

Case Reports

SOLITARY PLASMACYTOMA OF THE JAWS REPORT OF TWO CASES

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مرض في هذا البحث حالتين نادرتين للورم ذي الخلايا المصورة والذي يؤثر على الفكين حيث إن هناك (٢٧) حالة مثبتة في غضون ٢٠ عاماً. والمظاهر السريرية تكون عبارة عن ناتج غير مؤلم وبدون أي علامات إشعاعية في أكثر المناطق تأثراً في الفك السفلي هي المنطقة الوسطى من الفك. والمعالجة تتم بواسطة الجراحة، الأشعة العميقة، والعلاج المشترك ويجب عمل فحص دوري لإتمام المعالجة وحتى يتم السيطرة على المرض. وفي هذا البحث ناقش حالتين تم اكتشافهما في دبلن بأيرلندا، إحداهما في الفك السفلي والأخرى في الفك العلوي حيث تمت المعالجة للحالة الأولى بواسطة الجراحة، وفي الحالة الأخرى بواسطة الأشعة العميقة. وفي اعتقادنا أن الحالة المثبتة في الفك العلوي هي الثالثة من نوعها.

Solitary plasmacytoma is a rare condition affecting the jaws with only 25 reported cases in the mandible and two cases in the maxilla over the last 20 years. The most common site of involvement in the mandible is the premolar - molar region. Patients usually present with a firm, painless swelling with non-specific radiographic features. Monoclonal immunoglobulin in both serum and urine, combined with the histology, confirm the diagnosis. Treatment is either surgery, radiotherapy or a combination of both. Although the prognosis is good, there is a risk of developing multiple myeloma, hence long-term follow-up is necessary. This paper presents two cases of this anomaly, one in the mandible and one in the maxilla. We believe that the maxillary case is the third reported in this position. The mandibular case is also of interest as it is the eleventh case to have been treated with surgery alone.

Introduction

Plasma cell neoplasms usually present as a disseminated condition (multiple myeloma) involving multiple bony sites.¹ A solitary lesion is

an uncommon entity representing only 3-5% of all plasma cell tumors.² The most common sites of involvement of a solitary form of plasmacytoma are the body of the vertebrae and the pelvis.³ Plasmacytoma of the jaw is rare with only 27 reported cases⁴ of which 25 occurred in the mandible and 2 in the maxilla.^{5b} In this report, two cases of solitary plasmacytoma of the jaws are presented, one in the mandible and another in the maxilla for which literature was reviewed.

Case 1

A 46-year-old male was referred to the National Maxillofacial Unit at St. James Hospital in Dublin, Ireland complaining of swelling on his left jaw of a one-year's duration. The swelling had suddenly

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enlarged 6 weeks prior to presentation and was interfering with his lower denture. He also noticed his chin had deviated to the left [Fig. 1]. The patient's medical history was non-contributory and physical examination revealed no other abnormality. On examination, a swelling measuring 4cm x 4cm x 3cm was noted at the left angle of the mandible. It was bony hard, non-tender and obliterated the buccal sulcus and was not attached to the overlying mucosa or skin. There was no numbness of the lower lip and regional lymph nodes were not palpable. Computerized tomographic examination showed a large multiloculated lesion in the left body of the mandible with expansion of the buccal cortex [Fig. 2]. A clinical diagnosis of ameloblastoma was made and a biopsy was performed. Microscopic examination showed bone with extensive infiltration by sheets of plasma cells, with some atypical and binucleate forms [Fig. 3]. The cells demonstrated lambda light chain restriction (mouse anti-human monoclonal N10/12, Dako), confirming the diagnosis of plasmacytoma. The patient was investigated further for multiple myeloma. Serum and urine protein electrophoresis disclosed a monoclonal peak (M-protein), identified as IgG immunoglobulin with a lambda light chain. No Bence-Jones protein was seen. A skeletal radiographic survey showed no other lesion apart from the osteolytic lesion of the mandible. After consultation with the Oncology Department, it was decided to treat this isolated lesion by surgery. A hemimandibulectomy with preservation of the condyle was undertaken. The mandible was reconstructed using a Dacron urethane tray filled with cancellous bone harvested from the iliac crest. The patient made an uneventful recovery and was discharged from the hospital one week later. In the resection specimen [Fig. 4], there was a circumscribed 2.5 cm fleshy yellow mass expanding and almost replacing the entire circumference of the mandible with focal extension into adjacent soft tissue. Microscopically, the appearances were similar to the biopsy. Also noted was a perineural and intraneural invasion by the plasma cell infiltrate in the soft tissue. Serous red stain for amyloid was negative. Resection margins were free of involvement. The patient is now 23 months postoperative with no clinical or biochemical evidence of plasma cell dyscrasia. Postoperative orthopantomograph showed a satisfactory reconstruction of the left side of the mandible [Fig. 5].

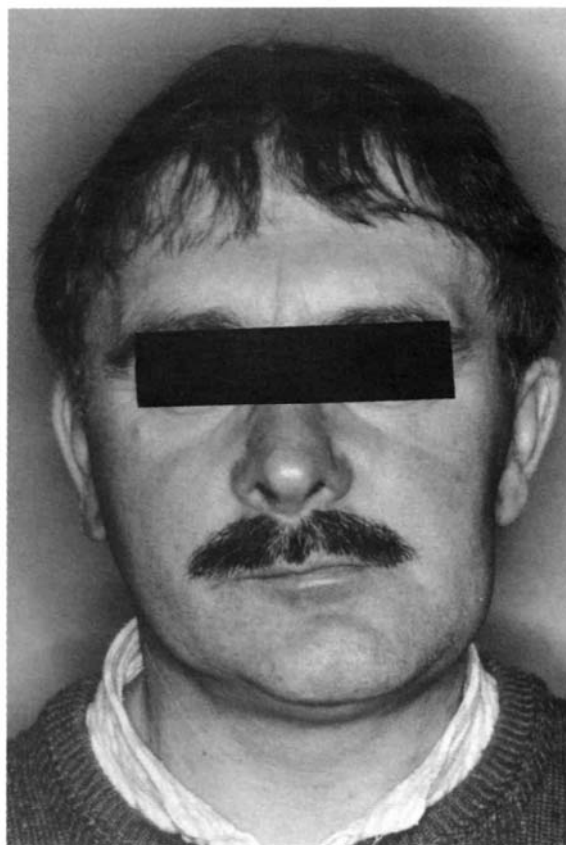


Figure 1. Clinical presentation with deviation of the mandible to the left.

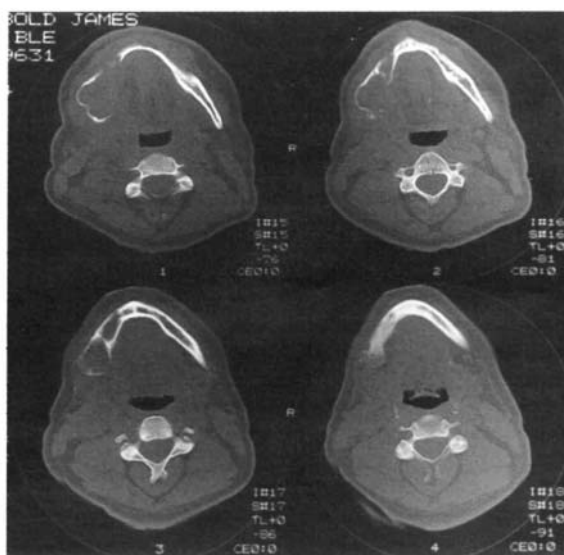


Figure 2. CT scan showing multilocular radiolucency in the left mandible with buccal expansion.

Case 2

A 53-year-old male was referred to the Oral Surgery Department at Dublin Dental Hospital in Ireland complaining of a right sided facial swelling and associated nasal obstruction of three years duration. The patient's medical history was

non-contributory and physical examination showed no other abnormality. On examination, an obvious swelling was noted on the right cheek which extended into the right nose and obliterated the buccal sulcus. It was firm and non-tender. Radiographic examination showed an extensive radiopacity involving the right maxilla, obliterating the antrum and extending into the nose [Fig. 6]. A biopsy was performed and microscopic examination confirmed the diagnosis of plasmacytoma. There was no radiographic or biochemical evidence of multiple myeloma. After consultation with the Oncology Department a decision was taken to treat this patient by local curettage and radiotherapy. The patient received a local field radiation with a total dose of 3500 cGy in 25 fractions over 30 days. The patient is now 10 years postoperative with no clinical or biochemical evidence of a recurrence or of a multiple myeloma.

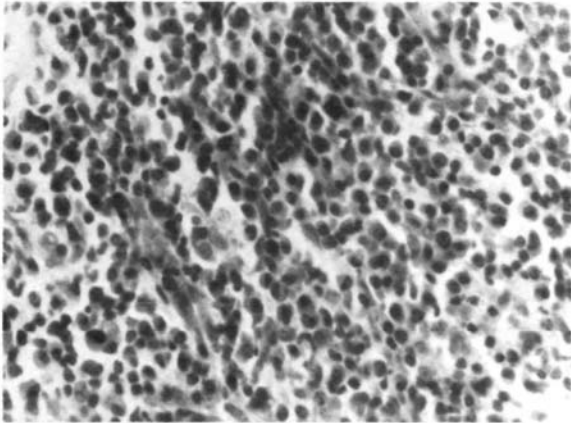


Figure 3. Photomicrograph of Case 1 showing dense cellular infiltrate, many with typical plasmacytoid features.



Figure 4. The resection specimen of Case 1 showing the expanding fleshy mass.

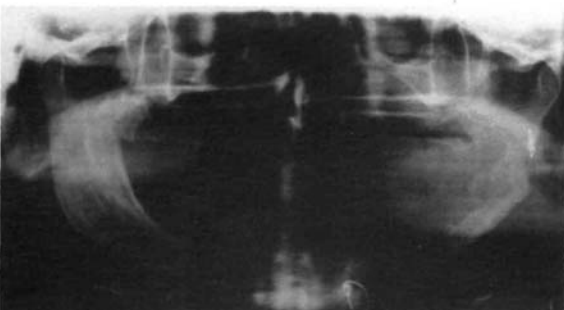


Figure 5. Postoperative OPG showing the grafted right mandible.

Discussion

Multiple myeloma is the most common type of plasma cell malignancy.¹ Other types include



Figure 6. Radiograph of Case 2 showing an extensive radiopaque lesion involving the right maxilla and extending into the nose.

extra-medullary plasmacytoma and solitary plasmacytoma. Solitary plasmacytoma of the jaws is rare. Review of the literature over the last 20 years showed a total of 25 reported cases in the mandible^{4,6} and two cases in the maxilla with the most common site being the premolar-molar region. Involvement of the anterior region is rare with only one case being reported.⁴ Two cases of solitary plasmacytoma has been reported in the maxilla.⁹ The criteria essential for the diagnosis remain similar to those developed by Bichel and Kirketerp¹⁰: the histology of the lesion, the skeletal survey to eliminate the presence of other foci, a negative bone marrow biopsy, serum and urine electrophoresis and immunocytochemistry. The histology of the lesion is predominated by a dense sometimes atypical plasma cell infiltrate, in which light chain reaction is evident. Serum M-protein has prognostic significance reflecting tumor activity and should, therefore, be checked regularly. Treatment may be surgery, radiotherapy or a combination of both.

Our literature review showed that only 11 cases had been treated by surgery alone.¹¹ Of the other cases, three were treated by radiotherapy and 13 cases had combined therapy.⁴ Progression of the tumor to multiple myeloma after treatment has been reported in two cases, both of which had combined therapy.^{9,12} Accordingly, long-term follow-up is recommended.^{7,8}

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