CASE SERIES OF DENTIGEROUS CYST WITH RARE ASSOCIATION OF MAXILLARY PREMOLAR, MAXILLARY LATERAL INCISOR AND MANDIBULAR CANINE

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ABSTRACT

Dentigerous cyst is the most frequently encountered odontogenic cyst which is epithelial lined and is developmental in origin. It usually affects young males below 30 years of age. Thus, its early diagnosis and treatment with rehabilitation becomes very important. Here a series of three cases with rare involvement of impacted maxillary lateral incisor, maxillary premolar and mandibular canine is presented with the discussion regarding clinical, radiological, histological features and treatment with a short review of literature.

Keywords: Rare, Maxillary premolar, Maxillary lateral incisor, Mandibular canine, Dentigerous cyst, Case series.

INTRODUCTION

Cyst is a pathological cavity having fluid, semifluid or gaseous contents, may or may not be lined by epithelium [1]. Odontogenic cyst derives its epithelium from odontogenic apparatus which is derived from basal epithelium of the stomodeum [2]. Dentigerous cyst (DC) is the most common developmental true odontogenic cyst and 2nd most common odontogenic cyst after radicular cyst. The literal meaning of dentigerous is ‘tooth bearing’ and it encloses the crown of an unerupted or impacted tooth, cyst lining being attached to the neck of the tooth at cemento enamel junction [3]. It usually occurs in young male patients involving mostly impacted mandibular third molars followed by maxillary permanent canine, mandibular premolars and maxillary third molar. Few cysts are associated with maxillary incisors, maxillary premolars and supernumerary teeth [4]. It is usually asymptomatic unless secondarily infected. Patient often reports with the complaint of either slow growing swelling with mild discomfort, retained deciduous teeth, missing permanent teeth or gradual malalignment of teeth in localized area. It usually follows a gradual course of increase in size though can attain a large size before they are diagnosed and is often found on routine radiographic examination. Diagnosis is made after correlating clinical, radiographic and surgical findings. Histopathology is done to confirm the diagnosis and to rule out the possibility of other odontogenic cysts and any malignant changes if any. Here we are reporting series of three cases of DC, which showed rare association with maxillary 1st premolar, maxillary lateral incisor and mandibular canine.

Case 1

A 14 year old female reported with a gradually increasing swelling over right side face since 1 year. On examination extensive swelling of about 5cm x 5cm x 4cm was present on right side mid face causing asymmetry. The swelling was extending superio-inferiorly from right infraorbital margin till base of right nostril and mediolaterally from right side of nose till cheek prominence. Right palpebromalar sulcus, alar nasal and nasolabial sulcus were completely obliterated with shifting of right nostril, nasal septum and upper lip...
toward left side. Overlying skin was normal in color [Fig. 1.1, 1.2].

Swelling was slightly tender and compressible on palpation. Intraorally, swelling was present in right buccal vestibule extending from 21 to 16 completely obliterating the sulcus. Right side palatal swelling was also noted. Overlying mucosa was normal in color. 11 was rotated with midline diastema. 52, 53 were retained and 12, 13 were missing [Fig. 1.3]. Buccal cortex and palatal expansion was present. No complaint of epiphora or epistaxis was present.

Radiographic examination revealed a large unilocular ovoid shaped radiolucency with well-defined borders involving right side maxilla extending from apices of 11, 52, 53, 14, 15, 16 till right side infraorbital margin with impacted 12 and 13. 13 was drifted to the right side infraorbital margin and 12 towards right side nasal septum medial to the right infraorbital margin. Root apices of 52 and 53 were resorbed and drifting of apices of 11, 14, 15 was also noted [Fig. 1.4].

Noncontrast computed tomography revealed expansile, unilocular radiolucency with respect to right maxilla with well-defined margins and impacted 12, 13 [Fig.1.5, 1.6]. On aspiration straw colored fluid was present. A provisional diagnosis of DC was rendered.

The cyst was enucleated along with 12, 13, 52 and 53 was submitted for histopathological examination. Post-operative healing was uneventful and symmetry of the face could be achieved to a larger extent [Fig.1.7]. Histopathology and macroscopy confirmed the diagnosis of DC.

Histopathology revealed cyst lined by thin non keratinized stratified squamous epithelium with scattered few inflammatory cells in the fibrous connective tissue.
Case 2

A 13 year old male child reported with a swelling on right side face since 1 year. On examination swelling was present over right side midface over maxillary region. Overlying skin was normal in color [Figure 2.1, 2.2]. Swelling was hard, non-tender and had gradually increased to the present size without causing any pain.

Intraorally, right buccal vestibule was obliterated and swelling extended from 11 till 15 with normal overlying mucosa. 53, 54, 63, 64, 65, 73, 74, 75, 83, 84, 85 were retained [Figure 2.3]. Panorex view revealed a unilocular radiolucency with regular defined borders, present on right side maxilla extending from the apices of 53, 54 till hard palate. 13, 14 were impacted, shifted superiorly and 14 was seen in the lumen of radiolucency. Drifting of apices of 12 and 15 were noted [Figure 2.4].

Maxillary occlusal view revealed expansion and thinning of buccal cortical plate [Figure 2.5]. In this case cyst enucleation along with extraction of 53 and 54 was done. Orthodontic treatment was done to guide the eruption of 13 and 14 along with the straightening of 15.

Case 3

An 11 year old male child reported with slight swelling in left lower jaw and malpositioning of lower teeth since 6 months. On examination the face was symmetric with no extra oral swelling. Intraorally, swelling was barely appreciable in the region of 31, 32 and 73. Overlying mucosa was normal in color. 73 was retained and 32 was buccally rotated [Figure 3.1]. On palpation, there was slight expansion of buccal and lingual cortex with respect to 31, 32 and 73. Panoramic view revealed well defined, unilocular radiolucency with regular borders at the peri apices of 42, 41, 31, 32, 73, 33 extending till mandibular alveolus. Drifting of roots of 31, 32, 41, 42 was noted. 33 was impacted with the crown surrounded by the radiolucent lesion [Figure 3.2, 3.3].

Considering the age, clinical presentation and radiographic features, a provisional diagnosis of central type DC was rendered with differential diagnosis of odontogenic keratocyst, radicular cyst, ameloblastic fibroma and unilocular ameloblastoma. The cyst lining was enucleated along with extraction of 73. Considering the young age, orthodontic treatment was planned to guide the eruption of canine along with alignment of mandibular anteriors. Histopathology revealed cyst lined by thin non keratinized stratified squamous epithelium with underlying connective tissue having few scattered inflammatory cells, thin fibers and vascular spaces. Gross specimen along with histology confirmed the diagnosis of DC.
Dentigerous cyst is a developmental odontogenic cyst occurs due to accumulation of fluid between reduced enamel epithelium and tooth enamel surface [5]. It is attached to the cemento enamel junction of the involved impacted tooth. Frequency of dentigerous cyst among odontogenic cysts range from 17.1% to 45% [4, 6-8]. Though, it can occur in any age group but mostly affects patients in first decade showing peak in second and third decade with gradual decline as the age progresses. Recently a male case as young as one year of age is being reported with dentigerous cyst associated with permanent mandibular first molar [9]. It affects males more significantly as compared to females varying from 1.6-1.8:1 [8-13].

In above mentioned cases all the three were in their early second decade and two were males and one was female.

Anatomically cyst is found in highest frequency in relation to mandibular third molar followed by maxillary permanent canine, mandibular premolars and maxillary third molar. Maxillary first premolar, maxillary incisors and supernumerary teeth are involved in very few cases [4, 8, 13, 14]. Out of three cases reported, two cases showed rare association with maxillary lateral incisor and maxillary first premolar. de Andrade Freitas Oliveira reported DC involving maxillary lateral incisor in a 3 year old male patient [15]. Rohilla et al., reported a 10 yr old male presenting with DC associated with maxillary central, lateral incisor and canine [16].

Third case showed association with mandibular canine which is also not very frequently reported.

Relative risk of development of DC in impacted teeth is more in mandibular third molar when compared to maxillary counterpart. Impacted maxillary canine have more relative risk of development of cyst than mandibular counterpart. Frequency of development of DC among impacted teeth varies from 1/150 to 1.44/100 [17, 18].

DC is usually asymptomatic unless secondarily infected. Patient often reports with the complaint of either slow growing swelling with mild discomfort, retained deciduous teeth, missing permanent teeth or gradual malalignment of teeth in localized area. It usually follows a gradual course of increase in size though can attain a large size before they are diagnosed. Two cases reported with the chief complaint of gradual swelling with retained primary teeth and missing permanent teeth. None of the reported case presented with severe pain and only slight discomfort was present.

DC is often found on routine radiographic examination as a well-defined unilocular radiolucency with sclerotic borders associated with unerupted teeth. Position of the impacted tooth and its association with radiolucent lesion varies in different cases. In central type, crown is enveloped symmetrically and may be pushed away from its direction of eruption. Lateral variant results from dilatation of the follicle on one aspect of the crown, commonly seen with partially erupted or impacted mandibular third molar. Circumferential type is, in which entire tooth appears to be enveloped by cyst [17,19]. All the three cases showed well defined radiolucencies with regular sclerotic borders. Two of them showed multiple teeth involvement with circumferential radiolucency and pushing of tooth superiorly away from eruption path. One case showed central type of radiolucency.

Radicular resorption of adjacent teeth is frequently seen and is suggested to be due to its origin from dental follicle which is known for resorption of primary teeth apices [20]. It was seen in all the three cases.

Though this cyst is considered to be developmental in origin, there are many theories in view of its origin, development and expansion.

Atkinson through various experiments concluded its origin due to degeneration in enamel organ, but Shear believes its formation in humans due to accumulation of fluid between reduced enamel epithelium and tooth enamel surface [5, 21-23].

Another theory of its origin proposes the rare eruption of permanent tooth into a developing radicular cyst around its deciduous predecessor [24].

Few authors found some evidence of an inflammatory etiology in the pathogenesis of some DC [25-27].
Main suggested the separation of follicle from crown due to increased hydrostatic pressure caused due to obstruction of venous outflow thus, supporting the haemodynamic concept of DC enlargement linked with dysfunctional eruptive forces [28, 29].

Few authors were in view of expansion due to accumulation of fluid by exudation through vessels in capsule [30, 31]. Skaug and Hofstad, Smith et al., suggested the diffusion of glycosaminoglycans in the cyst fluid to have an important role in expansile cyst growth by increasing the osmolality of the cyst fluid and hence raising the internal hydrostatic pressure of the cyst [32, 33].

Many pathologies like keratinizing cystic odontogenic tumor, unilocular ameloblastoma and early adenomatoid odontogenic tumor may mimic dentigerous cyst on gross specimen. Therefore, examination of gross specimen for any irregularity or nodularity on the cyst lining and its attachment on the neck of the tooth should be done thoroughly and the sections should be prepared from the same [5].

Histologically, cyst shows a thin fibrous wall with young fibroblast widely separated by stroma with overlying thin stratified squamous epithelium of 2–4 cell layers of flat or cuboidal cells. Epithelial hyperplasia and intense infiltration of inflammatory cells can be seen in infected cysts. Mucous cell may often be found in the epithelial lining specially in old age patients [34]. Presence of ciliated, sebaceous cells and hyaline bodies are found occasionally [35-37]. Present cases showed a thin epithelial lining with underlying connective tissue with few scattered inflammatory cells.

Complication of long standing DC may include development of malignant changes, ameloblastoma, mucoepidermoid carcinoma [38]. Bodner has analysed 116 reported case of Primary intraosseous squamous cell carcinoma (PIOSCC) in 2010 [39]. Zapala-Pospiech in 2013 reported a 66 year old male patient developing PIOSCC in long standing untreated DC in left hemimandible [40].

DCs can co-exist with other more serious conditions, such as odontogenic keratocyst or cystic ameloblastoma. Zhang et al, reported the first series of cases presenting clinically as bilateral DCs, but histologically as an odontogenic tumour or another type of odontogenic cyst. This association with more significant conditions reinforced the importance of histologically confirming any jaw cyst, even if it presents clinically as a classic DC [41].

DCs are treated either by complete enucleation along with extraction of the involved tooth/teeth or enucleation/ marsupialization of cyst with preservation of the impacted tooth guiding its eruption path. Treatment plan depends on the eruption potential of teeth; position, angulation, maturity/formation of root; extensiveness of the cyst, age of the patient, usefulness of tooth and the available space for eruption [42, 43]. Regular follow up is a requisite for both the treatment plans.

In the present case series, in one case cyst was fully enucleated along with the extraction of primary and involved permanent teeth followed by prosthetic rehabilitation, because of its location near maxillary sinus, thinning of bone, age and superior position of involved teeth. In other two cases, enucleation of cyst wall along with removal of primary teeth was done. Guidance to permanent teeth to erupt was planned considering the young age, incomplete root formation, availability of space. All the three cases are under regular follow ups in the concerned departments for the assessment of progress of resolution and development of complication if any.

CONCLUSION

Though DCs are benign, commonly encountered, asymptomatic cyst but considering the age it affects, its potential to gain large size leading to facial asymmetry, loss of esthetics, impacted teeth, thinning of bones with a probability to cause pathological fractures, diagnosis and treatment should be planned at the earliest. Importance of long follow up, careful examination of gross specimens and histopathological sections is a need of hour to avoid complications in long run, which may cause significant morbidity.

REFERENCES