to observe and describe nasopalatine duct cyst (NPDC [1]. It arises from remnants of nasopalatine duct which is an embryologic structure located in the midline of anterior palatal region of maxilla connecting the oral and nasal cavities. During fetal development, the duct gently narrows and finally disappears converting into incisive canal. The nasopalatine nerve, a branch of maxillary nerve is located within this duct, and emerges through the nasopalatine foramen [2].

NPDC is one of the most common nonodontogenic cyst and contributes about 1.7 - 11.9% of all jaw cyst. The cyst is mostly observed in patient of fourth to sixth decade with more affinity for male members [3]. The epithelial remnants of NPD either stimulated spontaneously or provoked by trauma or infection resulting in formation of NPDC [1,4]. Mostly they are asymptomatic, unless they are secondarily infected [5]. Swelling in anterior palatal region of maxilla is most frequently concern of patient for consulting the doctor. Surgical enucleation of cyst is the treatment preferred [6,7].

Case 1

A 35 year old male patient reported with the chief complaint of painless swelling with salty discharge from anterior region of palate from past one month. The swelling was sudden in onset, and was not followed by any trauma or pain in the tooth. The size of the swelling was constant in nature since inception. No history of paresthesia/difficulty in chewing/dysphagia could be elicited.

General physical and extraoral examination revealed no abnormalities and no lymphadenopathies. On intraoral examination, single round shaped swelling of size approximately 1 × 1 cm in anterior part of hard palate extending from incisive papillae to mid palatine raphe region. No observable defects with respect to maxillary anterior teeth. The overlying mucosa appeared normal in colour (Figure 1). On palpation the swelling was non tender and firm hard in consistency. Pus discharge was noted from the swelling. Vitality test revealed positive response with respect to 11, 21, 13, 21, 22 and 23.



Figure 1: Swelling in the mid palatal region..

Well defined heart shaped radiolucency between two central incisors of maxilla was seen on intraoral radiographs (IOPAR). No resorption of roots or displacement of teeth noted with respect to 11 and 21. Based on clinical signs and symptoms and after radiographical analysis a tentative diagnosis of Nasopalatine Duct cyst was given (Figure 2).



Figure 2: Heart shaped radiolucency in between two incisors.

Surgical enucleation under local anesthesia was done by gaining access through palatal mucoperiosteal flap. The cystic lining was curetted and sent for histopathological examination (Figure 3 and 4). Suturing was done and post-operative instructions were given to patient. Patient was called for suture removal one week post operatively and healing was satisfactory and uneventful (Figure 5).



Figure 3: Intraoperatively after cyst enucleation.



Figure 4: Excised cystic lining.



Figure 5: Post operatively after enucleation.

The final diagnosis of Nasopalatine duct cyst was made on histopathological examination of excised cystic lining.

Case 2

A 25 year old female patient reported with a chief complaint of swelling and pus discharge from anterior palatal region from past 2 months. The swelling was pin head sized which had gradually increased to attain the present dimension. History of accident was present in which maxillary anterior teeth were injured for which patient consulted a doctor. Radiograph of 12, 11 and 21, 22 were taken and root canal treatment was done in respect to 11 and 21 assuming it as any radicular cyst. But the swelling still persisted and 2 months later she noted a yellowish colour discharge from swelling and salty taste in mouth.

On extra oral examination no abnormality was detected and lymph nodes were not palpable. Intraoral examination revealed a single, well defined oval shaped swelling of approximately 1 X 2 cm in size (Figure 6). The swelling was tender on palpation and soft in consistency. Intraoral radiographs showed root canal treated maxillary incisors and in between both the incisors a well-defined heart shaped radiolucency with sclerotic borders was seen (Figure 7). Aspiration of the cyst was done which showed a straw-coloured fluid. Based on clinical and radiographic interpretation, we made a provisional diagnosis of radicular cyst. Differential diagnosis of nasopalatine cyst was also given.



Figure 6: Midpalatal swelling.



Figure 7: Radiographic presentation.

Enucleation of the cyst was done under local anesthesia via palatal mucoperiosteal flap (Figure 8 and 9). The enucleated cystic lining was sent for histopathological evaluation where it was diagnosed as nasopalatine duct cyst.



Figure 8: Cavity after enucleation.



Figure 9: Excised cyst.

Discussion

Meyer was first to describe the nasopalatine duct cyst [1]. It is a fissural non-odontogenic cyst occurring in oral cavity which corresponds to the 1% of all maxillary cyst [8,9] and the 26.6% of all non-odontogenic cysts [10]. Although being a developmental cyst, it is rarely seen in the first decade of life, mostly discovered in the fourth to sixth decade. The cyst affects three times more to males as compared to females [3].

The etiology of the cyst is unknown, although it is believed to develop from epithelial remnants of nasopalatine duct. Nasopalatine duct is an embryologic structure located in the midline of anterior palatal region of maxilla connecting the oral and nasal cavities. During fetal development, the duct gently narrows and finally disappears converting into incisive canal but its remnants in the form of epithelial cord remains. The epithelial remnants of NPD either stimulated spontaneously or provoked by trauma or infection resulting in formation of NPDC [1,4].

Most of NPDC are asymptomatic or cause minor symptoms that are tolerated for very long periods. Mostly patients may complain of a small asymptomatic swelling just posterior to incisive papilla. Larger cysts may also result causing bony resorption resulting in a fluctuant swelling of labial alveolar mucosa and anterior hard palate [15]. An uncommon case of a massive NPDC with nasolabial protrusion has been reported by Tanaka, *et al* [15]. The swelling may be associated with a burning sensation, numbness over the palatal mucosa and pain as a result of pressure on the nasopalatine nerves.

Discharge from the cyst may cause salty taste in mouth and upon infection may cause a purulent or foul taste [14]. The vitality of adjacent teeth remains unaffected. Since the location of NPDC and its less frequent occurrence as compared to radicular cyst it is often clinically misinterpreted as radicular cyst and several times endodontic therapy of the incisors has been observed [13]. A rare case of bilateral nasopalatine duct cysts has been reported by Ciccio, *et al.* in a 35 year old black man [16].

Radiologically, NPDCs appear as a well-defined, rounded or heart-shaped radiolucency in between two incisors. Superimposition of the anterior nasal spine causes the heart-shape representation in radiograph. CT scan reveals a midline located smooth expansion with sclerotic margins. T1W1 images (MRI) of NPDC depicts a high signal intensity owing to presence of keratin and viscous fluids [13].

Histologically, NPDC may show squamous, columnar, cuboidal or pseudostratified lined epithelium. Peripheral nerves, arteries and veins, mucous glands are also observed which are native to this region [3].

The differential diagnosis for NPDC is radicular cyst, dentigerous cyst associated with a supernumerary tooth (mesiodens), central giant cell granuloma, keratocystic odontogenic tumor and nasoalveolar cyst [16].

Enucleation is the treatment of choice as epithelial lining of longstanding NPDC may undergo metaplastic changes resulting in squamous cell carcinomas, so early intervention and surgical removal is done with the aim of reducing the risks and complications [14]. Another rare complication after a surgery which can be observed only in 10% of cases is the paresthesia of the anterior part of the palate [13]. Recurrence is infrequent and has been reported in 11% of the patients [17].

Conclusion

Nasopalatine duct cyst is a common nonodontogenic cyst which is commonly misdiagnosed as radicular cyst because the site of location of the cyst and radicular cyst being more common in occurrence as compared to nasopalatine duct cyst. Clinical and radiographic expertise of the clinician is must for proper diagnosis. Surgical enucleation is the gold standard treatment for nasopalatine duct cyst.

Conflict of Interest

None.

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