



## Intraosseous squamous cell carcinoma in the anterior part of mandible: A case report

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### ABSTRACT

Intraosseous squamous cell carcinoma is a rare type of squamous cell carcinoma that requires accurate and early diagnose which is important due to its invasive feature. This article presents a rare case report on intraosseous carcinoma in a 33-year-old patient.

The case was a 33-year-old man who was referred to oral disease department of Babol College of Dentistry complaining of chin pain. In the radiographic view there was a well-defined unilacunar radiolucency extended from the first molar on the right side of mandible to the opposite premolars which caused perforation. Incisional biopsy showed a destructive lesion and finally, the lesion was removed by excision and curettage.

**Conclusion:** In the present study, accurate and early diagnose of Intraosseous squamous cell carcinoma was very important due to the young age of the patient who was under 50 years old. In addition, the large size of the lesion that has passed the midline makes this case noticeable.

**Keywords:** Squamous cell carcinoma, intraosseous squamous cell carcinoma, intraosseous carcinoma, oral cancer, mandible.

### Introduction

Squamous cell carcinoma constitutes about 94% of oral cavity malignancies [1, 4-6] and is the 10<sup>th</sup> common cause of death [4]. This carcinoma is more common in men [5,7] and has diverse clinical variants such as exophytic, endophytic, leukoplakic and erythroplakictypes [1 and 4]. Its etiology is not clear, but some factors such as anemia, iron deficiency, genetic polymorphism, alcohol and tobacco consumption, UV and some viruses such as HPV can be involved in its formation [1, 2,6,8,9]. Intraosseous squamous cell carcinoma is a rare type of squamous cell carcinoma that develops from the odontogenic epithelial remnant, rests of Malassez or lining of a cyst or odontogenic tumors [3, 10-13] and can invade bones. Radiographic features reveal a radiolucent lesion with moth-eaten borders [1]. Squamous cell carcinoma

is common among individuals older than 50 years [5,7] and it is rarely (1-6%) seen in individuals younger than 40 years. It is more important in younger patients compared to older individuals regarding its invasive nature and likelihood of local recurrence and poorer prognosis, as a result, its accurate diagnosis is very important [7].

### A case report

The patient was a 33-year-old man who was referred to oral Health department of Babol College of Dentistry complaining of chin pain. The patient had pain in the anterior part of mandible for last year and had pus drainage. Three months before, one of his mandibular incisors was extracted because of mobility before, one of his mandib-

ular incisors was extracted because of mobility (Fig.1). Intraoral examination showed a bucco-lingual expansion with labial ulcer (Fig.2). The remaining incisors were mobile and displaced.

Radiographic features showed a well-defined unilocular radiolucency in anterior mandible which was extended from the first molar on the right side to the left premolars. The lesion displaced anterior teeth and surrounded an impacted canine. Expansion of the inferior border of mandible was seen with thinning of the inferior cortex in some areas. Presumably because of the downward displacement of the inferior alveolar nerve canal, the canal was not seen within the lesion. Perforations of the cortex were seen on CT scan views (Fig.3,4).

Incisional biopsy was performed. Histopathologic findings showed epithelial islands and sheaths in a fibrovascular stroma. Peripheral cells were similar to ameloblasts and central cells looked like stellate reticulum. Most of the islands showed squamous changes in central areas. Clear cells also could be seen. Based on histopathological findings, ameloblastoma was reported. (Fig 5).

The patient was treated by excisional biopsy and curettage. Microscopic evaluation of excisional biopsy showed sheaths of epithelial cells with individual cell keratinizations, hyperchromatic and vesicular nuclei. Clear cells aggregations were also evident. Based on these findings, intraosseous carcinoma was considered (Fig. 6) and in order to identify the type of carcinoma, immunohistochemical evaluation was done. The sample was positive for p63, CK 7, ki-67 and was mildly positive for PAS staining, but negative for CK 20. These findings supported an intraosseous squamous cell carcinoma (Figure 7-10). A whole body scan and lateral neck sonography were ordered for metastasis evaluation, but the patient refused to cooperate. On 6- month follow up, there was no sign of recurrence clinically, but precise evaluation is not possible due to the patient poor cooperation.

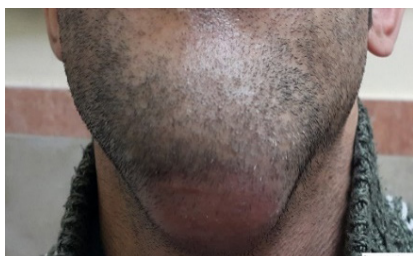


Figure 1: Clinical feature. Note the submandibular.



Figure 2: Intraoral examination demonstrated Swelling. ill-defined swelling in mandibular vestibule.



Figure 3: Radiographic feature shows a well-defined unilocular radiolucency that displaced teeth.

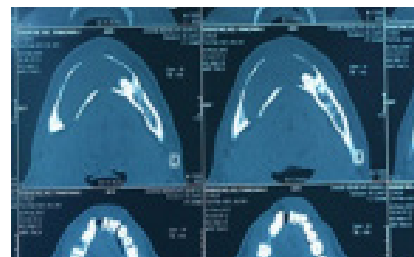


Figure 4: CT feature (Axial) shows that the lesion Perforated the cortex.

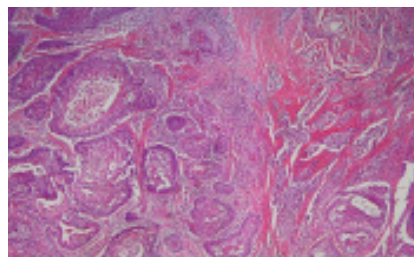


Figure 5: Microscopic view of incisional biopsy islands with keratinization (H&E×100).

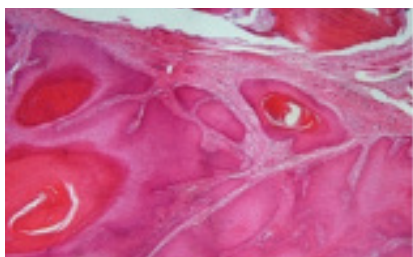


Figure 6: Excisional biopsy. Note the neoplastic epithelial (H&E×40).

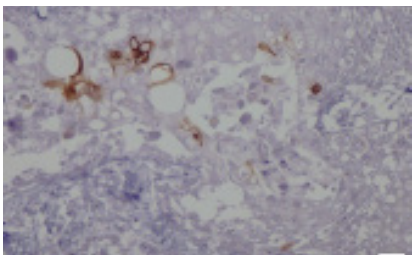


Figure 7: CK 7 (IHC×400).

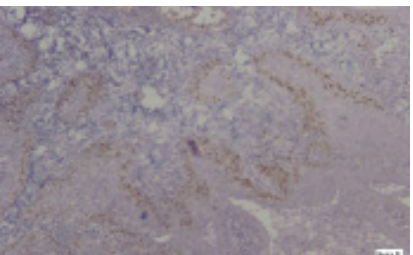


Figure 8: ki 67 (IHC×400).

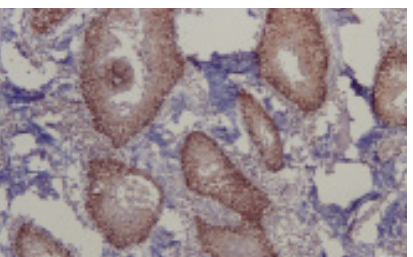


Figure 9: p 63 (IHC×400).

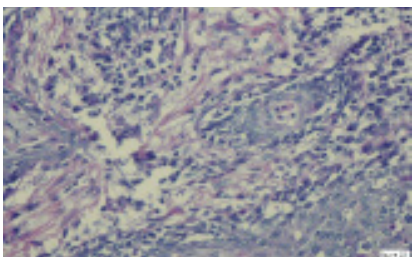


Figure 10: PAS (IHC×400).

## Discussion

Intraosseous carcinoma has been observed in patients with 1.3 to 90 years old (mean age of 30 years). There is a male predilection. The primary symptoms are swelling, pain, tooth pain and rapidly growing mass; however, delayed symptoms can be more serious such as trismus and anesthesia [13]. An important subtype of intraosseous carcinoma is squamous cell carcinoma which is known as keratinization of tumoral cells [12], the carcinoma is typically invasive and has a very poor prognosis [15]. Although the carcinoma mostly affects posterior area of mandible, its solid form has been reported in maxilla [3]. This carcinoma can primarily originate from the jaw bones and additionally the epithelial lining of the odontogenic cysts, and the rests of Malassez [10-13]. In radiographic features, the lesion causes radiolucency with moth-eaten borders [3]. Mitsuyoshilino et al [20-13] reported a case of intraosseous squamous cell carcinoma in Japan that developed in the maxilla of a 36-year-old Japanese man [3]. Shintaro and Sukegawa [20-15] described a case of intraosseous squamous cell carcinoma in the maxilla originating from an infected residual cyst [10]. If the lesion originates in the bone, patients' prognosis and overall survival are poor, so early and accurate diagnosis help proper treatment [15]. Squamous cell carcinoma is more common in individuals over 50 years old and if it occurs in younger individuals, it has a more invasive behavior and has a poorer prognosis; therefore, clinical examination, diagnosis and analysis in these patients are highly recommended [7]. Histopathologic grading of squamous cell carcinoma is based on the similarity of the tumoral cells to normal squamous epithelium and the amount of keratin production. Lesions are graded on a three-point scale; well differentiated, moderately differentiated and poorly differentiated squamous cell carcinoma [1,16]. Low-grade or well differentiated tumor is mature enough to resemble tissue of origin and often grows slowly and metastasizes later in its course. In contrast, a tumor with marked pleomorphism and little or no keratin production that often enlarges rapidly, metastasizes early are termed high-grade or poorly differentiated or anaplastic [1].

Differential diagnosis includes ameloblastic carcinoma, central mucoepidermoid carcinoma and a malignant variant of calcifying epithelial odontogenic tumor. Similarity to ameloblastoma, such as peripheral palisading of peripheral cells has been shown in some intraosseous squamous cell carcinomas [19]. The negativity for Ck 7 in ameloblastic carcinoma is diagnostic [20]. If central mucoepidermoid carcinoma is suspect-

ed, mucin stains should be performed. Intraosseous SCCs usually do not contain mucin-producing cells. Malignant variant of calcifying epithelial odontogenic tumors may show features of SCC, but they also have some diagnostic features, such as amyloid deposition and Liesegang ring calcifications [19].

Cytokeratin-7 (CK-7) is a protein that in human is encoded by the KRT7 gene which is specifically expressed in the simple epithelial lining of internal organs and in glandular ducts and blood vessels [17]. The p63 is a nuclear transcription marker used in the identification of tumor subtypes such as squamous cell carcinoma [1]. Ki-67 is a proliferation marker and higher proliferative activity is confirmed by higher Ki-67 expression [21]. Keratin 20 is a type I cytokeratin. It is a major cellular protein of mature enterocytes and goblet cells which is specifically found in the gastric and intestinal mucosa [18]. Periodic acid-Schiff (PAS) staining is mainly used for staining structures containing glycogen, glycoprotein and proteoglycan that typically are found in mucus and basal laminae [22].

Intraosseous squamous cell carcinoma is considered a highly malignant tumor which must be treated aggressively [19]. Unfortunately, due to the socioeconomic status the patient refused to cooperate; Therefore, the precise evaluation is not possible.

### Conclusion

Tumors of the head and neck are important group of malignancies with their prevalence increasing worldwide [17]. The present study discussed a case of intraosseous squamous cell carcinoma that is important due to the young age of the patient and lesion progress.

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