wjpmr, 2017,3(11), 259-265

SJIF Impact Factor: 4.103

WORLD JOURNAL OF PHARMACEUTICAL AND MEDICAL RESEARCH

www.wjpmr.com

Case Report

ISSN 2455-3301

WJPMR

DENTIGEROUS CYST ASSOCIATED WITH IMPACTED SUPERNUMERARY TOOTH

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Article Received on 20/10/2017 **Article Revised on 10/11/2017** **Article Accepted on 01/12/2017**

ABSTRACT [118]

Dentigerous cysts [DC] are the most common type of developmental odontogenic cysts arising from the crown of impacted, embedded, or unerupted teeth, but less common with supernumerary teeth. They are most commonly associated with third molars and maxillary canines. 5% of dentigerous cysts are reported to be involved with supernumerary teeth, where mesiodens is common. This case report describes dentigerous cyst associated with an impacted mesiodens in a patient with swelling in the maxillary anterior region. On radiographic examination well defined radiolucency was seen surrounding impacted mesiodens. Enucleation of the cyst was done along with the removal of the mesiodens. The final diagnosis of dentigerous cyst associated with an impacted mesiodens was confirmed with the help of histopathological examination.

KEYWORDS: Dentigerous cyst, anterior maxilla,supernumerary teeth, mesiodens.

INTRODUCTION [340]

In 1853 Paget coined the term "Dentigerous cyst"[DC]. They are the most common type of developmental odontogenic cysts which arise from the crown of impacted, unerupted or embedded, teeth.[1] They are the second most common cystic lesion of the jaws, after radicular cysts. The word dentigerous means "tooth bearing". They are mainly associated with the crowns of permanent teeth; however, few rare cases are reported in association with the crowns of deciduous teeth, along with complex odontoma, and supernumerary teeth. The association of dentigerous cysts with supernumerary teeth constitutes about 5-6% of all dentigerous cysts and about 90% are associated with a maxillary mesiodens.[2]

Dentigerous cysts occur due to expansion of dental follicles which results from accumulation of fluid between tooth crowns and epithelial components. Occurrence of this cyst[3] ranges from 10 to 30 years of ages with a greater incidence in males having a ratio of 1.6:1. The impacted mandibular third molars are most commonly involved teeth with this cyst, followed by maxillary canines, mandibular premolars, and occasionally supernumerary teeth or odontomas. Stafne first described the association of dentigerous cysts with supernumerary teeth with the incidence of 5.5% out of 200 supernumerary teeth.[4]

The anterior maxillary region is the most common site for supernumerary teeth. The supernumerary tooth which appears in the maxillary midline is known as mesiodens due to its position in the center of the maxilla. On clinical examination a missing tooth or teeth and occasionally a hard swelling, which sometimes may result in facial asymmetry and possible pathologic fracture can be observed. On routine dental radiographs Dentigerous cysts are initially diagnosed. Radiographically, they appear as a unilocular radiolucent area with a well-defined sclerotic border surrounding the crown of an unerupted tooth, but an infected DC will show ill-defined borders[5]. In the present study Medline search was made from 1988 to 2016 using key words like dentigerous cyst, supernumerary teeth and mesiodens. This literature search of 28 years was mainly focused on dentigerous cyst associated with supernumerary teeth in the anterior maxillary region describing the present case.

CASE REPORT: [2148]

A 23 year old male patient had reported to the department of Oral Pathology and Microbiology with a chief complaint of painless swelling in the roof of the mouth since 1 year. Initially swelling was small and it gradually increased to the present size. Pain was not associated initially but after 6 months patient complained of swelling pain with mild loosening of the tooth structure in the upper front tooth region.There was no associated history of trauma and pain. The medical history was noncontributory.

Intraoral clinical examination revealed presence of a single diffuse swelling in the mid-palatal region of hard palate measuring approximately 1.5x 1 cm in size, extending anteriorly from gingival margins in the centre

of rugal region and about 2 cm from the gingival margins bilaterally. No surface discharge or ulceration was seen. The surface had a bluish white tinge. The swelling was smooth, pulsatile in nature, non-compressible, non-reducible, soft in consistency, tender on palpation with mild rise of temperature. A clinical provisional diagnosis of mid palatine cyst was made. [Figure 1].

Figure 1: A single diffuse swelling present in the mid-palatal region of hard palate with bluish tinge.

Radiological investigations included Intra Oral Periapical (IOPA), Occlusal, Ortho PentamoGraph (OPG).

Figure 2: IOPA: well-defined heart shaped or pear shaped unilocular radiolucency with 11, 21,12.

IOPA radiograph with respect to 11, 12, 21, 22 showed widening of periodontal ligament space in the periapical region of 11, 21, 12, with loss of lamina dura. A well-defined heart shaped or pear shaped unilocular radiolucency with hyperostotic borders of 2x3 cm in size present in the periapical region of 11, 12, 21, 22. A diffuse radiopacity in periapical region of 11 mimicking a tooth structure is present. Root of 11 is displaced distally. These features are suggestive of dentigerous cyst. [Figure 2].

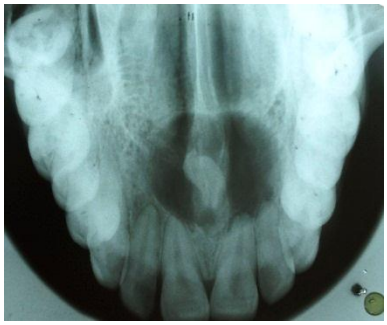


Figure 3: Occlusal Radiograph. Well-defined, Heart shaped, corticated, unilocular radiolucent lesion extending from 11 to 22 region.

Maxillary occlusal radiograph revealed a well-defined,Heart shaped, corticated, unilocular radiolucent lesion measuring approximately 2x2 cm extending from 11 to 22 region mediolaterally, and anteroposteriorly. It extended from the apices of the maxillary incisors to a few cm into the palate. Inverted impacted mesiodens was also seen in relation to the maxillary central incisors in the intermaxillary suture and naso-palatine foramen/canal region. The radiolucent lesion circumscribed the cementoenamel junction of the impacted tooth. Slight resorption of the roots of 11 and 21 was noticed [Figure 3].

Figure 4 OPG.

OPG revealed well-defined radiolucency in the periapical region extending from 12 11 21 22 measuring 3x2 cm in size with a distinct radiopacity in the mesial aspect of 11 in the periapical region was noted suggestive of dentigerous cyst.

Figure 5: NCCT PNS.

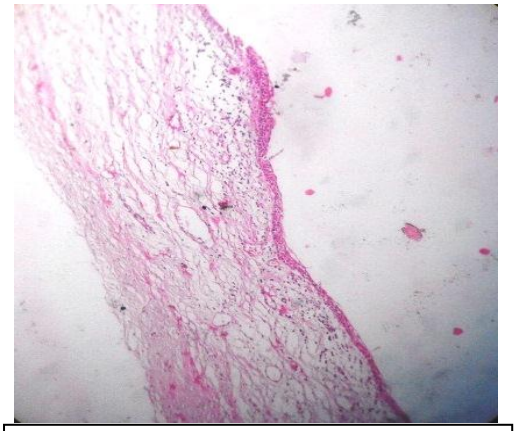
A Non-Contrast Computed Tomography (**NCCT**) scan of the Para Nasal Sinuses (**PNS**) revealed well defined oval hypodense lesion in alveolar process of maxilla 24x29 mm overlying the central incisors.

The overall radiological examination was suggestive of dentigerous cyst.

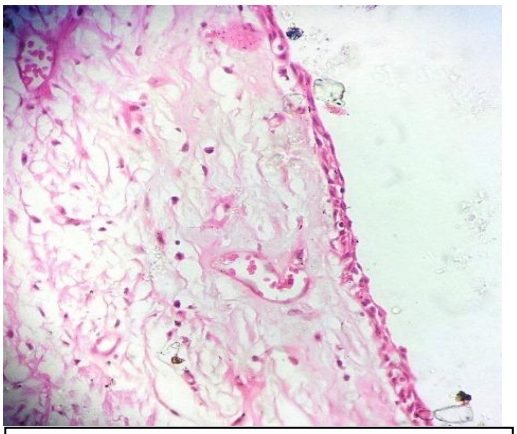
After the informed consent of the patient, an incisional biopsy was performed under local anesthesia and tissue was sent for histopathological evaluation.

Histopathology revealed odontogenic cystic lining along with connective tissue wall. The cystic lining consists of 2-4 layer thickness of nonkeratinized stratified squamous epithelium. The thin connective tissue wall consisting of young fibroblasts widely separated by stroma and ground substance resembling dental papilla and shows mild chronic inflammatory cell infiltrate with mild vascularity and areas of hemorrhage. Features are suggestive of dentigerous cyst [Figure 6a & 6b].

Dentigerous cyst was considered as the final diagnosis after correlating the history, clinical and radiographical features, coupled with histopathology. Cyst enucleation along with extraction of the supernumerary tooth was considered as the treatment plan; a surgical flap was raised in the maxillary anterior region, the labial cortex was raised and the cyst was exposed.



6a) Low power: Cystic lumen lined by epithelial lining which is supported by connective tissue capsule.



6b) High power: 2-4 layer thickness of nonkeratinized stratified squamous epithelium.

Figure 6: H and E examination.

Table I: Documented cases in the literature of dentigerous cysts associated with supernumerary teeth.

Case no.	Authors/Reference	Patient age(years)	Gender	Site (premaxilla region)	Symptoms	Treatment
1	Lustmann and Bodner (3) 1988	09	Female	Right central incisor	---	Enucleation
2	Lustmann and Bodner (3) 1988	12	Male	Left central and lateral incisor	--	Marsupialization
3	Lustmann and Bodner (3) 1988	37	Male	Entire premaxilla	Swelling, pain	Enucleation
4	Lustmann and Bodner (3) 1988	38	Male	Midline to the left, maxillarysecond premolar	Swelling	Enucleation
5	Lustmann and Bodner (3) 1988	68	Female	Midline to the left maxillarypremolar	Swelling, pain	Enucleation
6	Lustmann and Bodner (3) 1988	71	Female	Left premaxillary region	Asymptomatic	Enucleation
7	Awang and Siar (6) 1989	34	Male	Midline to the upper leftfirst premolar	Swelling Enucleation	Enucleation
8	Awang and Siar (6) 1989	24	Female	From the upper right canine to the upper left central incisor	Swelling	Enucleation

9	Scolozzi, et al (21) 2005	42	Male	Premaxilla	Swelling	Enucleation and cancellous bone graft from the iliac crest
10	AyakoItoh et al (11) 2006	10	Male	maxillary right anterior mucogingival fold.	Swelling	Enucleation along with removal of the impacted mesiodens,
11	Dinkar, et al (15) 2007	14	Female	Maxillary anterior region	Pain, swelling	Enucleation
12	Khan, et al (20) 2008	24	Male	Incisor region	Swelling	Enucleation
13	Gulses, et al (13) 2009	10	Male	Right central incisor	Asymptomatic	Enucleation
14	Kumar, et al (10) 2010	14	Male	Central incisor region	Swelling	Enucleation
15	John, et al (7) 2010	22	Male	From the maxillary right central incisor to the right canine	Swelling	Enucleation
16	John, et al (7) 2010	24	Male	Maxillary anterior region	Swelling	Enucleation
17	John, et al (7) 2010	46	Male	Maxillary anterior region	Pain, swelling	Enucleation
18	Qian Jiang,(5) 2011	55	Female	Central incisors	Swelling	Enucleation
19	Qian Jiang,(5) 2011	46	Male	Midline to the upper right canine	Swelling	Enucleation
20	Qian Jiang,(5) 2011	53	Male	From the maxillary right centralincisor to the right canine	Swelling	Enucleation
21	Qian Jiang at el,(5) 2011	23	Male	From the maxillary right first molar to the right central incisor	Swelling	------
22	NehaKhambete et al (6) 2012	55	Male	Maxillary anterior region	Pain and swelling	Enucleation along with removal of the impacted mesiodens,
23	NehaKhambete et al (6) 2012	46	Male	Maxillary anterior region	Pain and swelling	Enucleation along with removal of the impacted mesiodens,
24	Preeti Nair et al (7) 2013	30	Male	Maxillary anterior region	Pain and swelling	Enucleation
25	Dhir et al (9) 2013	--	--	Maxillary anterior region	Pain and swelling	Enucleation
26	Kaushal Mahendra Shah, et al (13) 2013	18	Male	Upper jaw	Swelling	Enucleation along with removal of the impacted mesiodens,
27	Beatriz González Navarro et al (3) 2015	23	Male	Upper right central incisor to upper left canine	Swelling	Enucleation along with extraction
28	Shamimul Hasan et al (4),2014	32	Male	Maxillary anterior region	Swelling	Enucleation along with extraction
29	John Hearty Deepak	32	Male	Maxillary right and	Pain ,	Enucleation

	et al (1) 2015			left central incisors	Swelling	along with extraction
30	Sk. Abdul Mahmud et al (2) 2015	56	Female	Maxillary right canine region to the left premolars	Swelling	Enucleation along with extraction
31	Sulabha A. Narsapur et al (10) 2015	40	Male	Palate anterior region	Swelling	Enucleation along with extraction
32	Andrade, et al.(12) 2016	28	Male	Maxillary anterior region	Pain and swelling	Enucleation along with removal of the impacted mesiodens,
33	Dr. Soumithran C.S (8) 2016	08	Male	Maxillary anterior region	Pain and swelling	Enucleation along with removal of the impacted mesiodens,
34	OUR STUDY CASE	23	Male	Maxillary right central incisor region to the left lateral incisor	Swelling	Enucleation along with extraction of mesiodens.

DISCUSSION

Supernumerary teeth are commonly present in the maxillary anterior region and can usually cause developmental and eruption disturbances of adjacent permanent teeth, leading to diastema, crowding, displacement, and in some cases, radicular resorption and dentigerous cyst formation.[4,6,7] Developmental abnormalities and hyperactivity of the dental plate is the most accepted theory in order to explain their formation.[8]

It is associated most frequently with mandibular third molars, followed by maxillary canines, mandibular second premolars and maxillary third molars.[9,10] It may also occur around unerupted supernumerary tooth; but rarely can be associated with primary dentition.[10] In present case, there is association of dentigerous cyst with impacted mesiodens.

Association of dentigerous cysts with supernumerary teeth is an unusual pathology, with the prevalence varying between 1% and 9.9%. According to some published reports, the frequency of impacted supernumerary teeth developing dentigerous cysts varied between 5.5% (Stafne) and 13% (Frietas et al.).[11] Mesiodens, having an overall prevalence of 0.15-1.9%,[12] when fail to erupt, is responsible for causing a sequelae of pathological situations like median diastema, dentigerous cyst and delayed eruption.[12]

Dentigerous cysts are the second most common odonto-genic cysts of the jaws after radicular cysts, whereas dentigerous cyst associated with a supernumerary tooth is a rare entity. A review of the literature since 1988 disclosed 33 reported cases of dentigerous cysts associated with premaxillary supernumerary teeth.[2,13,14,15,16,17,18,19] The present case brings the total number of documented cases to 34 (Table I). The age range for reported cases varies widely, from 9 to 71 years of age. Among the 34 cases summarized in Table I, the incidence is significantly higher in men (n=26) compared to women (n=7). This suggests that dentigerous cysts appear to have a distinct predilection for the male gender.[20,21]

Dentigerous cysts associated with supernumerary teeth in the premaxilla are easily diagnosed radiographically due to their radiopaque image. Panoramic and upper occlusal radiographs are easy and inexpensive methods, where the location of dentigerous cysts, the structure of the impacted teeth, the effect on adjacent teeth and causing the resorption of adjacent roots can be easily determined.[22] However, there are some limitations of radiograph films. To overcome these limitations, Computed Tomography [CT] is necessary and valuable to obtain more information about the lesion. In CT, not only pathology and the exact location of the impacted tooth is identified but also full extension of the lesion is identified, thus helping in proper treatment planning. CT can also be helpful in identifying bone erosion and adjacent soft tissues invasion. In the present case, the dentigerous cyst with the supernumerary tooth appeared to be within the anterior maxilla in the OPG or/and upper occlusal radiograph. For the exact location of the impacted tooth and the degree of bone destruction along with the extension of the lesion Non-Contrast Computed Tomography [NCCT] of Para Nasal Sinuses [PNS] was taken. Hence for exact location and extent of the lesion followed by appropriate treatment which was made based on accurate information regarding dentigerous cysts associated with supernumerary teeth. Hence,we

recommend panoramic radiograph and/or upper occlusal radiograph as basic diagnostic tools, with CT for further evaluation of the lesion.

The standard treatment protocol for a dentigerous cyst is enucleation along with extraction of the associated supernumerary tooth.[14,23] According to the documented cases in the literature, for the second case (Table I) marsupialization was done due to the close proximity of the lesion to the apices of the incisor teeth. Marsupialization is recommended for a large cyst where simple enucleation may not be effective and complete removal of the surrounding structure is not desirable.[24] Scolozzi et al recommended enucleation followed by an immediate bone grafting procedure for large cysts.[25] In the present case, enucleation was done with surgical removal of the impacted supernumerary tooth without using bone grafting.

A broad range of conditions may lead to a clinical presentation of painless swelling along the lingual surface of the palate or on the upper lip. Differential diagnosis of a radicular cyst, median palatine cyst, nasopalatine duct cyst, odontogenic keratocyst (OKC) or adenomatoid odontogenic tumor (AOT) was considered in the present case. Median palatine cysts and nasopalatine duct cysts are associated with vital teeth as non-odontogenic cysts of the hard palate.[25] Most radicular cysts exhibit round or pear-shaped, unilocular, lucent lesions in the periapical region. Associated tooth may have a deep restoration or large carious lesion radiographically.[25] Usually OKCs contain an impacted tooth, with the lumen containing 'cheesy' material and has a parakeratinized epithelium lining. They are more likely to show aggressive growth compared to the other odontogenic cysts and may have multilocular appearance with undulating borders on radiographic examination.[26] Approximately 75% of cases are associated with an unerupted tooth, and the most common location is in the anterior maxilla. AOTs are more common in young people with female predilection and most importantly, the radiolucency in cases of AOTs extends apically beyond the cementoenamel junction.[22]

CONCLUSION

In conclusion, dentigerous cysts arising from impacted supernumerary teeth in the anterior maxilla should be considered in the differential diagnosis for painless swelling along the lingual surface of the palate or on the upper lip. To prevent the development of a dentigerous cyst and to avoid unwanted effects on adjacent teeth, early detection consisting of a thorough clinical and radiographical examination is necessary for accurate diagnosis and proper treatment planning.

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