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Case Report

Solitary Plasmacytoma of the Mandible: Report of a Rare Case

Adebayo Ibikunle[1*], Abdurrazaq Taiwo[1], Ramat Braimah[1], kabir Abdullahi[2], Michael Adeyemi[3]

1. Department of Dental & Maxillofacial Surgery, Usmanu Danfodiyo University Teaching Hospital, Garba Nadama Road, Sokoto, Nigeria.

2. Department of Histopathology, Usmanu Danfodiyo University Teaching Hospital, Sokoto, Nigeria , Aliyu Quarters, Sokoto.

3. Department of oral & maxillofacial Surgery, Lagos University Teaching Hospital, Lagos, Nigeria. Staff quarters, Lagos university Teaching Hospital, Lagos, Nigeria.

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Solitary bone plasmacytoma belongs to a group of lymphoproliferative clinical disorders typified by clonal proliferation of plasma cells. Solitary plasmacytoma typically involves a single bone or extramedullary site. It commonly occurs in the vertebrae or thorax, and rarely involves the mandible. The aim of this report is to present a rare case of solitary plasmacytoma of the jaws mimicking periodontal disease. A solitary plasmacytoma involving the right mandibular ramus/angle was observed in a 60-year-old woman, which was a coincidental finding following persistent excessive post-extraction hemorrhage. The patient was referred for radiotherapy, and subsequent follow up has been uneventful. Although solitary plasmacytoma of the mandible is a rarity, it may occur nonetheless. It may mimic periodontal disease; hence clinicians should have a high index of suspicion when managing patients.

Keywords: Solitary bone plasmacytoma, lymphoproliferative disorders, mandible

Solitary bone plasmacytoma (SBP), multiple myeloma (MM), and extra medullary plasmacytoma (EMP) are a group of lymphoproliferative clinical disorders typified by clonal proliferation of plasma cells (1, 2). They may be systemic or localized in nature (2). MM is a disseminated form of plasmacytoma which is relatively more commonly encountered than its solitary form (2, 3). Localized plasmacytomas may be solitary plasmacytoma (SP) which denotes single bone involvement or EMP which signifies involvement of the soft tissues (2, 4). SBP commonly manifests as spinal disease, frequently affecting the thoracic vertebrae (2). Other bones such as the sternum, clavicle, rib and humerus may be involved, but that of the mandible is a rarity (3, 4).

Patients might present with pain, tooth mobility, severe bleeding or pathological bone fractures (2, 4). SBP may progress to become systemic MM; this has been observed to occur within 3 years in as much as two-thirds of cases (3). Regardless of its course, it has significant effect on the patients' quality of life, and mortality rates are still significant (5). We report a case of a 60-year-old female who was managed on account of severe hemorrhage following routine extraction; a diagnosis of solitary plasmacytoma of the mandible was subsequently made. To the best of our

*Correspondence: Department of Dental & Maxillofacial Surgery, Usmanu Danfodiyo University Teaching Hospital, Garba Nadama Road, Sokoto, Nigeria. E-mail: adebayoibikunle@gmail.com

knowledge this is the first report from Sub-Saharan Africa.

Case presentation

A 60-year-old female presented at our out-patient clinic with toothache of 2 weeks duration. The pain was said to be spontaneous, throbbing, and intermittent. It was aggravated by mastication. She had had an uneventful extraction of the tooth #48, and medical history was found to be non-contributory. The patient was not pale, not jaundiced, and not febrile. There was no obvious facial asymmetry, and the patient's interincisal mouth opening was 3.3 cm. Grade III mobility, and tenderness to both lateral and vertical percussion of tooth #47 was observed. Periapical radiograph showed periradicular radioluscency in association with the tooth #47.

Forceps extraction of the #47 was done under local anaesthesia; hemostasis was achieved, and postoperative instructions delivered. However, the patient presented at the clinic after 13 hours with significant post-extraction hemorrhage, which she claimed commenced about 4 hours after hospital discharge. On examination, she was slightly clinically pale, mouth opening was satisfactory, but there was a persistent oozing of blood from the extraction socket of tooth #47, which was recalcitrant to digital pressure. An iodine soaked, adrenaline impregnated gauze strip was inserted into the extraction socket, and secured in place with a horizontal mattress suture.

Thereafter, hemorrhage subsided, and an immediate packed cell volume (PCV) was determined, which gave a value of 22%. Additionally, she was placed on 200 mg ferrous sulphate tabs, as well as 100 mg vitamin C tabs, both 8 hourly for 2 weeks, and 100 mg paracetamol tabs, 8 hourly for 3 days.

The patient was reexamined after 24 hours, no sign of ongoing hemorrhage was observed, and the suture and gauze dressing were removed. The patient was then sent for postero-anterior radiograph of the skull, which revealed a unilocular radiolucency involving the right mandibular angle/ramus region with poorly defined borders (fig. 1). The radiolucency extended postero-anteriorly from the angle of the right mandible in close proximity to the extraction socket of the right mandibular third molar to just apical to the roots of the right mandibular first premolar (fig. 1).

A test needle aspiration which was negative was performed to evaluate the vascularity of the lesion. An incisional biopsy of the radiolucent lesion was done via an intra-oral access under local anaesthesia.

Histological examination of the specimen revealed sections of highly cellular tumor sheets, composed of medium sized cells with hyper-chromatic round to ovoid nuclei, which were irregular, and eccentrically located (fig. 2 A-D). The cytoplasm was basophilic to amphophilic in nature, with occasional binucleation (fig. 2 A-D). Immunohistochemical studies were negative for both keratin and CD45 (fig. 3 A and B).

Values for electrolyte, urea/creatinine (EU/Cr), and white blood cell (WBC) count were within reference ranges. Skeletal survey with plain radiographs of the spine, chest, pelvis, and long bones were performed to rule out the presence of bone lesions. No other osteolytic bone lesions were

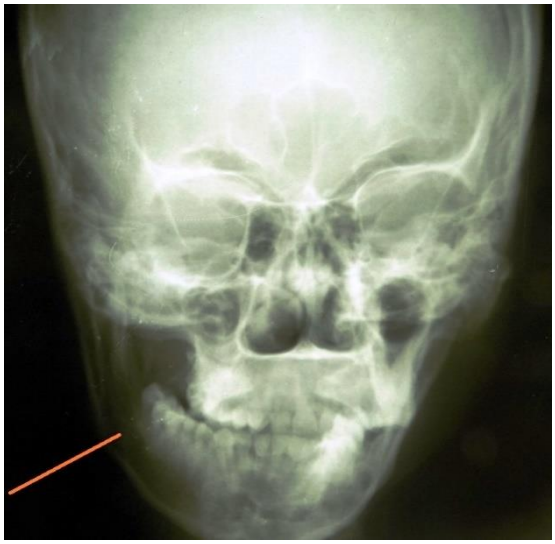

Figure 1. Postero-anterior radiograph of the skull. The arrow indicates the radiolucent lesion.

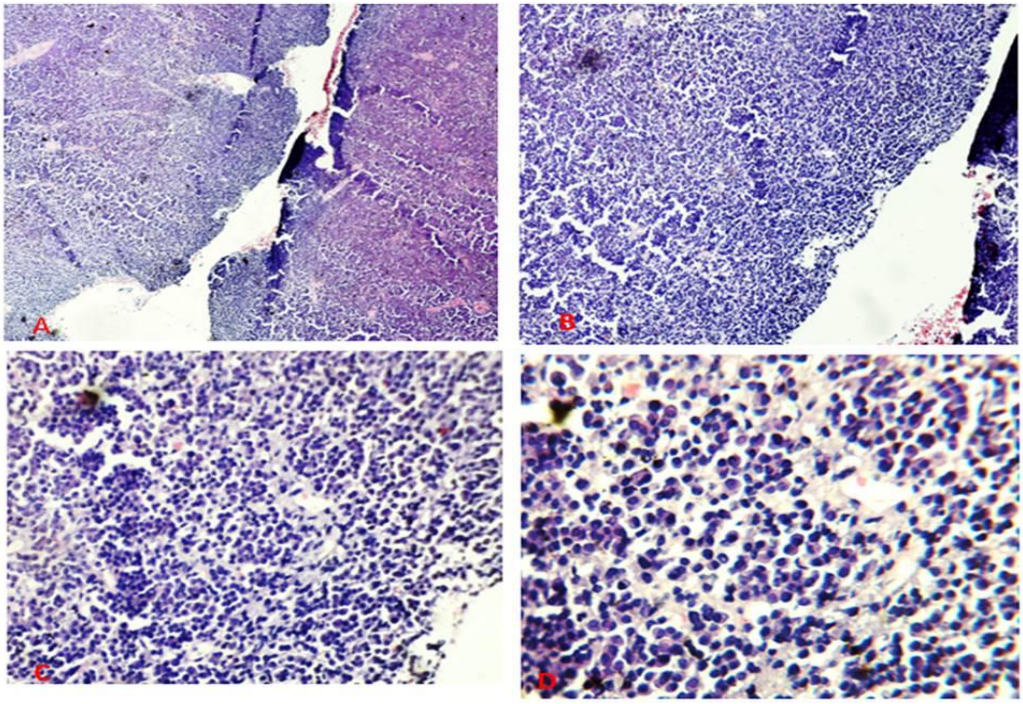

Figure 2. Histological examination of the lesion. High density cellular regions with hyperchromatic nuclei, and basophilic to amphophilic cytoplasm are observed.

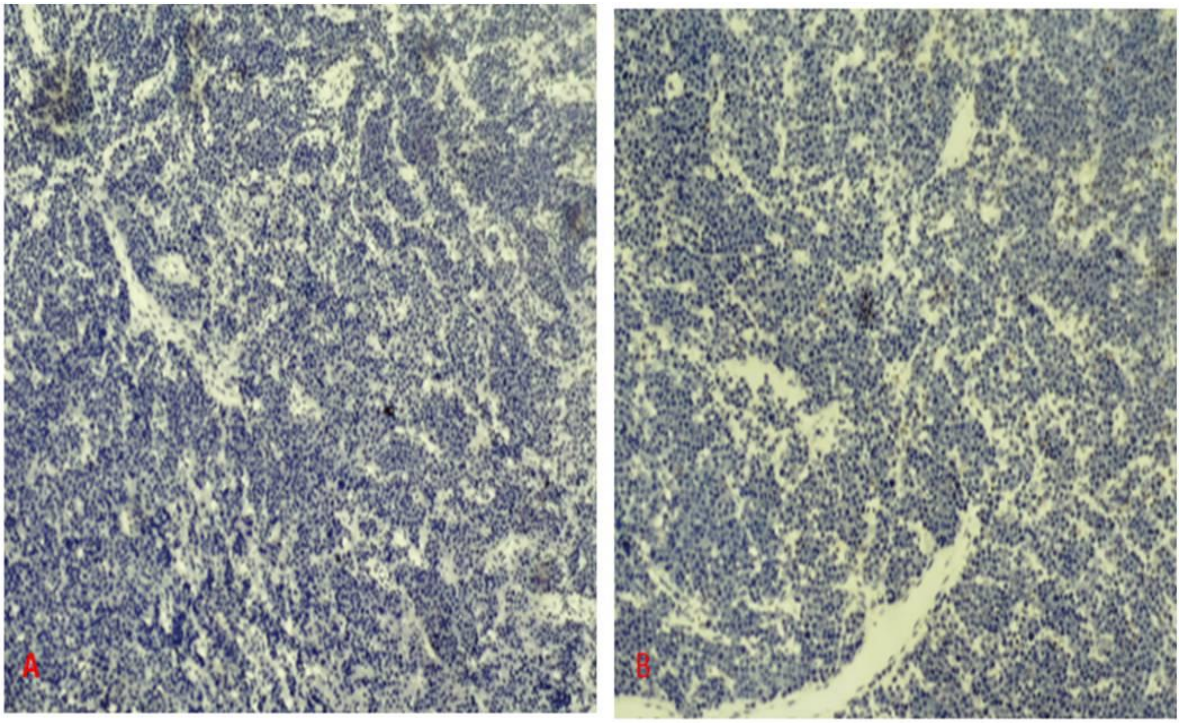

Figure 2. Immunohistochemical examination of the lesion. A: keratin; B: CD45. Both markers were negative.

seen on radiographs. Positron emission tomography (PET) scan was requested for, but was not procured due to financial incapacitation. Urinalysis was negative for Bence Jones proteins. Consequently, a diagnosis of solitary plasmacytoma of the mandible was made, and she was referred for radiotherapy. Consent was obtained from the patient for treatment, and for publication of this report.

Discussion

SBP is a localized malignant clonal proliferation of plasma cells (2, 6). It represents 3-10% of all plasma cell tumors (1). It is commonly seen among patients in the 5th and 6th decades of life, and has a male preponderance with a ratio of 2:1 (1). Although the patient reported in this case is a female, her age correlates with reported cases in the literature (2, 7). The most common sites for SBP are the vertebrae, long bones, and skull (1-3). SBP of the mandible is a rare event. However, the specific site of occurrence in the mandible in this patient (angle/ramus) compares favorably with mandibular sites reported in the literature (3, 4). SBP is believed to have a penchant for marrow rich areas of the mandible such as the ramus, and angle of the mandible (2, 4).

Frequently encountered signs and symptoms of mandibular involvement include pain, paraesthesia, bony expansion, spontaneous bleeding, and tooth mobility (2, 4). Pain and tooth mobility were the principal features observed in this case, which were suggestive of localized chronic periodontal disease. No obvious bony expansion was noted, which may signify an antero-posterior rather than a bucco-lingual growth of the tumor. Additionally, postoperative bleeding was also encountered. Although, easy fatigability and/or fever have been described in some patients (4), they were not recorded in this case. Typically, SBP presents radiologically as a radiolucency, which may be unilocular or multilocular (2, 4, 8). In the present case a unilocular radiolucency with poorly defined borders was observed, the inferior part of the anterior ramal border was involved, signifying extension into the overlying soft tissue.

Bone imaging is a relevant investigation for diagnosing solitary plasmacytoma. Options that have been utilized include skeletal survey with plain radiographs, computed tomography (CT) scan, magnetic resonance imaging (MRI), and PET scan (1, 8). Skeletal survey with plain radiographs was the imaging investigation of choice in this report because of financial incapacitation and/or non-availability of facilities. Plain radiographs are less expensive than CT scan, MRI and PET scan, and it has been shown to possess greater ability to detect cortical bone lesions than MRI (6, 8, 9). Bone marrow biopsy has also been used in characterizing this disease with absence of clonal plasma cells considered to be in agreement with a diagnosis of SBP as opposed to systemic myeloma (10).

Apart from imaging, diagnosis of SBP also relies on histology (2, 7, 11). Typically, histology reveals sheets of atypical plasma cells exhibiting varying degrees of differentiation, nuclear eccentricity, inclusion bodies, and cartwheel chromatin pattern (1, 2, 4, 7, 11). This description is akin to the histological picture observed in this case.

Differential diagnoses of SBP may include osteolytic metastasis, osteoid osteoma, and osteoblastoma (10, 12). Metastatic lesions are often osteolytic in nature, often resulting in localized radiolucency that imitate SBP (10, 12). Osteoid osteoma is a solitary benign osseous lesion that frequently affects long bone cortices, and typically presents with history of nocturnal pain that is relieved by the use of non-steroidal antiinflammatory drugs (12). Osteoblastoma is a rare benign osseous lesion that is characterized by bone formation in the presence of abundant osteoblasts (12). Patients typically present with a painful bony swelling (12). History, examination, and investigations such as the use of imaging techniques, biopsy, and the use of special stains are helpful in differentiating SBP from other lesions (10, 11). Advanced diagnostic techniques, such as flow cytometry studies, and heavy and light chain deposits assessment are gaining popularity (11).

Treatment of SBP may be radiotherapy, surgery or a combination of the two (1, 2). Some researchers recommend radiotherapy as the mainstay of treatment; reverting to surgery only when there is an excellent chance of tumor removal with minimal aesthetic or functional compromise (2, 3). The role of chemotherapy is guarded, and some

authors have limited its usefulness to disseminated diseases (6). This patient was referred for radiotherapy. Dimopoulus et al. reported that a significant relief of symptoms was achieved in all cases, while local control of the disease was achieved in 90% of cases (3). No report of local failure was observed by Frassicca et al. in their series of 46 patients with SBP, who were given 4,500 cGy or more through radiotherapy (10). However, few of these studies were based on SBP of the mandible.

Patients with SBP require constant follow up, especially due to the chance of disease progression into MM (2). Other known patterns of failure are occurrence of new lesions, and local recurrence of the lesion (10). As much as 85 % of SBP evolve into MM within months to years post diagnosis (4, 9). Increased risk of progression into MM is associated with age greater than 60 years, and tumor size of greater than 5 cm (4). The 10 year survival rate of SBP is 50-80%. However, recurrence worsens prognosis significantly (4).

In conclusion, although, solitary plasmacytoma of the mandible is a rarity, it may occur, masquerading as periodontal disease. Therefore, clinicians should have a high index of suspicion, and meticulously evaluate patients for optimal treatment.

Conflict of interest

The authors declare that they have no competing interest.

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