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PRIMARY INTRAOSSEOUS CARCINOMA ARISING FROM AMELOBLASTOMA OF MANDIBLE- A CASE REPORT

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ABSTRACT:

Primary intraosseous carcinoma is a very rare neoplasm arising from the cell rests of odontogenic epithelium with no communication to surrounding mucosa. These can be arising from an existing odontogenic lesion and extend within the jaw bone.Because of this, the dentist is now more accountable for making the correct diagnosis and should focus on the entire maxillomandibular complex. Few of similar malignant transformation have been documented in literature as well but the presence of primary intraosseous carcinoma has been reported infrequently. Here, we report a case of primary intraosseous carcinoma in a 80 year old male patient with gross destruction of hemimandible with local metastasis.

Key words : Intraosseous carcinoma, Odontogenic epithelium, Maxillomandibular complex

INTRODUCTION:

Primary intraosseous carcinoma (PIOC) of the jaws is a very rare neoplasm arising from cell rests of the odontogenic epithelium with no communication to surrounding mucosa.[1-3]They can be arising from an existing odontogenic lesion and extend within the jaw bone. It may be arising from odontogenic keratocyst, dentigerous cyst, ameloblastoma or other odontogenic tumors.[2,4] Only a few cases have been reported in the literature where they have been seen malignant transformation of OKC into such primary intraosseous carcinoma. Some literature[3] include an exhaustive review of the published cases of such rare entity to define the diagnostic criteria. But, details or protocols about treatment modalities, patient outcomes, and comparisons with other oral cavity primary sites of squamous cell carcinoma are rarely available. This tumour usually presents as a painful swelling in the jaw and may be asymptomatic for years also which can be diagnosed on routine dental examination and most commonly misunderstood as some odontogenic lesion.[3,5,6] The diagnostic point which needs to be noticed for any suspicion regarding primary intraosseous carcinoma includes:lesion with no ulceration of oral mucosa for more than 4 weeks[3] and those lesion to be sent to histopathological examination with adequate margin.[7] Histopathologically, most PIOCs show the same histological features as squamous cell carcinoma which might be sometimes difficult to arrive at the diagnosis. The tumor is composed of sheets; islands; and strands of squamous cells with marked cellular pleomorphism, nuclear hyperchromatism, and mitotic activity. In a few cases, however, the tumor has a distinctly odontogenic pattern, with basal-type cells arranged in alveoli or in a plexiform pattern with palisading of the peripheral cells which might aid us differentiating between squamous cell carcinoma and intraosseous carcinoma. The standard treatment includes surgery with radiation therapy,[5,8,9] followed by chemotherapy if required as recommeneded by the tumor board if prognosis is severe.[3,5,8,10]

This article presents a case of ameloblastoma in the mandibular angle region turning out as primary intraossous carcinoma.

CASE REPORT

A 80- year - old male patient reported to the department of Oral And Maxillofacial Surgery, of Sree Balaji Dental College and Hospitals, Chennai, with the chief complaint of painful swelling in lower left back jaw for the past 2 months. Patient's general health condition was normal.During an intraoral examination, a swelling was observed in the lower left angle region, obliterating the buccal vestibule and extending from the distal aspect of 37 to the pterygo mandibular raphae . On palpation, the swelling was soft and fluctuant.Tenderness on palpation was positive.

A panoramic radiograph shows a well-defined, unilocular radiolucency.Generalized horizontal bone loss, with an impacted 38 noticed.

Incisional biopsy was done from 34-38 region. The histopathological report of the given specimen showed ameloblastoma of left mandible with pathological fracture in the left angle region of mandible. Considering the age of the patient a conservative plan for complete enucleation of ameloblastoma with removal of impacted teeth and fixation of pathological fracture was planned.

PROCEDURE:

Under GA, nasal tracheal intubation done. 2% lignocaine with 1:80,000 adrenaline administered from 33-38 region. A vestibular incision was made from the 35-38 region, the lesion was excised, and the affected teeth were extracted. Soft tissue adhering to the cystic lining was also removed. Peripheral ostectomy of the entire surgical bed done. The biopsy specimen was then sent for histopathological examination.

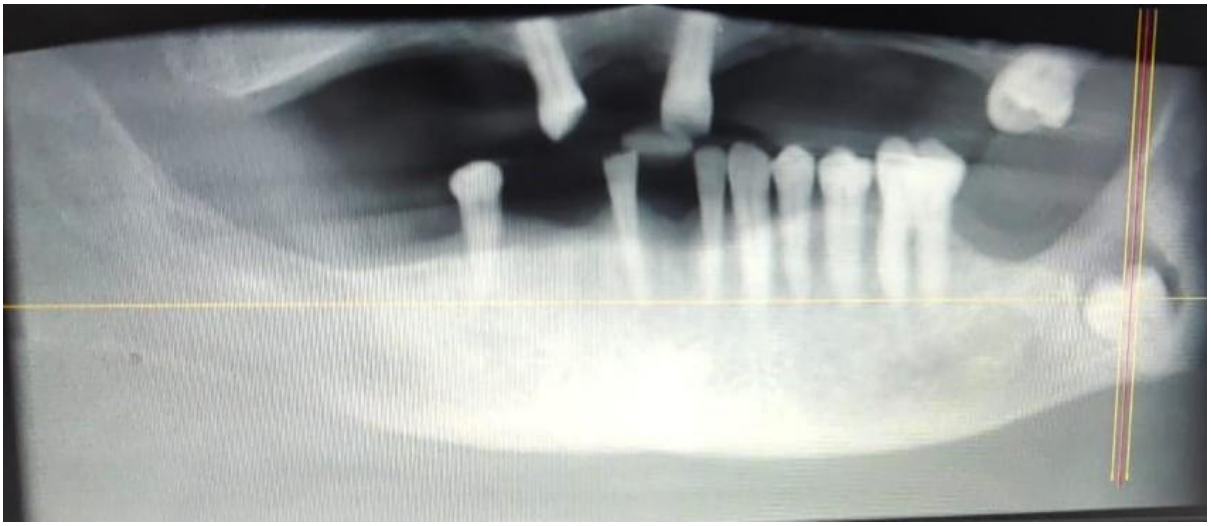

Fig.1: Pre operative radiograph reveals a well defined radiolucency surrounding impacted 38

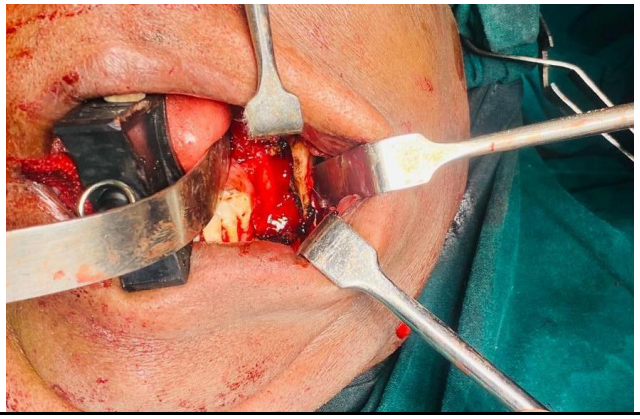

Fig.2: Enucleation of the lesion followed by peripheral ostectomy

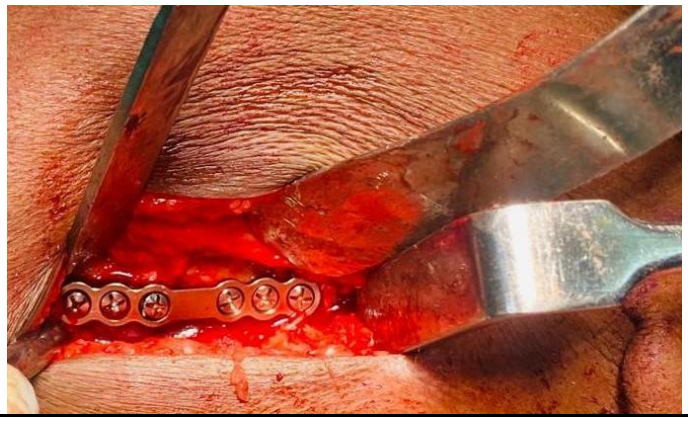

Fig 3: Placement of 6 hole with gap plate and secured with 2*8 mm screws in angle region

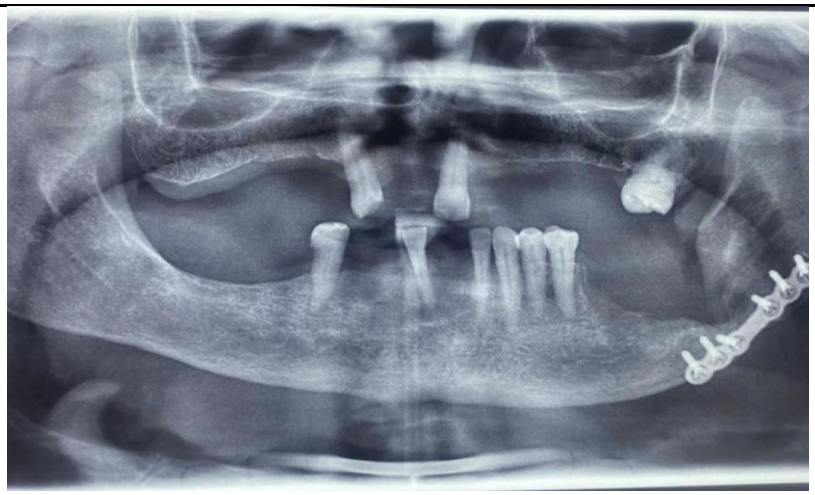

Fig 4: Three month post operative review

DISCUSSION:

On 3rd month follow up bony expansile lesion was noticed radiographically with restricted mouth opening and with complains of mild swelling in the angle region of mandible. A rebiopsy was advised and patient was hesitant for the procedure. On 6th week followup a well defined mass noticed intraorally in the angle region with significant swelling in the lower half of face with restriction of chewing food, dysphagia, lymph aggregation and systemic loss of weight. An incisional biopsy was planned along side with CECT. The biopsy report turned out as Primary intraosseous carcinoma of mandible from previously existing ameloblastoma. The CECT also shown the extent of lesion from the mandibular angle region extending all the way to ramus, condyle and extending slightly into the infratemporal fossa.

PIOC is predominantly found within the jaws, the only bones that may give origin to both connective and epithelial tumours in the head and neck region which grabs special interest in orofacial complex. The mandible is much frequently involved than the maxilla where in the exact etiology remains unknown.8 The association of the PIOC with odontogenic cysts has

already been reported, particularly with residual and radicular cysts, and less frequently with dentigerous cysts, odontogenic keratocysts and lateral periodontal cysts.7,9,10 In rare cases, PIOC results from dedifferentiation of a benign ameloblastoma which is a similar case in literature associating with this case study.4

Risk factors and causes are not well established, but since PIOC develops without initial communication with the oral mucosa, exogenous carcinogens such as tobacco and alcohol are unlikely to be involved which are predominant factors for development of oral squamous cell carcinoma.7 Other factors such as long-standing chronic inflammation and keratinization have been postulated to increase the risk of malignant transformation within an odontogenic cyst which are believed to be due to prolonged inflammation leading to cytological and genetic modifications of cells leading to malignant transformation of cells.8

A high prevalence on male population of the 5th decade of life has been previously documented in literature.11 Symptoms of the disease are non-specific and depend on the location, size and aggressiveness of the tumour and its subjective to vary from person to person. Pain, swelling, sensory disturbance (due to inferior alveolar nerve involvement) and odontogenic disorders are commonly found alongside with trismus and lymph node enlargement as well.11

Bodner et al 9 analysed the clinical and pathological features of 116 PIOCs arising in odontogenic cysts, and reported a large majority of well-to-moderately differentiated tumours (85%). In the same study, the overall survival rate was 62% at 2 years and 38% at 5 years.12

Several hypotheses have been postulated to explain the origin of PIOC over the period of time by different authors and clinicians. Presence of epithelial cells within the bone to give rise to a squamous cell tumour constitutes the most consensual theory.8 The jaw bone by histology are arising from the first pharyngeal arch and is posteriorly remodelled as maxillary and mandibular prominences giving rise to upper and lower jaw as separate entity,Both the prominences consist of cartilages made of neural crest cells. The mandible is formed from Meckel's cartilage with no stem cells of epithelium and hence this theory may not be a standard evidence for the formation of PIOC since it arises from malignant transformation of epithelial cells remnants. Due to this contrary opinion certain authors stated that the PIOC is directly related to transformation of remnants of odontogenic epithelium which may be of dental lamina or cell rests of Malassez or from reduced enamel epithelium.4,5,7,13–16

The diagnosis of PIOC is based on a proper clinical, imaging and histological correlation to rule out the possibility of other malignancy and it never relies on one particular parameter. Primarily clinically PIOC should never have any ulcerated lesion as it has no communication with the mucosa outside the bone and secondarily metastatic migration of SCC should be eliminated. Since there are no specific histopathological features that can distinguish a metastatic SCC from a PIOC, clinical and imaging correlation is mandatory.4

The choice of radiographic evaluation for PIOC is through ionising radiations like Orthopantomograph and Computed Tomography with or without contrast. Any severe cortical erosions noticed in CT might require a secondary evaluation with MRI. Some amount of perineural invasion has been noticed in high contrast studies.13

The radiographic findings of PIOC ranges varying from well defined mass to ill defined lesions with or without bony destruction. Some authors have mentioned the presence of ground glass appearance or small radiopaque foci as well mimicking fibrous dysplasia and ossifying fibroma.9,11,17

All these complexities in diagnosis needs evaluation of PIOC from experts from surgical,

pathological and radiological background as it has properties mimicking other pathological features and most cases will turn out to be misdiagnosed based on one single parameter reports. The PIOC makes it challenging for the team of doctors to rule out the presence of other pathologies which are very closely related to features exhibited by PIOC. Hence from clinical point of view PIOC can be suspected in elderly people with bony lesion with no ulcerative mass in the oral cavity preferabbly in the lower jaw. The radiological perspective would be from findings of extension and bone destruction in CT or MRI or OPG with or without ground glass appearances and finally the histological perspective is through ruling out any possible interpretation close to SCC.When the histological results represent SCC whereas the clinical examination revealed absence of mucosal lesion and radiological evaluation showed presence of metastasis the lesion should be primarily considered as PIOC.

CONCLUSION

Although PIOC is a lesion with a rare occurrence in the literature, it is known that its clinical features can vary. The significance of combining clinical, radiographic, and histopathologic aspects for definitive determination of appropriate pathology and treatment is required.

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