Primary Intraosseous Squamous Cell Carcinoma of The Mandible Arising De Novo
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ABSTRACT
Primary intraosseous squamous cell carcinoma is an odontogenic tumour with aggressive behaviour usually noticed in 6th to 7th decades of life. The tumour is characterized by progressive swelling of the jaw, pain and loosening of teeth. Microscopically, the lesion is showing foci of keratinising cells separated by collagenous connective tissue stroma. A case of primary intraosseous squamous cell carcinoma of mandible arising de novo in a 40-year-old man is reported.

Key words: Intraosseous squamous cell carcinoma. Odontogenic tumour. Mandible. De novo.

INTRODUCTION
Primary Intraosseous Squamous Cell Carcinoma (PISCC) is defined as a type of squamous cell carcinoma arising within the jaws, having no connection with the oral mucosa and presumably developing from the residues of the odontogenic epithelium.1 It may arise within the jaws either from a previous odontogenic cyst or de novo.2 It is usually noticed in 6th to 7th decades of life, with more cases reported in men and is characterised by progressive swelling of the jaw, pain and loosening of teeth.3 Majority of the cases occur in the posterior mandible as painful non-ulcerated lesions.4 The tumour is locally aggressive and metastasises to regional nodes.5

The present report describes a case of PISCC of mandible arising de novo in a 40-year-old man, which was initially treated by a general dentist, who mis-diagnosed the lesion as a periodontal lesion.

CASE REPORT
A male patient aged 40 years reported with a swelling on left lower half of face since one month. Extraoral examination revealed an oval swelling of 3x2 cm involving the left parasymphysis of mandible. Overlying skin was normal with smooth surface and raised temperature. The swelling was bony hard, tender, non-fluctuant and non-pulsatile. Regional lymphadenopathy was present. The patient reported a history of mobility of posterior teeth in the left mandible 6 months prior to referral. The patient had undergone scaling and extraction of 34 showing grade III mobility from the general dentist. Even after oral prophylaxis, there was no improvement and the patient noticed swelling of gums in relation to teeth of left side of mandible. There was no history of tobacco use or alcohol consumption.

Intraoral examination revealed a diffuse swelling in relation to 33, 35, 36, 37 and 38. Gingiva was detached in relation to 33, 35, 36, 37 and 38. (Figure 1). The overlying mucosa was normal but slightly erythematous. The teeth (33, 35, 36, 37 and 38) present in the region were showing grade III mobility and grade II furcation involvement. Periodontal examination showed pocket depth ranging from 5-7 mm buccally and 6-8 mm lingually. Buccal and lingual expansion was evident. Panoramic radiograph revealed a radiolucent lesion in relation to teeth 35, 36, 37 and 38 involving the angle. The possibility of malignant tumour was considered on the basis of clinical findings. The lesion was surgically excised and the specimen was subjected to histopathological examination. The lesion showed foci of keratinising cells separated by collagenous connective tissue stroma and some islands of epithelial cells showing pleomorphism, nuclear hyperchromatism, loss of polarity, altered nuclear cytoplasmic ratio and bizarre mitotic figures (Figure 2). The surface epithelium did not show connection with the tumour nests. Based on the
conventional microscopy, a diagnosis of keratinising squamous cell carcinoma of mandible was made. The patient was scheduled for further investigations like ultrasonography, chest X-ray and bone scan and the results were non-contributory. On general examination, the patient showed no clinical evidence of systemic disease. Based on clinical and pathological findings, the diagnosis of primary intraosseous squamous cell carcinoma was formulated.

The patient was referred to Regional Cancer Centre, Trivandrum and underwent hemimandibulectomy in conjunction with radical neck dissection. Every month follow-up for 5-year period was advised for the patient.

**DISCUSSION**

PISCC was first described by Loos in 1913. Willis suggested the term intra-alveolar epidermoid carcinoma and Shear later revised the term intra-alveolar epidermoid carcinoma to primary intra-alveolar epidermoid carcinoma. WHO classified this tumour in 1972, which was later reviewed by Elzay in 1982. Waldron and Mustoe in 1989 histologically classified PISCC arising de novo into Type 3a (keratinising type) and Type 3b (non-keratinising type) with addition of intraosseous mucoepidermoid carcinoma. The age of affected patients ranges from 4 to 81 years, with male predilection. The most commonly affected site is the posterior mandible. These features are consistent with those in the present case. The histologic features of the present case were confirmed to be those of keratinising PISCC arising de novo. Histologically, 2 patterns of growth have been described. The margins of PISCC lesions are poorly defined, diffuse and irregular in most cases. In the present case, a radiolucent lesion was evident in relation to teeth 35, 36, 37 and 38 involving the angle.

In the present case, the diagnosis of PISCC was delayed and the general dentist has given top priority to dental problems and misdiagnosed as periodontal disease. Tooth mobility may be due to both periodontal disease and invasion of the mandible by the neoplasm.

If PISCC is suspected, a multidisciplinary diagnostic approach should always be adopted. The diagnosis of PISCC rests on several criteria including intact oral mucosa before diagnosis, microscopic evidence of squamous cell carcinoma without a cystic component or other odontogenic tumour cells, absence of another primary tumour on chest X-ray and exclusion of the possibility of metastatic lesion from a distant primary tumour. All these diagnostic criteria were satisfied in the case reported here.

To conclude, the present case calls attention to a PISCC of mandible arising de novo, which was misdiagnosed as a periodontal disease. Since tooth mobility, pain and progressive swelling of the jaw seem to be important presenting symptoms of PISCC, these diagnostic criteria have to be considered in all cases, where initial dental treatment has failed. Long-term follow-up is mandatory to rule out regional and distant metastases in PISCC.

**REFERENCES**


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