



Research Paper

Maxillary Dentigerous Cyst: A Case Report and Brief Review of Literature

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ABSTRACT

Dentigerous cysts have been reported to make up around 24% of all the cysts occurring in the oral cavity. Frequently associated with unerupted tooth follicles, the exact histogenesis and origins of cysts still remains a topic of research. This cyst is known to occur mainly in the second to third decade of life with a predilection for males. The occurrence of Dentigerous cysts in association with premolars is believed to be less than 2.4%. In the maxilla Dentigerous cysts are noted to occur more commonly in association with unerupted or impacted canines. The following article attempts to review some key facts pertaining to the literature of this cyst and report a case of Maxillary Dentigerous cyst associated with a maxillary premolar.

Keywords:

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I. INTRODUCTION

One of the first documented cases of Dentigerous cyst was reported in 1847 and back then it was reported as a serous cyst/ osseous cyst/ distended capsule based more heavily on the clinical features that could be noted.¹ Today after nearly two centuries of one of the first few documented cases, Dentigerous cysts have been known to account for approximately 24% of all the cysts in the oral cavity.² Of all the cysts that have been reported in literature Odontogenic cysts make for the majority of the pathologies related to cysts occurring in the oral cavity.^{3,4} In a study conducted by Tekkesin et al where 5088 biopsy specimens of cysts were evaluated, they stated that around 98.3% of the cases were odontogenic cysts and the rest had non-odontogenic etiology.³ In odontogenic cysts radicular or periapical cysts happened to be the most predominant cyst, with Dentigerous cyst being the second most common of the diagnosed cysts followed by odontogenic keratocysts (which has now been reclassified as keratocystic odontogenic tumor) and residual cysts.^{3,5,6} Hence amongst the cysts of

developmental origin seen in the oral cavity, Dentigerous cysts are believed to be the predominating pathology.^{3,4}

Dentigerous cysts are frequently seen in close association with crowns of the unerupted teeth and developing tooth buds.^{2,7} They are generally more prevalent in the mandibular molar and ramus region with high propensity to occur in close relation to impacted third molars.^{8,9} But Dentigerous cysts have also been known to occur with almost similar frequency in association with maxillary canines followed by maxillary third molars and a few rare incidences associated with maxillary incisors.^{2,7} Hence they are also commonly located in the anterior maxillary region.^{2,7,8} But of all the cases reported in literature the occurrence of dentigerous cysts in relation to premolars or as Daley et al stated the association of dentigerous cysts to premolar follicles has been a rare citing.^{2,7,8,9} Various studies have also reported that the occurrence of dentigerous cysts in mandibular third molars is believed to be around 45.7% while the occurrence of dentigerous cysts in relation to premolars is believed to be only about 2.7%.^{10,11} Studies conducted in pediatric age groups additionally reported that Dentigerous cysts are seen more commonly in the maxilla while keratocystic odontogenic tumors were seen more commonly in the mandible for children and adolescents.⁶ This can be attributed to the fact that the samples evaluated in the various studies differed in various aspects. A large number of cases are incidental radiographic findings due to the benign nature of the cyst unless the cyst is secondarily infected, which may lead to a complaint of pain associated with the lesion.^{1,8,12} The agreed criteria to suspect a Dentigerous cyst radiographically has been laid down as an increase of more than 5 mm in the follicular space in regards to the concerned tooth which should then be investigated further.¹³ Majority of the cases of dentigerous cysts have been reported to occur in the second to third decade of life.^{8,12} This high occurrence of cases in pediatric and adolescent age groups can be attributed to its odontogenic origin since a number of factors and metabolic process involving growth and development of the individual are ongoing in those decades; while the third decade is closely associated with the eruption of the third molars.⁶ Due to its high occurrence rate in pediatric age groups dentigerous cysts are often misdiagnosed as periapical cysts when a carious etiology is suspected in the overlying deciduous tooth.¹⁴ Likewise a few studies have also demonstrated that the periapical inflammation due to carious involvement of the deciduous predecessor could result in formation of a dentigerous cyst in relation to the permanent successor.¹⁵ The incidence of dentigerous cysts in males is reported to be twice that of females.⁸ In support of the above stated data Daley et al had hypothesized that since the number of prophylactic third molar extractions done in females is higher, given the reduced jaw size compared to males they have a lower tendency to develop dentigerous cysts.^{8,12} A few studies have also mentioned that Caucasians have higher prevalence of Dentigerous cysts when compared to other races.⁸ Most of the cases reported in literature have reported only a unilateral incidence of Dentigerous cysts and the possibility of the occurrence of multiple dentigerous cysts is very rare and almost exclusively in relation with syndromes or systemic pathologies involving metabolic or developmental disturbances such as mucopolysaccharidosis and cleidocranial dysplasia.^{16,13} In a review of literature from 1943 to 2016 as minimal as 32 cases have been reported of multiple dentigerous cysts without any underlying syndromes with 24 cases occurring only in the mandible, 3 cases only in the maxilla while 3 cases were reported with multiple cysts in the maxilla and mandible.¹⁴

The histiogenesis of dentigerous cysts has been a topic of debate amongst researchers for quite a while now and different theories have been postulated regarding the same based on the multitude of observations of different studies.² The most common and widely approved theory is that dentigerous cysts develop as a result of fluid accumulation around the crown of the unerupted tooth within the follicle surrounding it.¹⁷ This fluid accumulation has been believed to be demonstrated, not just between the reduced enamel epithelium and the enamel of the unerupted tooth bud, but also between the different layers of the reduced enamel epithelium.¹⁷ This accumulation of fluid has been hypothesized to occur due to venous obstruction by the erupting tooth follicle which reduces the venous outflow leading to transudation of serum across the porous capillary wall.^{2, 17} This theory not only accounts for a plausible etiology, but can also be justified by the clinical finding that the fluid often aspirated in Dentigerous cysts is straw colored like serum and shows a biochemical composition close to it.¹⁷ An alternate hypothesis to this was stated by Toller et al that the breakdown of the continuously proliferating cells of the reduced enamel epithelium may lead to release of end products causing a rise in the osmotic pressure inside the follicle, resulting in fluid accumulation and cystic transformation.¹⁸ As per recent studies reported in literature, 8 cases of bilateral mandibular dentigerous cysts have been predicted to have occurred due prolonged use of cyclosporine A and Calcium Channel Blockers.¹⁹ In vitro studies have also shown that sub epithelial micro cysts could be induced beneath the secretory ameloblasts by ingestion of high concentrations of fluoride.²⁰ These small blisters could be reproduced, beneath a similar epithelium by transplantation of the tooth in a different subject that may be occurring due the lack of a basement membrane and presence of hemidesmosomes in the reduced enamel epithelium resulting in easy osmosis of the fluid due to high ion concentration.^{22,23} Hence this demonstrated the fact that the probability of developing a dentigerous cyst is much higher at the secretory stage or during the formative stage of the tooth bud; thus also justifying the enamel defects commonly seen in teeth associated with dentigerous cysts.²⁰⁻²³ This also supports the observation that the dentigerous cysts are

known to occur in the second to third decade more commonly and that the natural progression of the epithelium surrounding the follicle is from a specialized enamel epithelium to squamous epithelium while the tooth remains unerupted or impacted even with an increasing age of the patient does not increase the incidence of dentigerous cysts.^{20, 22, 23}

II. CASE REPORT

A 13 year old boy reported to the Department of Dentistry at Dr. R. N. C General Hospital, Mumbai with the chief complaint of swelling in the right cheek region since 4-5 months (refer to fig. 1). The swelling had gradually increased in size causing observable asymmetry of the face over a period of 4-5 months and was associated with slight pain on touching, chewing or closing the mouth. On further questioning the patient's parents gave history of being regular in following up with his daily oral hygiene. Patient had no history of sero-sanguinous or pus discharge from the nose or the oral cavity neither did he give a history of any systemic complaints or past illness.

Clinical Examination:

The extra oral examination of the patient revealed facial asymmetry (refer to fig. 1) on visual inspection with swelling present, overlying the right maxillary region, below the zygomatic arch and obliterating the nasolabial fold. The swelling measured approximately 2x2 cm in size. The skin overlying the area of swelling revealed no relevant changes and on palpation was free and afebrile. No draining sinus tract was observed either intraorally or extraorally on visual inspection and palpation. On further examination it was revealed that the swelling was firm to hard on palpation, non-tender and non-fluctuant. The swelling had no pulsation or bruit. There was no paraesthesia extraorally or intraorally in and around the area of the swelling. On further palpation it was revealed that the swelling was non mobile and attached to the maxilla. The Temporomandibular joint of the patient was non-tender on palpation and no clicking sounds were heard bilaterally. There was no deviation of the mandible observed on opening and closing of the mouth. The mouth opening of the patient was slightly restricted due to the discomfort caused by the swelling. In this case the mouth opening of the patient was approximately 35-40mm which is around 5-10mm less than the expected mouth opening for pediatric patients factoring in the age, size of the jaws and the overall growth of the patient.²⁴

The Intraoral examination revealed a diffused swelling on the right side of the maxilla extending approximately from the 11 region up to the periapical area of the 16 involving the alveolar ridges on both the buccal and lingual aspects and obliterating the buccal vestibule in its extent (refer to fig 2). On palpation egg shell crackling sound was heard over the buccal aspect of the swelling. It also had a hard surface on palpation with some areas also revealing a firm consistency on the buccal aspect which lead to the suspicion of a buccal cortical plate expansion overlying the lesion. The intraoral mucosa overlying the swelling appeared slightly translucent, stretched and shiny. There appeared to be no swelling or egg shell crackling palatally and soft tissue of the hard palate in the involved quadrant also showed no relevant abnormality. Dental Examination of the upper right quadrant revealed the presence of deciduous 53, 54, and 55. The permanent maxillary canine and the maxillary first premolar on the right side were noted to be missing. None of the teeth in the involved quadrant or involving the swelling showed any carious involvement. There was no pain on percussion in any of the teeth in the involved quadrant. No other relevant hard tissue or soft tissue finding were present.

Investigations:

An orthopantomogram (Refer to figure 3) was advised for the patient to begin with which revealed a well circumscribed radiolucent lesion with a corticated border that was seen on the right side extending from the upper right permanent first molar up till the permanent lateral incisor on the same side. The maxillary right first premolar was noted on the OPG within the cavity of the radiolucent lesion. The permanent maxillary canine was noted to be vertically impacted in the periapical region of the permanent maxillary right lateral incisor. The unerupted canine was present in close proximity of the lesion but did not show any involvement on the radiograph with the same. The roots of the permanent lateral incisor and the maxillary second premolar on the right side were displaced by the lesion. Root resorption was noted with respect to the deciduous 53, 54 and 55, with the root of the deciduous maxillary canine (53) showing resorption up to the middle third. Following this a CT scan was advised for the patient.

The CT scan (Refer to figure 4 and 5) confirmed the above mentioned findings of the OPG and additionally revealed that, the radiolucent lesion in the right maxilla was involving the right maxillary sinus and extending up to the infra-orbital rim with close proximity to the lateral nasal wall on the same side. The thinning of the buccal and lingual cortical plates was clearly visible on the scan.

The hematological and urine investigation carried out for the patient to eliminate were within normal limits. A Fine needle aspiration was done for the swelling following the radiological investigations and it revealed a straw coloured fluid (Refer to figure 6). The provisional diagnosis of Dentigerous cyst was taken into

consideration based on clinical and radiological examination and Odontogenic Keratocyst, Aneurysmal Bone Cyst and Unicystic Ameloblastoma were considered as differential diagnosis.

Surgical Management:

Based on the provisional diagnosis and the extent of the lesion it was agreed upon to perform an excisional biopsy under General Anesthesia.

Saline with adrenaline in a ratio of 1:100000 was infiltrated from the permanent maxillary central incisor on the left side to the permanent maxillary second molar on the right side on the buccal and lingual surfaces of all the teeth. A gingival marginal incision was taken from permanent maxillary central incisor on the left side to the permanent second molar on the right side with a releasing incision at the permanent maxillary left central incisor. A full thickness trapezoidal flap was raised using periosteal elevators to expose the underlying bone (Refer to Figure 7) which was expanded and only a paperthin buccal cortical bone was remaining. Areas of dehiscence on the buccal cortex were also present throughout the lesion. The paperthin bone was removed using Rongeurs and the underlying cystic lesion was exposed. With the use of periosteal elevators, the lesion was separated from the underlying bone and the entire cystic lining was enucleated out in toto. Since the left maxillary first premolar was involved with the cystic lining the enucleation of the lesion involved the extraction of the associated premolar (Refer to Figure 8,9). The permanent maxillary right second premolar also showed significant involvement with the lesion and a poor prognosis and hence was extracted. Since radiological scans and clinical evaluation revealed resorption with respect to the roots of deciduous 54 and 55 they were also extracted (Refer to Figure 10). The residual bony bed was inspected for any remnant tissue. Thorough curettage was done followed by irrigation using betadine diluted with saline. Hemostasis was achieved post curettage and irrigation.

The erupting permanent maxillary canine which was seen beyond the root of the permanent lateral incisor was left as it is to erupt by its natural course and since the deciduous maxillary canine (53) showed no significant involvement and minimal resorption hence it was left in place until the eruption of its permanent successor.

The incision was closed using 3-0 vicryl. Basket Sling sutures were given throughout the incision along with simple interrupted sutures wherever it was indicated (Refer to Figure 11).

The patient was kept in In-Patient post-operative care for 2 days and then discharged with instructions to follow up on an OPD basis.

Histopathology:

The enucleated lesion along with the extracted maxillary premolars and the deciduous maxillary molars was sent for histopathological analysis.

The histopathological report revealed that the lesion showed the presence of an ulcerated, attenuated thin layer of stratified squamous nonkeratinizing epithelium lining the cystic cavity, around 2-3 cells in thickness. The entire cyst was surrounded by a connective tissue wall. The surrounding fibrocollagenous wall showed a dense chronic inflammatory infiltrate. The infiltrate consisted of lymphocytes mainly. The dental specimens revealed no abnormality although the cystic lining was associated with the maxillary first premolar. No dysplastic changes were observed.

Hence histopathology of the specimen confirmed the diagnosis of a Dentigerous Cyst in relation to the unerupted first premolar.

Follow up:

The patient was asked to follow up weekly for one month followed by monthly visits for the next 6 months. Post-operative recovery was good and the healing was uneventful. The patient was followed up with for 1 year and did not show any evidence of recurrence.

There was good bone formation in the cystic cavity, and the canine had started erupting in its physiologic position (Refer to Figure 12).

III. DISCUSSION

In case, an underlying pathology is suspected and enucleation of the lesion is planned it is imperial to advise 3D imaging and panoramic radiographic scans for the patient; to visualize the approximation or the involvement of the lesion with respect to the surrounding anatomical structures.^{1, 13} These scans serve as an aid in locating the defect caused by the lesion while also aiding in estimating the pattern of growth of the lesion.^{1, 13} For the above mentioned case report we had advised a panoramic radiograph followed by CT scan of the face. The follow up scan was particularly more helpful in defining the borders of the lesion on 3 dimensional planes and also helped in ascertaining the non-involvement of the maxillary canine and the maxillary sinus with the lesion which was crucial for performing an adequate enucleation. The occurrence of a Dentigerous cyst in

relation to impacted teeth and unerupted tooth follicle is very high.²⁵ In this case although not impacted but the unerupted premolar follicle was associated with the Dentigerous cyst and hence it was extracted it to prevent recurrence.

In Indian populations Dentigerous cysts obey the general observation of distribution of cases in the second decade; a similar pattern was noticed in this case.^{3,21} Although Dentigerous cysts are not as aggressive as Keratocystic Odontogenic Tumors or Ameloblastomas; the progressive enlargement of the cystic cavity can still lead to thinning of bone if neglected and can often lead to involvement off the sinus especially when associated with the maxillary canines and premolars.²⁶ Hence surgical intervention is a must for patients suspected to have Dentigerous cysts.

The surgical line of management is the advised protocol for patient suffering from Dentigerous cysts given that they have been reported in literature to have a high rate of recurrence.¹¹ The preferred line of management for pediatric patients has been suggested as marsupialization which may or may not be followed by enucleation given the risk of hampering the growth of the maxilla and mandible that exist if radical surgical intervention is done for these patients.¹¹ But enucleation should also be taken into account due to secondary factors such as risk of recurrence, malignant transformation and displacement of anatomic structures due to aggressive growth resulting in hampered function of the patient.¹¹ For our patient since the surrounding bone was healthy and the lesion was not very extensive surgical enucleation was the treatment of choice. Although the risk of transformation of Dentigerous cysts is very low, clinical and radiographic follow up should be maintained for the patient periodically.¹¹ Dentigerous have been reported in literature to show transformation to ameloblastoma and keratocystic odontogenic tumour occasionally which may then grow aggressively.^{26, 27} The risk of Dentigerous cysts showing dysplastic transformation to squamous cell carcinoma although less still exists and has been documented in literature; hence it is essential that a histopathological examination of the specimen be conducted after surgical intervention to rule out the risk of any malignancy.²⁷

IV. CONCLUSION

In conclusion it can be said that Dentigerous cysts have a high frequency of occurring in the second decade of life with a close association of the cysts to unerupted tooth buds.^{3, 13, 5} Hence it is imperial that any patient with missing teeth in the suspected age group be examined additionally with radiographic scans to avoid the risk of neglecting a cyst at an early stage. This becomes even more important in cases such as the one stated above where the general opinion would suggest that a maxillary canine would have been involved, but actually the causative tooth was found to be maxillary first premolar; which in itself is a rare citing in literature. For patients diagnosed with dentigerous cysts the surgical enucleation of the lesion should be considered primarily over marsupialization and the lesion must be followed by histopathological examination of the lesion.^{11, 13, 27} For our patient we maintained a clinical and radiologic follow up of up to 1 year to avoid the risk of recurrence and to monitor the eruption of the teeth that were impacted due to the cyst. As regards to its origin further research into the systemic and local factors involved in the formation of the cyst may be able to give a clearer picture in the future.

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Fig 1

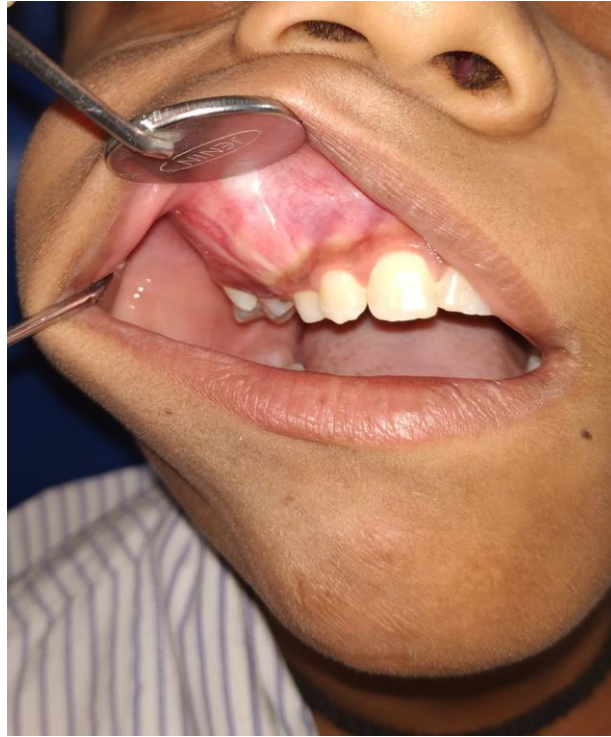


Fig 2



Fig 3

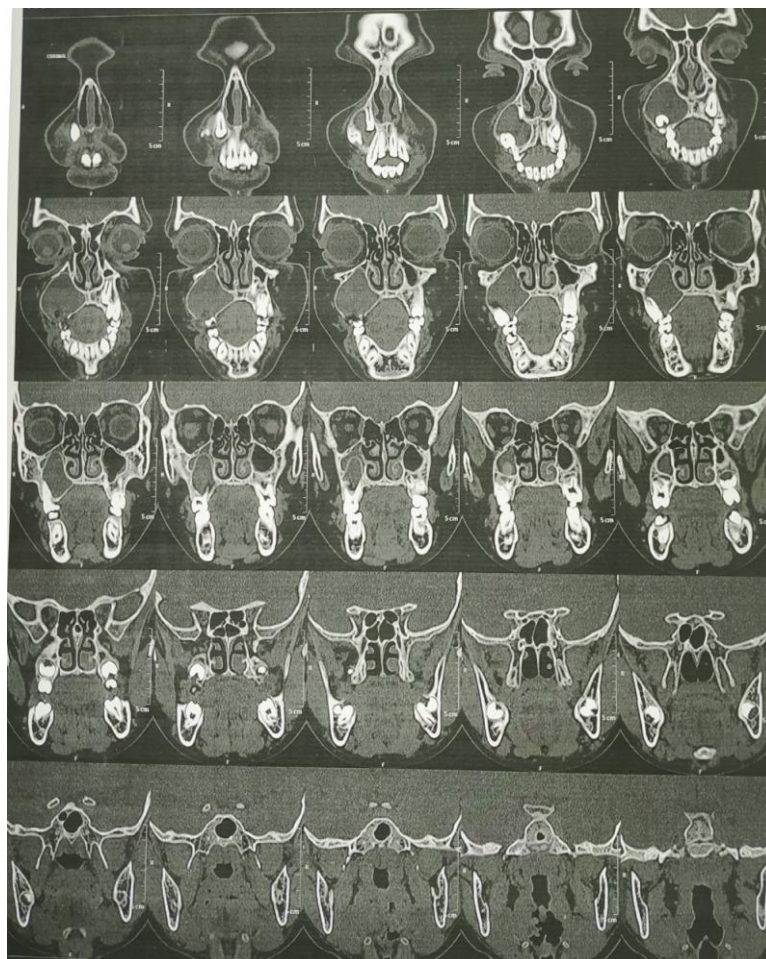


Fig 4

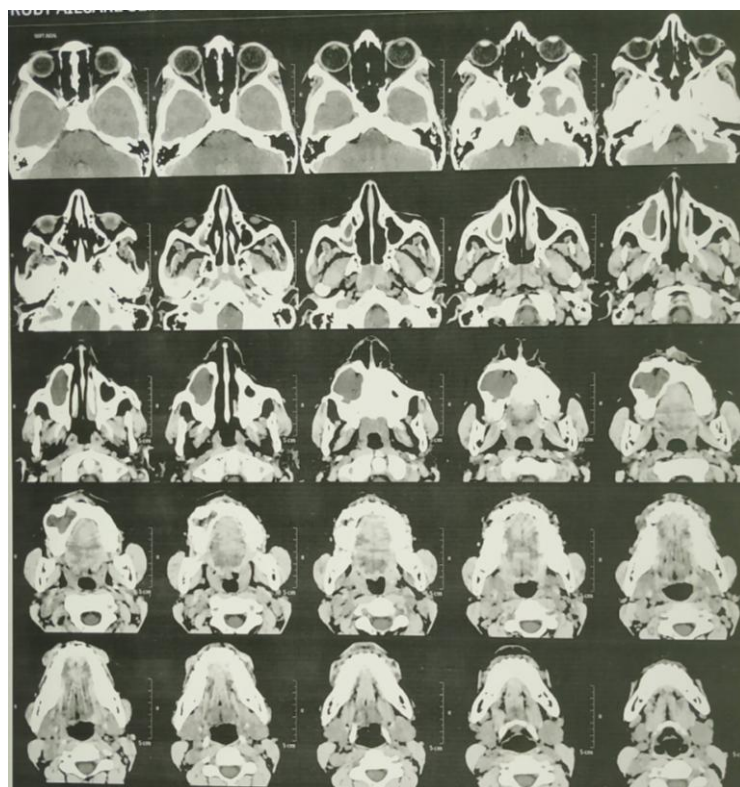


Fig 5



Fig 6

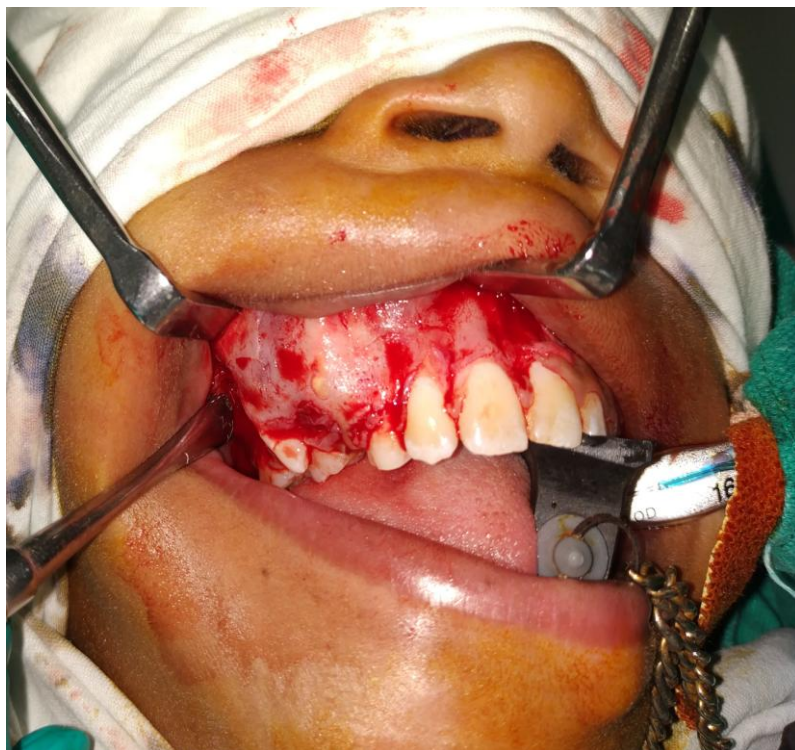


Fig 7



Fig 8

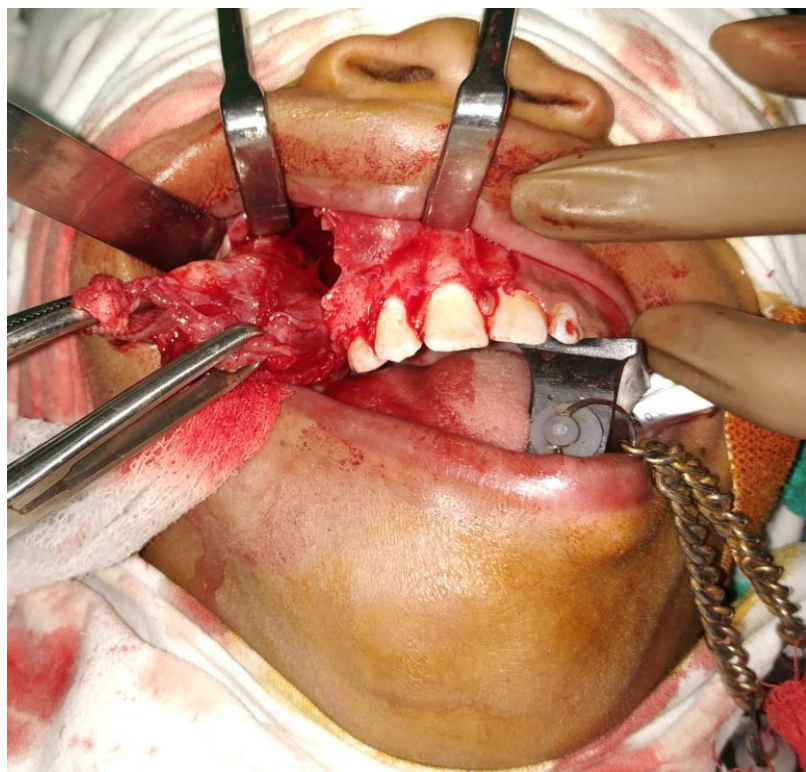


Fig 9

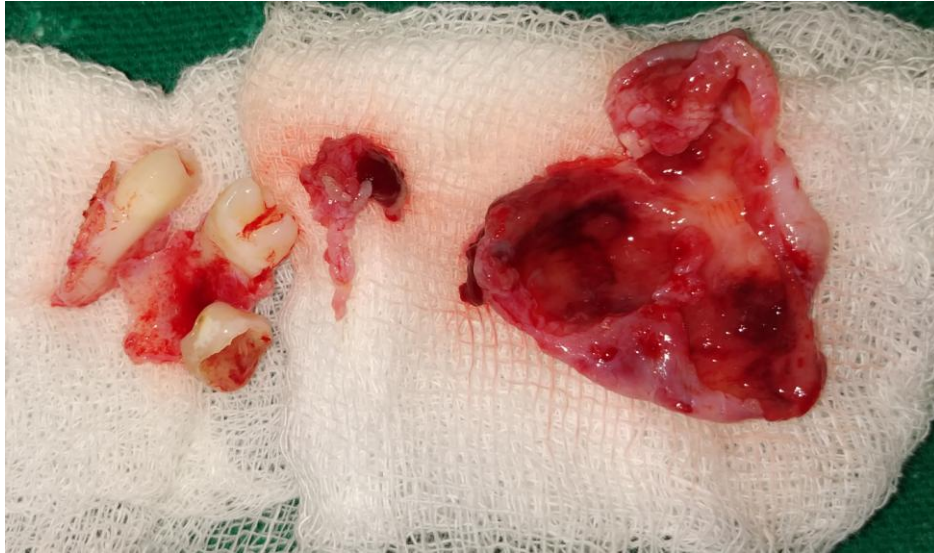


Fig 10



Fig 11



Fig 12