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### RESEARCH ARTICLE

#### DENTIGEROUS CYST TRANSFORMING INTO MUCOEPIDERMOID CARCINOMA OF THE MAXILLARY SINUS IN A 10 YEAR OLD CHILD – A RARE CASE REPORT.

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#### Abstract

Mucoepidermoid carcinoma of the jawbones is an extremely rare malignant tumour involving maxillary sinus. We report a case of mucoepidermoid carcinoma of the maxillary sinus in a 10-year old male patient developing from a previously diagnosed dentigerous cyst. An excisional biopsy was performed and the histopathological features confirmed mucoepidermoid carcinoma. The origin of central mucoepidermoid carcinoma could be suggested to be from the epithelial lining of previously diagnosed dentigerous cyst. Thus, emphasizing the need for careful examination of the entire excision specimen to rule out such neoplastic transformation of epithelial lining of odontogenic cyst and provide appropriate and effective treatment.

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#### Introduction:-

Dentigerous cyst is a most common developmental odontogenic cyst that originates by separation of dental follicle around the crown of unerupted tooth. Usually asymptomatic and associated with dentigerous cysts unless there is secondary infection. Any tooth may get involved, but most commonly mandibular third molar is involved (Neville et al.2006). The age range varies widely, from 5 to 57 years (Shear 2007). The highest incidence of dentigerous cyst occurs during second and third decades of life and rarely seen during childhood. According to Browne et.al<sup>1</sup> the sex predilection of dentigerous cyst is 1.6:1(male: female). Sometimes, lining of dentigerous cysts may transform into malignancies like squamous cell carcinoma(SCC)<sup>2,3,4,5,6</sup> (50%), mucoepidermoid carcinoma(MEC)<sup>7,8</sup> (50%), ameloblastoma<sup>9</sup> (14.6%) and adenomatoid odontogenic tumour(AOT)<sup>10</sup> (73%).

Malignant tumors of the paranasal sinus are uncommon, constituting less than 1% of all malignancies<sup>11</sup> and 3% of all head and neck cancers and in that malignant transformation of odontogenic cysts into mucoepidermoid carcinoma is estimated to be between 0.13% and 2%, with most of the cases involving the mandible<sup>12</sup>. Mucoepidermoid carcinoma accounts for 13% of all malignancies occurring in maxillary sinus<sup>13</sup>. 7 cases of mucoepidermoid carcinoma which was transformed from dentigerous cyst have been reported in children younger than age 16 of which 4 cases involved mandible and 3 cases involved maxilla<sup>14,15</sup>.

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We report a case of mucoepidermoid carcinoma of maxillary sinus arising from previously diagnosed case of dentigerous cyst involving impacted permanent maxillary canine in a 10 year old male child.

### Case Report:-

A 10 year old boy reported to GITAM Dental College and Hospital with a painful swelling in the right upper posterior region producing a feeling of dullness in the right maxillary sinus region. It has been present for about 3 months. Pain was continuous, radiating, aggravated on slight touch, during mouth opening and on lying down position and relieved by itself. There was no significant past medical history, family history or personal history. On general examination, patient was moderately built, nourished, co-operative and responsive. No rise in temperature and vitals were normal.

Extra-orally, on inspection, gross asymmetry on right side of the face was noticed (Fig.1). Swelling was irregular measuring about 2x3 cm, with obliteration of nasolabial fold. It extends from lateral wall of nose anteriorly to tragus of the ear posteriorly. It extends from 1cm above the ala-tragal line superiorly to 1cm below the ala-tragal line inferiorly. On palpation, swelling was very tender, firm, non-mobile, non-compressible, non-reducible, non-fluctuant, no sinus openings over the swelling.

Intra-orally, a swelling was seen on buccal and palatal portion of maxillary posterior region. Swelling was irregular extending from 53 to 16, completely obliterating the buccal vestibule (Fig.2).



**Fig. 1:-** Facial asymmetry



**Fig. 2:-** Swelling.

On palpation, swelling was very tender, firm, non-fluctuant, non-mobile, non-compressible, non-reducible. Teeth with tender on percussion were 53, 54, 55 and 16. The swelling was seen as an ill-defined bony mass, hard in consistency, erythematous surrounded by pinkish red area.

Panoramic radiograph revealed a massive radiolucent lesion from 53 to 16 involving maxillary sinus. The radiolucency was regular, well defined, trabecular pattern of alveolar bone was completely lost and lesion was invasive. Impacted canine (13) was displaced by the cyst to the roof of maxillary sinus. (Fig.3)

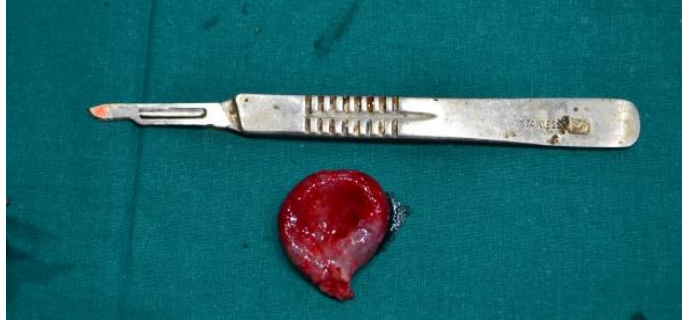


**Fig. 3:-** OPG: Unilocular well defined radiolucency.

Provisional diagnosis was dentigerous cyst and incisional biopsy was done. Histopathology revealed a 3-4 layered thick stratified squamous non-keratinized epithelium with fibrous connective tissue wall and few inflammatory cells suggestive of dentigerous cyst. A surgical approach was planned immediately after investigating blood reports. The standard treatment for dentigerous cyst of the maxilla involving the sinus is enucleation and extraction of the associated teeth via Caldwell-Luc procedure under general anaesthesia, which was followed for the present case (Fig.4). A crevicular incision was given from 21 to 16 region with an anterior releasing incision and the mucoperiosteal flap was reflected. The extraction of 53, 54, 55, 14, 15 and 16 followed by cystic enucleation and then by the extraction of 12 and 13 was done. There were no post-operative complications and the wound healed uneventfully. The excisional biopsy that was performed intraorally consisted of a bit of soft tissue specimen measuring about 2.5cm x 2cm x 1cm (Fig.5) which was grayish white in color and firm in consistency has been taken for processing. The H&E stained soft tissue section shows cystic lining epithelium with underlying connective tissue.



**Fig. 4:-** Surgical approach.

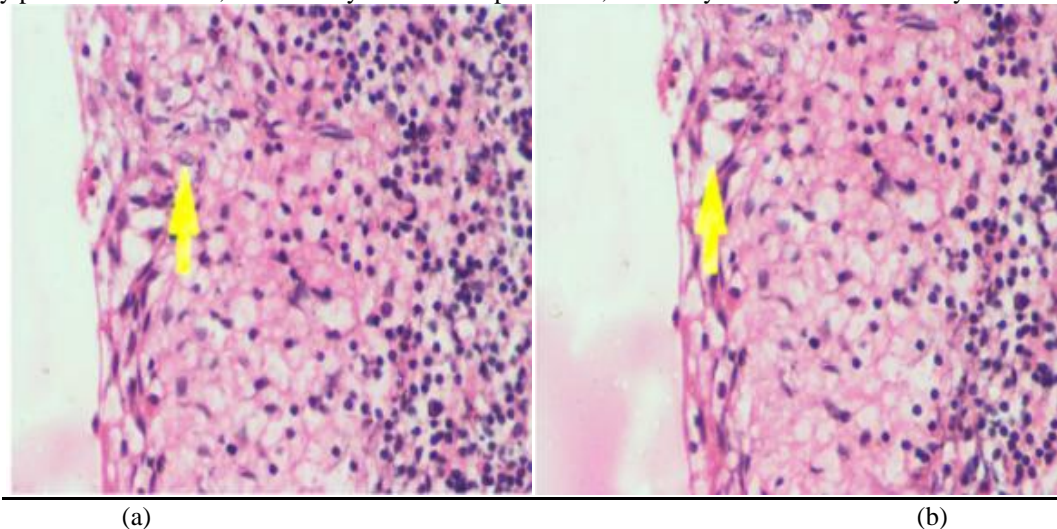


**Fig 5:-** Specimen.

Epithelial lining showed 3-4 layer reduced enamel epithelium. Cystic epithelium is transforming into hyper-proliferative epithelium with characteristic clear cells, intermediate cells and epidermoid cells arranged in nests pattern Fig 6 (a), (b). These cells are also present in connective tissue interspersed with chronic inflammatory cells. The findings were suggestive of Mucoepidermoid carcinoma arising from the lining of the dentigerous cyst. The radiographic evidence of lesion involvement of maxillary sinus along with histological features of clear cells, intermediate cells and epidermoid cells made us suggestive of “Dentigerous cyst transforming in to Mucoepidermoid carcinoma”. 2weeks post-operatively, a temporary obturator was given. Patient was on regular follow-up for the last 18 months without any evidence of recurrence.

### Discussion:-

Dentigerous cyst can be defined as one which encloses the crown of an unerupted tooth and is attached at the cemento-enamel junction. It is a cyst arising by separation of follicle from the anatomical crown of an unerupted tooth within the jaws. The substantial majority of dentigerous cyst involves the mandibular third molars and the maxillary permanent canines, followed by mandibular premolars, maxillary third molars and rarely third molars<sup>16</sup>.



**Fig. 6:-** (a) H&E stained photomicrograph showing cystic spaces & mucous cells at 40x

(b) H&E stained photomicrograph showing cystic spaces, mucous cells & epitheloid cells at 40x

Unlike other odontogenic cysts, epithelial cells that line the lumen of dentigerous cyst arise from reduced enamel epithelium origin and possess an unusual ability to undergo metaplastic transition into other epithelial cell types such as SCC, MEC, AOT and ameloblastoma. Radiographically, the dentigerous cyst presents as a well-defined unilocular radiolucency, often with a sclerotic border. Since the epithelial lining is derived from the reduced enamel epithelium, this radiolucency typically surrounds the crown of the tooth.

Malignant tumors such as mucoepidermoid carcinoma (MEC) of the paranasal sinuses are uncommon, constituting less than 1% of all malignancies. MEC is a malignant epithelial neoplasm composed of both mucus secreting cells and epidermoid type cells in varying proportions. It was first studied and described as a separate entity by Stewart et.al in 1945 (Rajendran et al<sup>17</sup>). Central mucoepidermoid carcinoma is an uncommon lesion in children and only 7

cases in patients younger than 16 years were reported according to the literature revealed. After a systematic review of its histology and degree of differentiation, the WHO Classification in 1991 recommended that the term “mucoepidermoid tumour” be changed to ‘mucoepidermoid carcinoma’. It comprises 5-10% of all salivary gland neoplasms and accounts for 13% of all malignancies occurring in the maxillary sinus.

The criteria for diagnosis of intraosseous mucoepidermoid carcinoma are the following:

- (1) Presence of intact cortical plate
- (2) Radiographic evidence of bone destruction
- (3) Histologic confirmation
- (4) Positive mucin staining
- (5) Absence of a primary lesion in the salivary gland and
- (6) Exclusion of an odontogenic tumor.

The pathogenesis of this tumor is still unknown. Four possible origins have been considered:

- (1) Entrapment of retro-molar mucous gland within the mandible, which later undergoes neoplastic transformation;
- (2) embryonic remnants of the submandibular and sublingual glands trapped within the mandible during development;
- (3) neoplastic transformation and invasion from the lining of the maxillary sinus; or
- (4) neoplastic transformation of mucous-secreting cells from the epithelial lining of dentigerous cyst associated with impacted teeth<sup>16,17</sup>.

MEC arising in the maxillofacial region can have its origin from maxillary sinus lining or central MEC arising from within the bone or from the minor salivary gland. In this case, histological picture is suggestive of MEC arising from the cystic lining of dentigerous cyst. Maxillary sinus malignancies are very difficult tumours to treat and traditionally have been associated with poor prognosis with the main reason being the close anatomic proximity of the nasal cavity and paranasal sinuses to the vital structures such as skull base, brain, orbit and carotid artery. This complex location makes complete surgical resection of sino-nasal tumours a challenging task.

### Conclusion:-

The prognosis for most histo-pathologically diagnosed dentigerous cysts transforming into mucoepidermoid carcinoma is excellent as it is in early stage; also recurrence is a rare finding. It can be therefore suggested that after thorough enucleation of dentigerous cyst, microscopic examination of the entire specimen by the pathologists must be done to determine transformation into mucoepidermoid carcinoma though rare, still possible.

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