Oral Diag. 2020;05:e20200020.

CASE REPORT

Case report on multiple solitary radiolucencies in the mandible - a diagnostic dilemma

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Abstract:

Radiolucent mandibular lesions seen on panoramic radiographs develop from both odontogenic and non-odontogenic structures and represent a broad spectrum of lesions with a varying degree of malignant potential. Here, we report a case of a forty-five-year-old female patient with a complaint of pain and pus discharge from the left side of the face. Based on the characteristic clinical appearance, a diagnosis of chronic osteomyelitis was made. However, on digital panoramic radiography, a rather unusual radiolucent appearance in the ramus and coronoid process of the mandible was observed. Several laboratory investigations and advanced diagnostic imaging techniques were carried out to narrow down the differential diagnosis, which was crucial for the identification of this lesion which was diagnosed as Primary Intraosseous Carcinoma. The objective of this clinical case report is to highlight this unusual radiographic appearance to aid in future diagnosis.

Keywords: Primary intraosseous carcinoma; mandibular radiolucency; multiple solitary radiolucencies; digital panoramic radiography; cone beam CT

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Article received on August 11, 2020.
Article accepted on September 28, 2020.

DOI: 10.5935/2525-5711.20200020

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INTRODUCTION

Primary intraosseous carcinoma (PIOC) describes a squamous cell carcinoma arising primarily from the jaw bone having no initial connection with oral mucosa, and presumably developing from residues of the odontogenic epithelium. Approximately 150 cases of PIOC have been documented till now.[1,2]

The tumour was first described by Loos in 1913 as a central epidermoid carcinoma. Several other terms have also been used including intra-alveolar epidermoid carcinoma and primary intra-alveolar epidermoid carcinoma. Pindborg et al. in 1971 were the first to use the term PIOC.[3,4,5,6]

It has been further classified by Waldron and Mustoe as Type 1: PIOC ex odontogenic cyst, Type 2a: Malignant ameloblastoma, Type 2b: Ameloblastic carcinoma arising denovo, ex ameloblastoma or ex odontogenic cyst, Type 3: PIOC arising denovo and Type 4: Intraosseous mucoepidermoid carcinoma.[7]

The definite diagnosis of PIOC is often considered difficult as the lesion must be distinguished from squamous cell carcinoma that may invade the bone from the overlying soft tissues or from the tumours that have metastasized to the jaw from a distant site.[8] PIOC most commonly presents as swelling and persistent pain in the mandible, or may be asymptomatic and discovered on routine panoramic radiographs.[9,10] The present report illustrates PIOC arising in the posterior mandible with an unusual radiographic appearance.

CASE REPORT

A forty-five-year-old female patient came to us with the complaint of pain in the left back cheek region since six months. She gave history of pain sudden in onset, severe, continuous, which was relieved on medication. It was associated with pus discharge from the cheek and difficulty in opening the mouth and difficulty in speech and swallowing since 6 months. She also gave history of weight loss in 6 months. Her personal history revealed she had the habit of chewing 5 packets of tobacco per day since 20 years.

On extra oral examination, a single left submandibular lymph node was palpable, 1 centimetre (cm) in diameter, firm, mobile and non-tender on palpation. A diffuse swelling was seen over left side of cheek measuring 2×3 cm, extending superoinferiorly from the ala tragal line to the lower border of the mandible and mediolaterally, 3cm from the commissure of the lip to the angle of the mandible. A sinus opening was present 1cm above lower border of mandible.

There was no local rise in temperature. Tenderness was present on palpation. The swelling was firm, non-fluctuant and fixed to underlying tissue. Pus discharge was expressed on palpation.

Intra oral examination revealed the mouth opening was restricted to 5 mm. Left lateral movement of tongue was restricted. Grade III mobility was present in relation to 36,37. No mucosal ulceration was seen irt. 36 and 37. No further examination could be done due to reduced mouth opening (Figure 1).

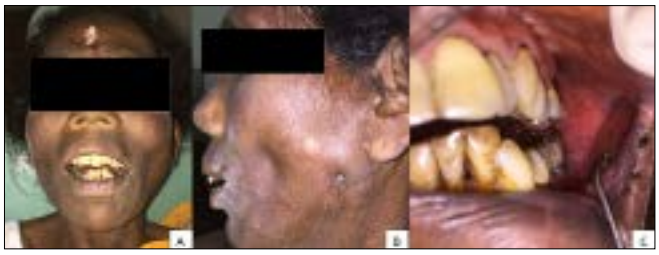

Figure 1. Clinical photograph revealing (A) restricted mouth opening, (B) Sinus opening 1cm above left lower border of mandible and (C) absence of mucosal ulceration

Based on the history and clinical examination a provisional diagnosis of chronic osteomyelitis in relation to the left ramus region and generalised periodontitis was made. Differential diagnosis considered were odontogenic infection, cellulitis, deep fungal infection and carcinoma of left alveolus/ ramus.

Investigations carried out were pus culture and sensitivity testing which revealed moderate growth of Pseudomonas aeruginosa. A digital panoramic radiograph (OPG) was taken. The OPG revealed two separate well defined radiolucencies in the left coronoid process and left ramus of the mandible. Generalised horizontal bone loss with radiolucency involving the furcation in relation to 17,26,27,36,37. Deep dentinal caries was seen in relation to 16,17,18,26,27,28,36,37,47 with loss of lamina dura in 37; 38 was missing (Figure 2).

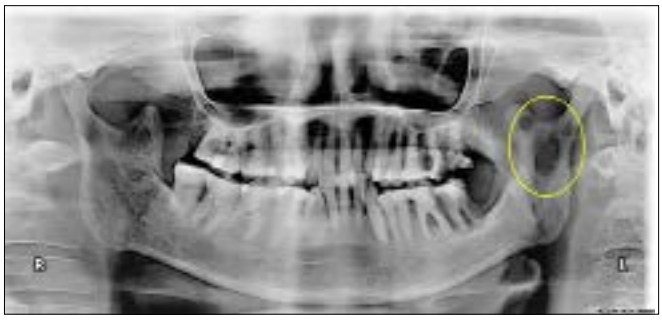

Figure 2. Pre- operative OPG revealing two separate well defined radiolucencies in the left coronoid process and left ramus of the mandible.

Radiographic differential diagnosis of Multiple myeloma, metastatic malignancy with secondaries in posterior mandible and multiple odontogenic keratocysts were considered.

The patient underwent a variety of examinations such as Lateral skull radiograph, Serum albumin globulin ratio, Bence- Jones protein, Chest X-ray and Abdomino-pelvic sonography, which were all normal (Figure 3).

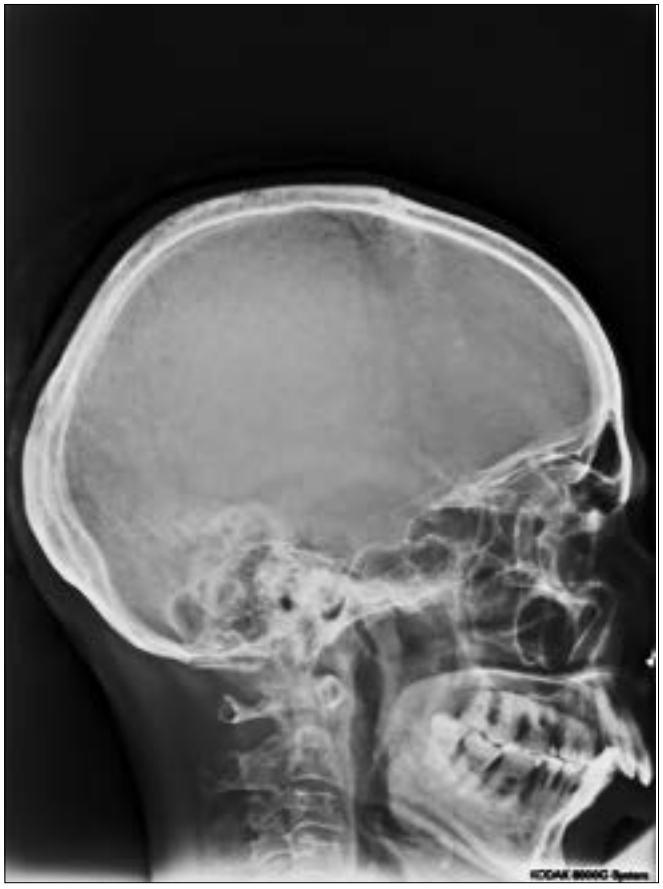

Figure 3. Lateral skull taken to rule out multiple myeloma.

A cone beam CT (CBCT) was advised to further evaluate the lesion. The CBCT revealed two well defined lesions measuring 13 and 6.7mm in the ramus and coronoid process of mandible. (Figure 4).

Incisional biopsy could not be performed due to the proximity of the lesion to the parotid gland and it was decided to proceed with an excisional biopsy followed by histopathology. A segmental resection of the left condyle, coronoid and ramus was performed under general anesthesia (Figure 5). The microscopic section showed strips of stratified squamous epithelium with proliferating round to polygonal cells. These cells showed abundant eosinophilic cytoplasm with large, vesicular nucleus with irregularly clumped chromatin

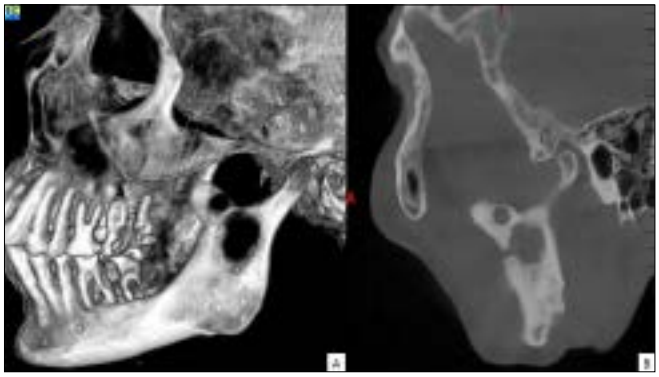

Figure 4. CBCT of the patient revealing two solitary well defined lesions measuring 13mmand 6.7mm in the ramus and coronoid process of mandible respectively.

and prominent nucleoli. Occasional mitotic figures were seen with centres of proliferating cell masses show keratin pearls. Sections from medial pterygoid and masseter showed fibrocollagenous tissue and muscle infiltrated by tumour. Sections from the parotid showed stratified squamous epithelium with proliferating islands of tumour tissue with central keratin pearls. Perineural invasion was present. Two lymph nodes exhibited reactive hyperplasia and sinus histiocytosis. Sections from sinus opening shows ulcerated stratified squamous epithelium with granulation tissue. The histopathological report was indicative of well – differentiated Squamous Cell carcinoma.

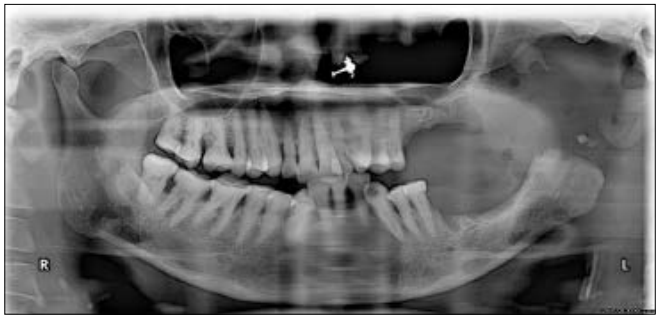

Figure 5. Post-operative OPG revealing segmental Resection of the left condyle, coronoid and ramus.

Based on the findings the case was diagnosed as primary intraosseous carcinoma of the left mandible. The patient was referred to the Department of Oncology for post –operative Radiotherapy. Six months of follow up showed no evidence of a primary tumour or recurrence.

DISCUSSION

To define a lesion in the jaws as PIOC, 3 specific criteria may be present: (1) Histological evidence of

squamous cell carcinoma, (2) Absence of ulcer formation on the overlying mucosa, and (3) Absence of a distant primary tumour at the time of diagnosis and at least six months during the follow-up period.11,12,13 The possibility of metastasis in the present case was eliminated by a careful history and comprehensive systemic evaluation. Furthermore, an intact mucosa made the possibility of direct extension of squamous cell lesions from the oral mucosa appear unlikely. Hence, the tumour described in this paper completely fulfilled the aforementioned criteria. 14

PIOC is commonly seen in the 6th to 7th decade with a mean age of 57 years. It affects men more than women with a ratio of 3:1. However, our patient was a female of 45 years. The tumour commonly occurs in the mandible with a striking predilection for posterior regions and presents with pain, swelling of jaws, trismus, sensory disturbances such as paraesthesia.15,16 Our case had an additional clinical feature of a sinus opening. This may have been due to the odontogenic infection from the mandibular second molar as seen in the OPG (Figure 2).

Radiographic examination of PIOC shows great variation in size and shape, and in the appearance of the borders. Nolan reported that PIOCs that grew slowly had a well-defined, smoothly contoured bordesr, while those that grew more rapidly showed a poorly defined, ragged border. Because the margins of PIOCs show great variation, they may resemble benign or malignant tumours. Our case had two rather well-defined, unilocular radiolucencies in the ramus and coronoid process of the mandible which resembled cysts. 17

Surgery is the treatment of choice and in most cases consisted of enbloc excision or radical resection of involved bone. Radiotherapy and chemotherapy are considered in lesions that cannot be surgically controlled.11

Variable radiographic features of PIOC and its resemblance to odontogenic cysts and tumours, emphasize that PIOC should be considered in the differential diagnosis of radiolucent lesions of jaws.2

CONCLUSION

The importance of this case is that it illustrates a rare radiographic appearance of primary intraosseous carcinoma. The diagnosis of such a lesion is challenging, and precise clinical and pathological details are important in deciding the treatment plan. As oral diagnosticians, the sole responsibility we have in case of PIOC is early diagnosis for which sufficient knowledge of this rare entity is a necessity.

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