

Solitary Plasmacytoma Involving Maxilla Treated by Multi-disciplinary Approach

SMA SADAT^a, N KHAN^b, J FARDAOUS^c, K GOLDER^d, SN RITA^e

Abstract:

Solitary plasmacytoma, a B-lymphocyte proliferating neoplastic rare entity can involve facial soft tissue and bone. The presenting case showed diffuse swelling in left mid face with maxillary bone destruction. The lesion was diagnosed as solitary plasmacytoma by conventional histopathology and immunohistochemistry. The case was successfully treated by multi-disciplinary approach with neo adjuvant

radiotherapy followed by wide surgical excision without any relapse in three years follow up.

Key words: Solitary Plasmacytoma, Plasma cell Myeloma, Plasmacytoma in maxilla, Neo adjuvant radiotherapy, partial maxillectomy

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Introduction:

Plasmacytoma can be defined as neoplastic proliferation of B-lymphocytes of bone marrow stem cells¹. The proliferation results in destruction of bone adjacent to the mass with cloning and multiplication of immunoglobulin secreting cells². Plasmacytoma occurs during the differentiation from primary and secondary B-lymphoblast cells to pathologic plasmablasts in the stage of maturation³. Plasma cell neoplasms can be classified into: Solitary Plasmacytoma of Bone (SPB), Extramedullary Plasmacytoma of soft tissue (EMP) and Multiple Myeloma⁴. Solitary plasma cell myeloma or solitary plasmacytoma converts into multiple myeloma in 50% cases³. Solitary plasmacytoma in maxilla has an infrequent incidence predominantly on males and at the age of 50-80 years⁵. The early manifestations of solitary

plasmacytoma in maxilla are pain, swelling and obstruction in oral cavity due to proliferative tissue mass and oedema⁶. In maxilla adjacent teeth can be affected due to the surrounding bone destruction⁷. Usually in case of solitary plasmacytoma in maxilla systemic proliferation is non evident⁸. The aetiology of SPB is unknown though chronic stimulation of radiation, viral disease, and genetic factor of reticulo endothelial system can be considered to initiate the tumour formation⁹. For diagnostic purpose, radiological and histopathological investigations are done. Neo adjuvant chemotherapy followed by surgery is preferred for management of the case. Here we are reporting a case of rare solitary plasmacytoma of maxilla of a Bangladeshi male patient who was successfully treated with multi-disciplinary approach.

Report of the Case:

A 35-year-old man, otherwise healthy, non-smoker reported to ENT and Maxillofacial Surgery department of Square Hospital Ltd on October 2020 with the complain of painful swelling on left mid face for one year. He had history of several tooth extraction in abroad with discharge of fluid from extraction socket. On physical examination, diffuse non tender, firm swelling was found on left midface (Fig 1) and intra orally, the lesion was diffuse, soft, non-tender in the palate extending from upper left 2nd premolar to upper left 2nd molar (Fig 2). There was missing of two molars and mild mobility of 3rd molar tooth with otherwise moderate oral hygiene.

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Figure 1: Extra oral front and inferior nasal view showing diffuse swelling in left mid face



Figure 2: Intra oral view showing swelling in the left palate

Initially, Orthopantomogram was done for the radiological evaluation. Irregular bone destruction as ill-defined radiolucency was seen on the left maxillary alveolar bone and maxillary walls (Fig 3).

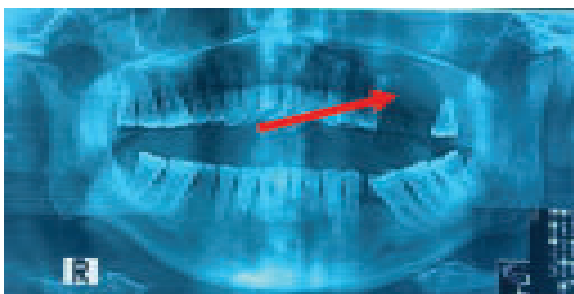


Figure 3: Orthopantomogram showing ill-defined radiolucent lesion in left maxilla

Computed Tomography (CT) scan of maxillae revealed fairly large strong heterogeneous enhancing soft tissue density mass lesion on the region of left maxillary sinus, left nasal cavity with extension upward into ethmoid air cells with destruction of medial lateral inferior and anterior walls and roof of the maxillary antrum (Fig 4).

The mass extended towards the left face. No definite intra orbital extension was identified.

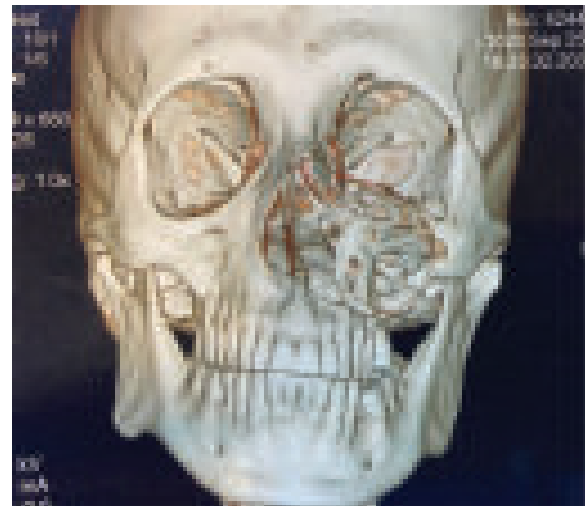


Figure 4: 3-D CT scan showing irregular bone destruction in left maxilla

Later on, suspecting as malignant lesion, incisional biopsy was done for histopathological evaluation. Specimen was collected from tissue of left maxillary alveolar ridge swelling. Specimen consisted of two small grey-white pieces of soft tissue measuring 0.7 cm and 0.6 cm in maximum diameter respectively. Sections showed soft tissue and large sheets of a hyper cellular tumour. The tumour was composed of small cells with round nuclei and moderate amounts of eosinophilic cytoplasm. Several cells had eccentrically placed nuclei imparting a plasmacytoma morphology. Occasional cells had large round nuclei with distinct nucleolus resembling a plasmablast. Occasional mitotic figure was seen (Figure 5). The tumour cells showed moderate to strong immunopositivity for CD 138 (Figure 6). All haematological values including CBC, ESR, Serum Creatinine, SGPT were within normal range. Chest Xray revealed normal findings. Bone marrow examination revealed secondary marrow mass without any evidence of Multiple Myeloma. A medical board consisting of Maxillofacial Surgeon, Haematologist and Oncologist recommended neo adjuvant radiotherapy followed by maxillectomy for the patient. Accordingly, Radiotherapy was given as 25 fractions each 200 cGy which was completed on 30.11.2020.

After periodic CT scan, residual lesions were identified and so partial maxillectomy was done on 30.9.2021

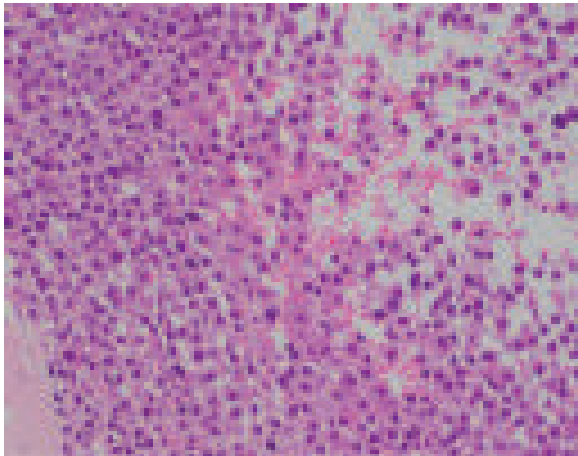


Figure 5: Histopathology (Haematoxylin & Eosin Stain) showed features suggestive of plasmacytoma

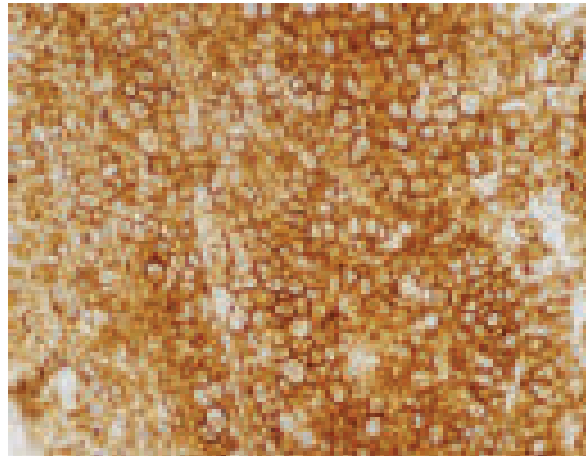


Figure 6: Immunohistochemistry shows moderate to strong Immunopositivity for CD 138

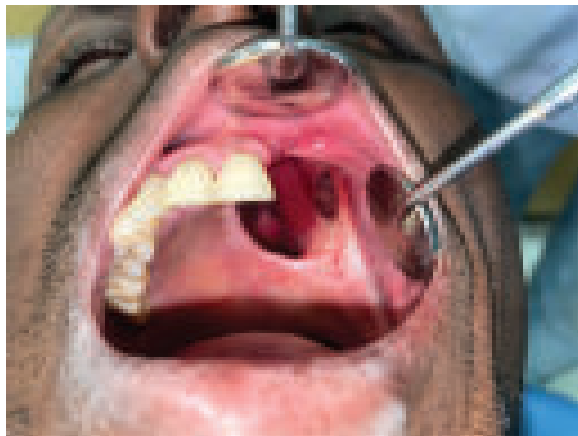
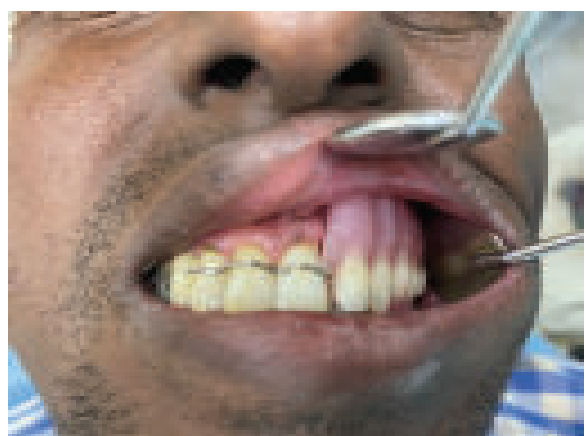


Figure 7: Post-Surgical Intra-oral defect



(Figure 7). After complete healing, acrylic denture with obturator was constructed for rehabilitation (Figure 8). Patient was in close follow up with oncologist and maxillofacial surgeon. Till October 2023, there was no evidence of recurrence clinically and radiologically and no evidence of dissemination of disease.

Discussion:

Solitary plasmacytoma is a neoplastic proliferation of monoclonal plasma cells without systemic characteristic of malignancy^{10,11}. It can be a result of low-grade progression of multiple myeloma in 3-10% of cases^{12,13,14}. Nasal cavity, paranasal sinuses, nasopharynx, palatal and alveolar region of maxilla are the frequent sites of solitary plasmacytoma of head neck region¹⁵. There are two types of light chain

immunoglobulin i.e.: κ - chain and λ - chain. κ : λ ratio is examined to monitor the monoclonal cell proportions and density along with maturity and stage of the tumor¹⁰. Clinical behaviour of Solitary Bone Plasmacytoma is relatively benign. The progression is slower than Extra Medullary Plasmacytoma (EMP) with approximately 70% of lesions converts into multiple myeloma^{1,3}.

In the present case, the origin of plasmacytoma was unknown. The reported symptoms were fullness in left face below eye with mild aching pain. Usually, the symptoms appear late and the lesion is painless until the secondary infection occurs^{10,16,17}. Nasal septum was deviated towards right side and patient did not show any evidence of epistaxis. Upper 1st and 2nd molar on the left side were missing. The prevalence of Solitary

Plasmacytoma of Bone (SPB) is more in male than female with a ratio of 2:1^{5,18}. In histopathological findings, tumor cells contained round nuclei and eosinophilic cytoplasm and occasionally placed nuclei with distinct nucleolus resembling a plasmablasts. The tumor cells showed moderate to strong immunopositivity for CD 138.

In a study of Etebarian et al¹², a 59-year-old male had painful slowly growing mass in the upper jaw which enlarged right after four months of second molar extraction. The histochemical studies showed plasma cell rich tumor positive for CD 138 in lambda light chain and negative for CD 45, CD 56 in kappa chain.

Another systematic review by Shirani et al.^u showed a 42-year-old woman had medical history of swelling in left zygomatic area treated with antibiotics; and dental granuloma with chronic abscess around porcelain fused metal bridge teeth was treated surgically. Due to misdiagnosis of the disease, patient had to undergo unnecessary Root Canal Treatment and bone augmentation following curettage. Later the case was diagnosed as plasmacytoma histologically.

In a study of Nguyen et al.^{1p}, a 37-year-old male had obstruction and pain in nasal sinus, as the tumor was of soft tissue origin (EMP). In histopathological report CD 45 staining showed positive result. The cells had large nuclei with prominent nucleoli and high nucleus-cytoplasm ratio. Cartwheel or clock face is the common terminology for the typical heterochromatin arrangement of plasma cells.

In our present case treatment option was neo-adjuvant radiotherapy followed by surgery. In a case report of Shirani et al.^u two-field conventional radiotherapy with extraction of tooth no 26 were performed. In another case, Etebarian et al¹². performed four sessions of chemotherapy with Bortezomib plus Dexamethasone (BD therapy) and also 10 sessions of radiation with planned dosing of 40 - 50 Gy.

Conclusion:

Solitary plasmacytoma can be specifically evaluated by both histopathology and immunohistochemistry. It can be successfully treated by multi-disciplinary approach with neo adjuvant therapy followed by wide surgical excision with reference to the reported case. Long term follow up should be considered to exclude its dissemination and also to see the recurrence and need for further adjuvant therapy.

Consent: Consent was taken from patient

Conflicts of interest: There is no conflict of interest

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