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Primary Intraosseous squamous cell carcinoma of mandible mimicking odontogenic lesion: An enigma to surgeons – A case report

[1]Dr. Shweta S. Sonwane (Kamble), Associate Professor, Department of Oral and Maxillofacial Surgery, Govt. Dental College and Hospital, Nagpur

[2]Dr. Pooja Umathe, Post Graduate Student, Department of Oral and Maxillofacial Surgery, Govt. Dental College and Hospital, Nagpur

[3]Dr. Abhay Datarkar, Professor and Dean , Govt. Dental College and Hospital, Nagpur

[4]Dr. Prashant Pandilwar, Professor & Head, Department of Oral and Maxillofacial Surgery, Govt. Dental College and Hospital, Nagpur

Corresponding Author: Dr. Pooja Umathe, Post Graduate Student, Department of Oral and Maxillofacial Surgery, Govt. Dental College and Hospital, Nagpur

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Abstract

Intraosseous squamous cell carcinoma (PIOSCC) is an exceedingly rare lesion primarily found in the jawbones, originating from remnants of odontogenic epithelium or odontogenic cysts or tumors. Herein, we report a rare case of PIOSCC initially presenting as an odontogenic lesion.

A 38-year-old male patient presented to the outpatient department (OPD) with swelling and trismus, resembling an odontogenic lesion, in the mandible. Surgical intervention was performed based on this presentation.

The posterior mandible exhibited a predilection for PIOSCC, particularly in males. Initial surgical intervention was pursued under the assumption of an odontogenic lesion, but histopathological analysis revealed intraosseous squamous cell carcinoma, necessitating hemi-mandibulectomy. Preceding this, a PET scan was conducted, revealing no significant findings, and palpation indicated no palpable lymph nodes. The patient is currently under follow-up.

Swelling and continuous pain are common presenting symptoms of PIOSCC, often leading to consideration of benign odontogenic conditions in diagnosis. This case underscores the importance of recognizing intraosseous carcinoma as a potential diagnosis in cases of persistent pain and swelling in the jaw, facilitating early and appropriate treatment interventions.

Keywords: Intraosseous Squamous Cell Carcinoma, Mimicking, Odontogenic Lesion, Rarelesion, Jaw Lesion

Introduction

Primary intraosseous squamous cell carcinoma is a very rare clinical entity affecting the jaws, especially in the elderly population. The condition develops from the remnants of the odontogenic epithelium or odontogenic cysts and tumors[1]. The tumor was first described by Loose in 1913. It was renamed by Willis in 1948 as intra-alveolar epidermoid carcinoma, and Shear modified the name again to Primary intra-alveolar epidermoid carcinoma[2,3]. WHO recommended the term primary intraosseous carcinoma, and in 2005, it was designated as a primary intraosseous squamous cell carcinoma[4]. The cases reported in the literature are very few; hence, a case of PIOSCC, which was difficult to diagnose, is presented here.

Case Report

A 38-year-old male patient reported to the Department of Oral and Maxillofacial Surgery, Government Dental College and Hospital, Nagpur, with a chief complaint of pain and swelling in the lower left side of the face, along with reduced mouth opening for one year. The patient did not have any past medical history. There was a history of areca nut chewing once daily, but he quit his habit five years back. On extra oral examination, there was diffuse swelling on the lower left side of the face. There was no sinus tract, and no color change over the swelling. On palpation, the swelling was soft and slightly tender, temperature over the swelling was normal. Mouth opening is restricted up to 1 finger, and no associated lymphadenopathy. TMJ movements bilaterally palpable and reduced due to trismus. Intraorally, the overlying mucosa was intact without cortical expansion. All these clinical findings point towards an odontogenic lesion involving the left side of the mandible. So, we performed further radiographic investigations.

The orthopantomogram shows a unilocular radiolucency at the left ramus area, just below the condyle and coronoid process, measuring 10mm by 8mm, with no other bony changes (Figure 1). CBCT scans in axial view show a unilocular hypodense area in the left ramus area with intact buccal and lingual cortex (Figure 2).

From all these clinical and radiographic features, considering it as an odontogenic lesion, we decided to proceed with the surgical removal of the lesion under general anesthesia.

The excised lesion was sent for biopsy, and it was diagnosed as Primary Intraosseous Squamous Cell Carcinoma (PISCC). H&E-stained sections revealed infiltrated proliferative stratified squamous dysplastic epithelium, invading the underlying connective tissue in the form of small and large islands and dissociated cells. These dysplastic cells exhibited features such as nuclear cellular pleomorphism, altered N:C ratio, hyperchromatism, individual cell keratinization, keratin pearl formation, and mitotic figures (Figure 3).

The intervening connective tissue showed collagen fibers interspersed with fibroblasts, multiple endothelial-lined blood vessels, and a moderate to severe chronic inflammatory cell infiltrate consisting of lymphocytes and plasma cells (figure 3).

Based on clinical features such as no history of substance abuse, absence of intraoral lesions, lack of buccal vestibule obliteration, absence of clinical signs of Oral Submucous Fibrosis, presence of normal oral mucosa, intraosseous radiolucency observed in the ramus area in the non-odontogenic zone, and findings from radiographic and histopathologic examinations, we arrived at a final diagnosis of Primary Intraosseous Squamous Cell Carcinoma.

Discussion

According to the World Health Organization's histopathologic classification of tumors, PIOC is a squamous cell carcinoma arising from the jaws, having no initial connection with the oral mucosa and possibly developing from residues of odontogenic epithelium[1,5]. Hence, they are also referred to as odontogenic carcinomas. WHO separated this entity from malignant ameloblastoma and other carcinomas. One should always exclude squamous cell carcinoma arising from the oral mucosa, alveolar ridge, and maxillary sinus lining before giving the conclusive diagnosis of PIOSCC[6].

In the present condition, it was observed that the tumor was arising within the bone because the overlying mucosa was intact and normal. It originated from the bone, and there was an early invasion of the inferior alveolar nerve, rather than invasion from the surface epithelium. In the present case, it is called primary intraosseous squamous cell carcinoma as it originated from odontogenic epithelium or remnants of epithelium or epithelial rests of molasses. Hence, the fact that PIOSCC is rare in bone other than jaw bone supports the concept that PIOSCC is odontogenic in origin.

PIOC arising within is rarer than the one arising from the odontogenic cyst. There are only 28 well-documented cases of PIOC from the year 1984 to 1997. In these patients, the age range reported was 4-81 years with a mean of 53 years. The male-to-female ratio observed was 2.2:1. The majority of the lesions were in the posterior part of the mandible, where remnants of the dental lamina are more likely to be the source of epithelium[3,6,7].

Most patients present with complaints of pain, swelling, and trismus of the jaw, and they were often initially treated as dental problems. This leads to a delay in the diagnosis of PIOSCC. McGowan and Coonar suggested that the poor prognosis of PIOSCC is due to delayed diagnosis. There are no special histopathologic features of PIOSCC, and a diagnosis can be made only if there is no evidence of a tumor arising from the oral epithelium or the odontogenic cyst[8,9].

Suei et al suggested that serial sections of the main histological specimen must be made to demonstrate squamous cell carcinoma without cystic components or other odontogenic tumor cells to rule out the possibility of it arising from odontogenic carcinoma[10].

To EH et al reviewed the literature regarding metastasis to the jaw and concluded that investigation of the primary tumor should include chest radiography and that the patient must be followed for at least 6 months[7]. In our case, a full clinical examination and routine hematological, biochemical, and radiological examination showed no evidence of a primary site, and the patient is still under follow-up with a regular PET scan with a recent report suggesting no evidence of metabolically active disease.

Surgical excision is accepted as the preferred method of treatment. Hemimandibulectomy may be required for extensive mandibular lesions. Involved lymph nodes require block dissection combined with the excision of the primary lesion. In lesions that cannot be surgically excised, other treatment modalities, such as radiotherapy or chemotherapy, may be considered.

Conclusion

PIOSCC is a rare tumor. Whenever a case presents with persistent pain and swelling along with an intraosseous osteolytic lesion with irregular borders, one should also consider the possibility of intraosseous squamous cell carcinoma and rule out other conditions to reach the final diagnosis of PIOSCC. The prognosis of patients with PIOSCC is difficult to determine because of the small

number of reported cases, the different treatment modalities, and variable follow-up times. Additional well-documented examples of PIOSCC should be reported, as this will eventually yield better information on the management and prognosis of these uncommon neoplasms.

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Legend Figures

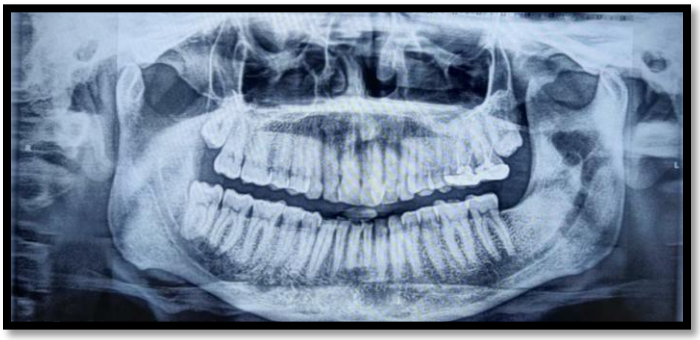

Figure 1: Panoramic image showing the radiolucent area of size 1 by 1 cm in left ramus region of the mandible

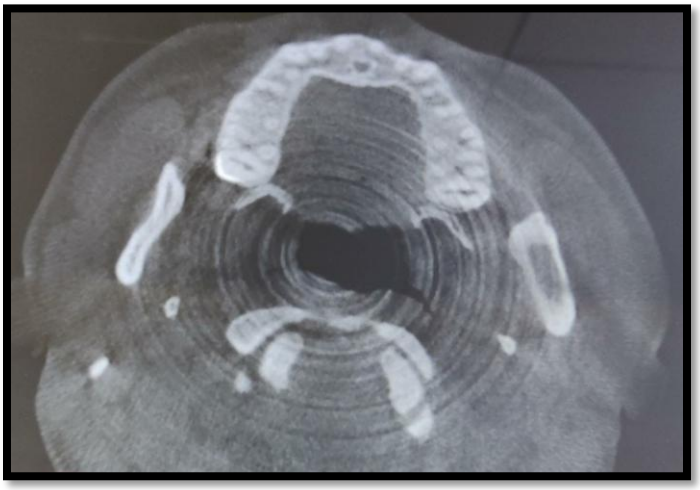

Figure 1: CBCT of the patient with axial view showing 1 by 1 cm hypodense area in left ramus region

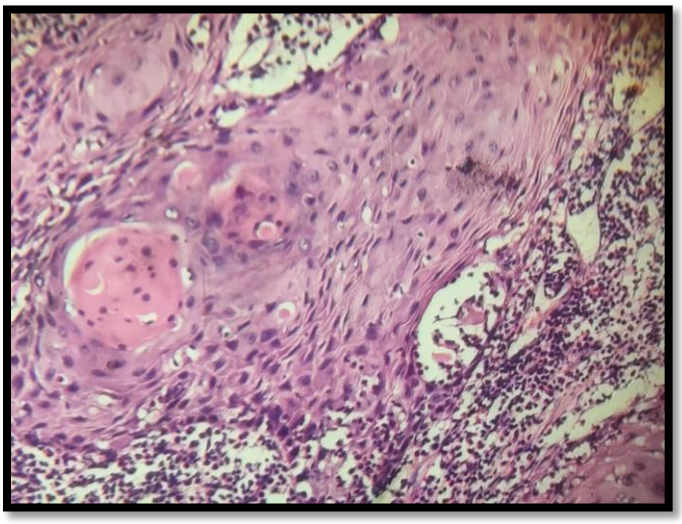

Figure 3: Histopathological picture (10x view)