Case Report

Sclerosing odontogenic carcinoma misnomering previously as central odontogenic fibroma-A case report with review of literature

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Abstract

Sclerosing odontogenic carcinoma (SOC) was first described by Koutlas *et al*. in 2008. Despite its inclusion in the World Health Organization (WHO) as a distinct entity, it is a tumour that remains poorly defined in the literature, with only 10 reported cases to date. The mandibular premolar and molar region is more commonly affected compared to the maxilla. In the maxilla, the anterior and the molar regions are most commonly affected. This article describes a case report of a Sclerosing Odontogenic Carcinoma in a 50 year old male patient in the mandibular region. The radiograph showed a well-defined radiolucency extending from the left ramus of the mandible to the right lower molar region. SOC is low grade with mild atypia and frequent mitosis and diffused infiltrative and perineural spread.

Keywords: Central odontogenic fibroma, sclerosing odontogenic carcinoma, WHO 2017

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INTRODUCTION

Sclerosing odontogenic carcinoma (SOC) was first described by Koutlas *et al.* in 2008.[1] SOC is a low-grade odontogenic carcinoma, a rare primary intraosseous carcinoma of the jaw added to the most recent fourth edition World Health Organisation (WHO) classification of head and neck tumours.[2]

Despite its inclusion in the WHO as a distinct entity, it is a tumour that remains poorly defined in the literature, with only 10 reported cases to date.[3]

SOC is present in both females and males. The mandibular premolar and molar region is more commonly affected compared to the maxilla. In the maxilla, the anterior and the molar regions are most commonly affected.[4] It presents as an expansile radiolucency that causes tooth displacement and root resorption. SOC is a low grade with mild atypia and frequent mitosis and diffused infiltrative and perineural spread. The histologic overlap/resemblance of this tumour with other head and neck entities such as desmoplastic ameloblastoma and primary intraosseous carcinoma also render it a diagnostic challenge, and it should therefore currently be regarded as a diagnosis of exclusion.[5,6]

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CASE REPORT

A 50-year-old male patient came to the outpatient department with a chief complaint of swelling in the mandible for 15 years associated with the habit of smoking.

- Extra oral examination of the face showed in Figure 1. Asymmetrical solitary swelling measuring approximately 4 × 5 cm on the left lower one-third of the face. swelling extended anteroposteriorly from the corner of the mouth to 5 cm in front of the tragus of the ear superoinferiorly, 2.5 cm away from the ala tragus line to 0.5 cm above the base of the mandible.
- A firm to hard swelling is seen on the edentulous ridge on intraoral examination, obliterating the buccal vestibule. The colour was the same as the surrounding mucosa, as shown in Figure 2. Round in shape measuring approximately 2 cm anteroposteriorly, 1 cm superoinferiorly in size. The surface was smooth and the edge was indistinct.

Figure 1: Asymmetrical solitary swelling measuring approximately 4 × 5cm on the left lower one-third of the face

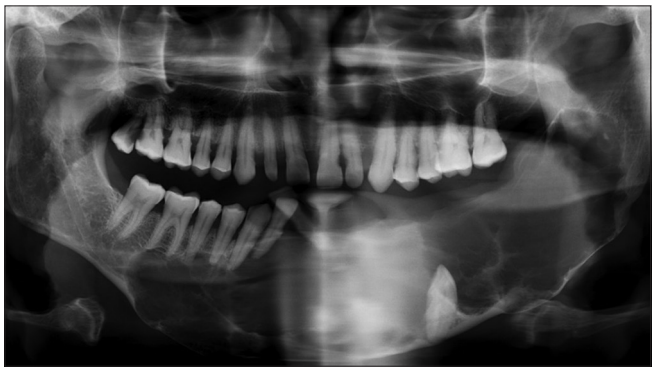

Figure 3: Well-defined radiolucency extending from the left ramus of the mandible to the right lower molar region

On orthopantamogram analysis is shown in Figure 3, a well-defined radiolucency extending from the left ramus of the mandible to the right lower molar region.

An excisional biopsy was performed. Two bits of the specimen were received as shown in Figure 4, which were sectioned into four bits, grayish–brown in colour and firm inconsistency.

The hematoxylin and eosin stained soft tissue section shows odontogenic epithelial islands arranged in strands and clusters as shown in Figures 5 and 6 surrounded by sclerotic connective tissue stroma. Perineural infiltration is also seen as shown in Figure 7a and b.

DISCUSSION

SOC is a rare and relatively new entity, recently added to head and neck tumours WHO classification.[3] With locally aggressive behaviour and bland histology with discrete areas of tissue invasion, which extend beyond this entity deserves a review of the potential causes for the discrepancy and an approach to the differential diagnosis on biopsy. Moreover, we will also emphasise the difficulties in accurate margin status assessment on histopathology. The clinical features of SOC mainly were described as being non-specific and ranged from entirely asymptomatic to

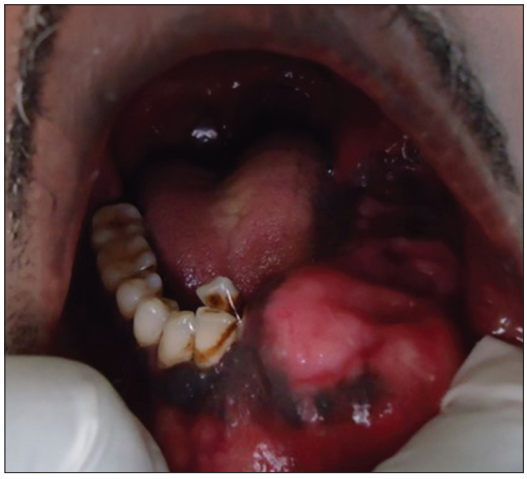

Figure 2: A firm to hard swelling was seen on the edentulous ridge on intraoral examination, obliterating the buccal vestibule

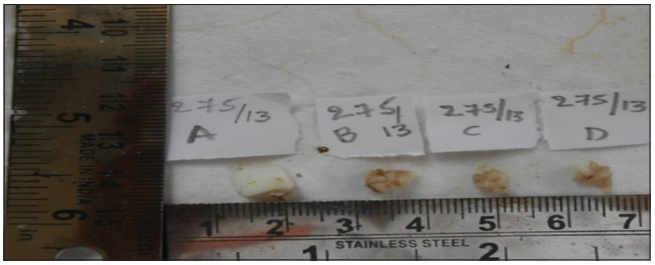

Figure 4: Excisional biopsy was performed

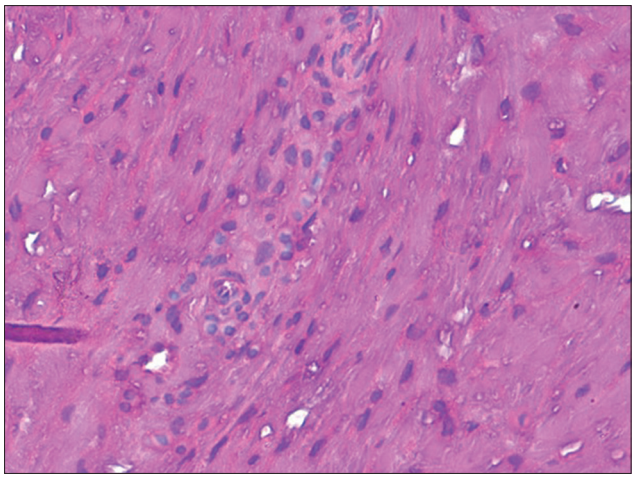

Figure 5: Hematoxylin and Eosin stained soft tissue section shows odontogenic epithelial islands arranged in strands

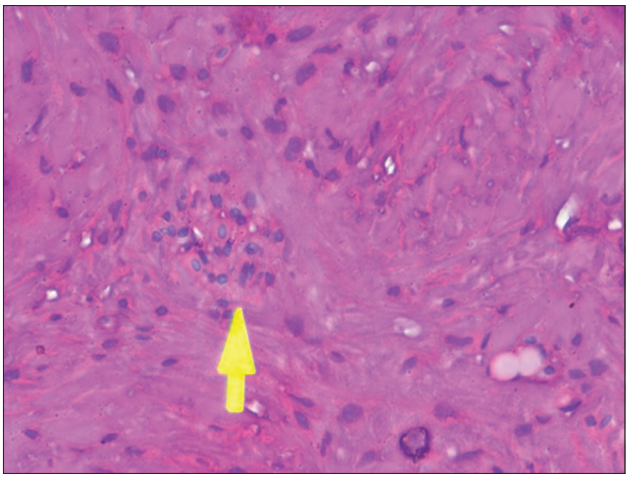

Figure 6: Hematoxylin and Eosin stained soft tissue section shows odontogenic epithelial islands arranged in CLUSTERS

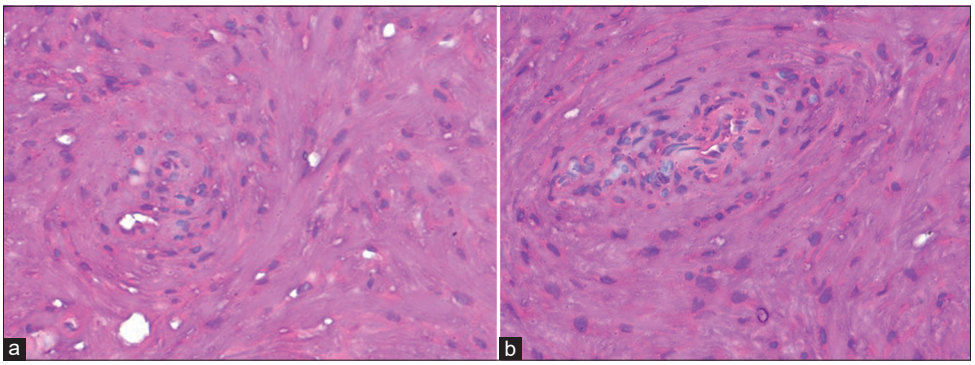

Figure 7: (a) Neural tissue, (b) Large perineural invasion making prognosis poor

cases where the patient had noticed a lump or swelling.[7] On low-power examination, all tumours lack a capsule and show an infiltrative tumour front. The epithelial cells are polygonal with low-moderate atypical nuclei and eosinophilic cytoplasm. These epithelial cells' appearance is reminiscent of the cells of odontogenic epithelial rests when first described.[8]

Based on histomorphology, the differential diagnosis for this tumour is broad as per the recommendation in the latest WHO; care does exclude other diagnostic entities before arriving at a SOC diagnosis, including metastases. By morphology, SOC has shown to be a great mimicker of other neoplasms in the head and neck.[3,9,10]

It also resembles the jaw primary intraosseous carcinoma (PIOC), a rare tumour with a poor prognosis; however, it does not have the significant cellular atypia seen in PIOCs. Hussain *et al*. reported the initial diagnosis of their SOC by core biopsy as a poorly differentiated squamous cell carcinoma.[5] In contrast, Tan *et al*. reported an initial diagnosis of a PIOC.[4] Similarly, Wood *et al*. reported an initial diagnosis of adenocarcinoma.[9] Consideration must also be given to metastatic malignancies and, in particular, neoplasms of epithelial origin. We initially gave the diagnosis of Central Odontogenic Fibroma due to the presence of infiltrating cords and nests of the epithelium with cytoplasmic clearing in the histopathological slide. But, after we revisited the WHO Head & Neck's new classification, we concluded it as SOC.[11,12]

CONCLUSION

Although SOC remains a recently described and rare entity with a few case reports to date, it is an important differential diagnosis to consider for head and neck tumours and a mimicker of other tumours in this region, including metastatic tumours. Finally, recognised as a distinct tumour type in the WHO, SOCs will require more reported cases to standardise a diagnostic approach and treat these locally aggressive neoplasms.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and

other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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