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Anterior Maxillary Dentigerous Cyst with Supernumerary Tooth- Case Series and Review of Literature

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Abstract

Dentigerous cysts are thought to be caused by a developmental abnormality derived from the reduced enamel epithelium of the tooth forming organ. Most typical dentigerous cysts are those associated with the third molar teeth of the mandible, but rarely involve impacted supernumerary teeth in the anterior maxilla. We hereby present a series of case reviews from 1997 to 2017 as well as two cases of enucleation and curettage dentigerous cyst of the anterior maxilla which we performed, which had a challenging surgical treatment on account of its close proximity to the vital structures and degree of bone perforation.

Keywords: *Dentigerous Cyst; Supernumerary Tooth*

Introduction

In 1974, Kramer defined cyst as "a pathologic cavity having fluid, semi fluid or gaseous contents and which is not created by accumulation of pus. Most cysts, but not all, are lined by epithelium".

Dentigerous Cyst is a developmental cyst of odontogenic origin, these cysts involve impacted, unerupted permanent teeth, supernumerary teeth, odontomas, and rarely deciduous teeth [1]. Dentigerous cysts around supernumerary teeth account for 5% of all dentigerous cysts, most developing around a mesiodens in the anterior maxilla. Treatment of these cysts ranges from marsupialization to enucleation and curettage [6].

We here by present 2 cases with Dentigerous cysts associated with impacted supernumerary teeth in the anterior maxilla and a literature review of similar cases reported from 1997 till 2018.

Case Report

Case Report 1

A 34 -year old male patient reported to the department of Oral and Maxillofacial surgery at our tertiary center, with the chief complaint of painless swelling in the palatal region since 3 - 4 months with no history of trauma. Examination of the swelling revealed its extension from 12 to 22 region, measuring approximately 3 × 2 × 2 cm in size (Figure 1). It was firm and non-tender on palpation; and adjacent teeth were immobile. Occlusal radiograph revealed an unilocular, well defined radiolucency, circumferentially involving a supernumerary tooth (Figure 2 and 3). On electric pulp testing 12, 11, 21 and 22 were found to be non-vital. Aspiration of the swelling yielded straw-colored viscous fluid. Based upon the clinical and radiographic features a provisional diagnosis of dentigerous cyst associated with supernumerary tooth was made. Prior to surgical phase, routine blood investigations were carried out, which were within normal limits. Endodontic treatment was planned with 12, 11, 21 and 22 followed by surgical enucleation and curettage under general anesthesia following all aseptic precautions.

Figure 1: *Pre-operative palatal view.*

Figure 2: *Pre-operative IOPA.*

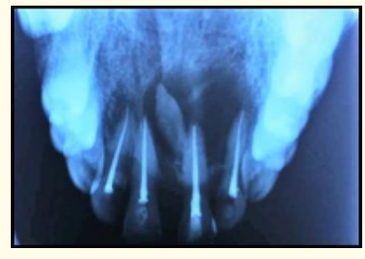


Figure 3: *Pre-operative maxillary occlusal radiograph showing impacted supernumerary tooth surrounded by ill-defined radiolucency.*

Procedure

Patient was taken under General Anesthesia and prepared as per standard surgical protocol. 2% lignocaine with adrenaline was used for local infiltration. Full thickness mucoperiosteal flap from 13 to 23 with bilateral vertical releasing incisions was raised (Figure 4). The cyst was de-roofed and enucleated with extraction of mesiodens followed by curettage (Figure 5). Hemostasis was achieved. Sharp bony edges were smoothened. Apicoectomy was performed with 12, 11, 21 and 22 followed by retro grade filling with MTA. Cavity was packed with DFDB graft and absorbable gelatin sponge and flap was repositioned and sutured with 3-0 black silk sutures (Figure 6). Patient was followed up regularly for more than a year with satisfactory healing and no recurrence.

Case Report 2

A 44-year old male patient reported to our department with a complaint of a painless palatal swelling. History suggested that the swelling was asymptomatic and gradually increased in size over a period of 6 - 7 months to its current size with no history of trauma. On examination a 3 × 2.5 × 2 cm, well-defined, ovoid swelling was seen extending from the mesial aspect of 21 to the mesial aspect of 24 with normal and intact mucosa overlying the swelling (Figure 7). On palpation, the swelling was non-fluctuant, non-tender and firm, mobility of adjacent teeth absent. Aspiration of the swelling yielded straw-colored viscous fluid. On CBCT evaluation, a single, ovoid radiolucency

Figure 4: *Incision marked with surgical ink.*

Figure 5: *Extraction of impacted supernumerary tooth and enucleation of cystic lesion.*

Figure 6: *Placement of DFDB graft.*

with well-defined sclerotic borders was seen encasing a tooth-like radiopaque structure within itself in relation to the roots of 21, 22, 23 extending close to the floor of the nasal cavity (Figure 8-10). The roots of the involved teeth were not resorbed. A provisional diagnosis of Dentigerous cyst associated with an impacted supernumerary tooth was made. Prior to surgical phase, routine blood investigations were carried out and found to be within normal limits. Endodontic treatment was planned with 21, 22, 23, 24 followed by surgical enucleation and curettage of cystic lesion under general anesthesia.

Figure 7: *Palatal swelling.*

Figure 8-10 : *CBCT image showing a single, ovoid radiolucency with well-defined sclerotic borders seen encasing a tooth-like radiopaque structure within itself in relation to the roots of 21, 22, 23 extending close to the floor of the nasal cavity.*

Procedure

Patient was taken under General Anesthesia following all aseptic protocols. Local infiltration using 2% Lignocaine with 1:200000 Adrenaline was administered. A crevicular incision with two vertical releasing incisions were made and a full thickness mucoperiosteal flap was raised (Figure 11 and 12). The cyst was carefully enucleated along with the impacted supernumerary tooth (Figure 13). Curettage of the bony cavity was carried out, hemostasis was achieved (Figure 14) followed by apicoectomy and retrograde filling with MTA with 21, 22, 23, 24. The bony cavity was packed with DFDB graft and resorbable gelatin sponge. The flap was repositioned and sutured with 3-0 Vicryl (Figure 15). Patient was discharged and followed up regularly for a period of 1 year with satisfactory healing and no recurrence.

Figure 11 and 12: *A crevicular incision with two vertical releasing incisions were made and a full thickness mucoperiosteal flap was raised.*

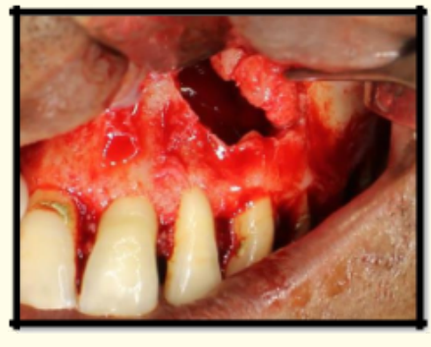


Figure 13: *The cyst was carefully enucleated along with the impacted supernumerary tooth.*

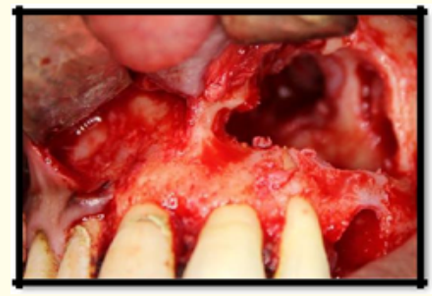
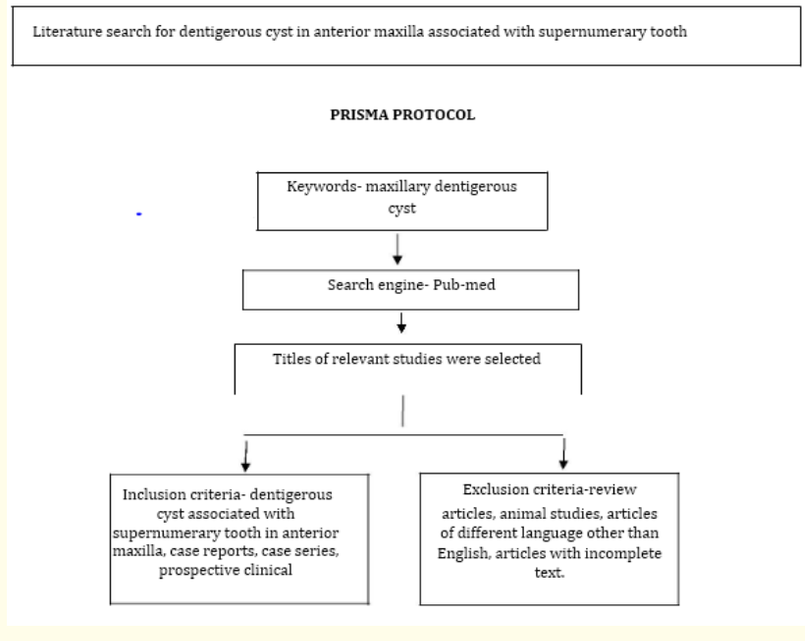


Figure 14: *Curettage of the bony cavity was carried out, hemostasis was achieved. reoperative photograph.*

Figure 15: *The flap was repositioned and sutured with 3-0 Vicryl.*

Review of Literature

The PRISMA protocol was followed for the review. Search engines and medical databases Pub-med were tapped for information related to the subject. The search word Maxillary Dentigerous Cyst" was employed for retrieval of data. An analysis of the treatment modalities, follow up periods and proclaimed success rates was done. A total of 331articles related to maxillary dentigerous cysts were found on Pub-med search from 1997 to 2018. Out of these 331 articles, 59 articles described about anterior maxillary dentigerous cyst. Among these 59 articles (maxillary anterior region) dentigerous cyst associated with supernumerary teeth were described in 20 articles. Based on the inclusion and exclusion criteria 14 articles were studied for review (Refer Table 1).



Sr no	Author	Study design	No. of patients	Age/ gender	Surgical procedure	Complications	Follow-up
1	Nascimento RD., *et al.* 2015	Case report	1	8 yr	Decompression, surgical extraction of supernumerary tooth and orthodontic traction of permanent canine	No complications	5yrs
2	Shamimul Hasan., *et al.* 2014	Care report	1	32 yr/male	Cyst enucleation and extraction of supernumerary tooth	No complications	6 months
3	Ashwini Ramakrishna., *et al* 2013	Case report	1	10 yr/male	Cyst enucleation	Sinus wall perforation intraoperatively	6 months
4	Navarro BG., *et al.*	Case report	1	-	Cyst enucleation, apicectomy and retrograde filling of affected teeth. Cavity filled with xenograft	No complications	-
5	Kiran Patel., *et al.*	Case report	1	30 yr/male	Cyst enucleation, apicectomy and retrograde filling of affected teeth. Cavity filled with hydroxyapatite crystals	No complications	-
6	Karthik Rajaram mohan., *et al.*	Case report	1	32 yr/male	Cyst enucleation	No complications	-
7	Kaushal Mahendra shah., *et al.*	Case report	1	18 yr/male	Endodontic treatment of affected teeth and cyst enucleation	No complications	6 months
8	Neha Khambete., *et al.*	Case report	2	a) 55 yr/male b) 46 yr/male	Cyst enucleation for both cases	No complication for both cases	1 yr
9	Neeraj k Aggarwal., *et al.*	Case report	1	11 yr/male	Cyst enucleation	No complications	9 months
10	Qian jiang., *et al.*	Case series	4	55 yr/female 46 yr/male 53 yr/male 23 yr/male	Cyst enucleation in all cases except the last case as surgery was not performed due to low hemoglobin level	No complications	-
11	Ritesh Kalaskar., *et al.* 2011	Case reports	2	12 yr/male 12 yr/male	Cyst enucleation and orthodontic extrusion of permanent tooth in both cases	No complications	-
12	Vosough Hosseini., *et al.* 2011	Case report	1	18 yr/female	Cyst enucleation	No complications	6 months
13	Deepak Sharma., *et al.* 2010	Case report	1	12 yr/male	Cyst enucleation	No complications	1 year
14	Khan MH., *et al.* 2008	Case report	1	24 yr/male	Cyst enucleation	No complications	-

Table 1: *Review from 1997-2017.*

Materials and Methods

In our institutional experience of last 10years, we have treated 16 other cases of Dentigerous cyst associated with supernumerary teeth in anterior maxilla accompanied with palatal swelling. Enucleation and curettage of the cystic lesion with extraction of involved supernumerary tooth was carried out in all cases (Refer Table 2).

Case No	Male/ Female	Age	Palatal Bone Perforation	Bone Graft	Follow-Up
1	M	20	Present	DFDB bone	5 yrs
2	M	19	Absent	_	3 yrs
3	F	23	Present	DFDB bone	5 yrs
4	F	24	Absent	_	1 yr
5	M	30	Present	DFDB bone	8 yrs
6	F	20	Present	DFDB bone	2 yrs
7	M	44	Present	_	1 yr
8	M	19	Present	DFDB bone	1 yr
9	M	30	Absent	_	7 yrs
10	M	27	Present	_	4 yrs
11	M	29	Absent	DFDB bone	5 yrs
12	F	19	Present	DFDB bone	8 yrs
13	F	29	Present	DFDB bone	4 yrs
14	F	31	Present	_	3 yrs
15	M	34	Absent	DFDB bone	1 yr
16	M	44	Present	_	1 yr

Table 2: *Cases treated in our institute.*

Discussion

Dentigerous cyst is a developmental, epithelium- lined cyst of odontogenic origin, accounting for about 20% of the cysts of jaws and second most common in occurrence after radicular cyst [7]. Toller stated that the likely origin of the Dentigerous cyst was the breakdown of proliferating cells of the follicle after impeded eruption [9]. Bloch suggested that from the overlying necrotic deciduous tooth, periapical inflammation spreads to involve the follicle of the unerupted permanent successor; an inflammatory exudate ensues and results in the formation of a cyst [10]. Dentigerous cyst most frequently occurs in patients between 10 and 30 years of ages and there is a slight male predilection, with a 1.6:1, M:F ratio [7]. The cysts most often involve impacted mandibular third molars, followed by maxillary canines, mandibular premolars, and occasionally supernumerary teeth or odontomas [7]. Only 5% of dentigerous cysts are associated with supernumerary teeth [11]. Most dentigerous cyst are solitary, dentigerous cysts which are multiple or bilateral are usually associated with syndromes such as cleidocranial dysplasia, Maroteaux-Lamy syndrome and Gorlin Goltz syndrome. All of our patients were non-syndromic and were diagnosed with dentigerous cyst associated with supernumerary tooth in anterior maxilla.

A Supernumerary tooth is formed due to disruption in the process of odontogenesis. This may occur due to the splitting of the enamel organ or from an uncoordinated cell proliferation. Hyperactivity of dental lamina, genetic mutation, dichotomy or environmental factors may also play a contributory role [24,25]. Supernumerary teeth may lead to impaction of adjacent teeth, crowding, spacing, displacement and rotation of teeth, occlusal interferences, caries, periodontal problems, mastication problems, and esthetic concerns [24,26]. Most common pathologic lesion associated with supernumerary teeth is the formation of a dentigerous cyst. Radiographically, the dentigerous cyst shows a well-defined unilocular radiolucency with sclerotic borders that is associated with the crown of an unerupted tooth, although, once infected, it may show ill-defined borders. The cyst-to-crown relationship shows several radiographic variations such as central, lateral and circumferential33. About 90% of dentigerous cysts from supernumerary teeth develop around a mesiodens in the anterior maxilla and they present as palatal swellings [3].

Clinically dentigerous cyst in anterior maxilla may be small, asymptomatic or may be large and symptomatic involving nasal floor, maxillary sinus associated with symptoms such as upper lip swelling [12], sino-nasal and orbital symptoms [15], epiphora and nasal obstruction [14], nasolacrimal duct obstruction [13]. All of our cases showed cyst located palatally. Palatal bone perforation with an intact palatal mucosa was seen in 11 cases and only one case showed nasal floor perforation with intact nasal mucosa.

Treatment of dentigerous cyst with anterior maxilla is challenging due to close association of the cyst with nasal floor, vital adjacent teeth in the esthetic zone and high vascularity. Treatment varies from marsupialization to enucleation. Dentigerous cyst associated with supernumerary teeth are usually treated with enucleation of cyst along with the extraction of the supernumerary tooth [3-6,11]. Maltoni., et al. and Nascimento RD suggested that marsupialization is usually preferred in cases where cyst is extensive, in close proximity with vital structures or when permanent teeth involved can be aligned with orthodontic treatment planning [16,17]. However, in none of our cases, we encountered any of these conditions, so the preferred line of treatment was enucleation and curettage. Kaushal Mahendra Shah., et al. and Kalaskar., et al. has discussed about need of orthodontic treatment required for impacted permanent teeth displaced due dentigerous cyst associated with supernumerary tooth [22,23]. In none of our cases there was displacement of adjacent teeth hence orthodontic treatment was not required.

We could salvage adjacent teeth in 10 of our cases with endodontic treatment, apicoectomy followed by a retrograde filling with MTA since these patients were relatively young, with good oral hygiene and had a healthy periodontium. In 6 cases, extraction of adjacent teeth was unavoidable due to external root resorption and poor periodontal status. In our opinion, teeth involved with cyst should be extracted to prevent recurrence. Hence, we performed enucleation and curettage with extraction of impacted supernumerary tooth in all our cases.

Scolozzi., *et al.* recommended enucleation followed by an immediate bone grafting procedure [12]. In 9 of our cases we carried out a surgical removal of the impacted supernumerary tooth and cyst enucleation with DFDB graft with gelatin sponge were used to allow faster healing and better bone-fill and in the 7 cases, we placed only resorbable gelatin sponge to eliminate dead space and to support the palatal mucosa. There is a certain degree of controversy as to whether the remaining cavity should be filled or not with bone grafts. Some authors are against filling the cavity and argue that, with the blood clot, it is possible to regenerate bone, also reducing the chances of encountering infections during treatment. In other cases, authors recommend using bone grafts to fill the cavity, whether with autologous grafts, allografts, or xenografts [31]. We used allograft in 9 of our cases and in the follow-up period we noticed no significant difference between bone filled with the graft and without the graft on radiograph after one year. Other authors state that the cystic cavity should not be filled, except in cases where the lesion is small in size and in patient where bone regeneration potential is compromised.

Mc Donald., *et al*. [7,10], in their studies state that dentigerous cysts do not recur after complete excision. MHK., et al. studied the management of extensive dentigerous cysts during 11-year period where 40 cases of extensive (involving three or more teeth) dentigerous cysts of the maxilla and mandible were studied and none of the cases showed recurrence. Our cases did not show any recurrence.

Potential sequelae of dentigerous cyst includes its transformation to squamous cell carcinoma, mucoepidermoid carcinoma or ameloblastoma [28-30]. None of our cases showed any of these complications.

Conclusion

Based on the cases managed in our institute and the literature that has been reviewed, we can conclude the following: The occurrence of a dentigerous cyst in the anterior maxilla associated with a supernumerary tooth is infrequent and the treatment is challenging due to the close association of the cyst with vital anatomical structures such as nasal floor and vascularized area around upper lip and nose. Enucleation of the dentigerous cyst followed by thorough curettage along with extraction of supernumerary teeth is a must line of treatment to avoid further complications and recurrence.

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