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UNUSUAL CASE OF OSTEOBLASTOMA IN AN ELDERLY FEMALE: A CASE REPORT AND A REVIEW

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ABSTRACT

REVIEW ARTICLE

Osteoblastoma is a rare entity encountered as a benign osseous tumor with the propensity of occurrence towards the vertebral column and long bones. Commonly occurs in young middle-aged males with mandible as a site of predilection in the facial skeleton. Differential diagnosis of osteoblastoma holds a challenge in attaining a final diagnosis. Osteoid osteoma, cementoblastoma, osteosarcoma, aggressive fibrous dysplasia, and ossifying fibroma share a similar clinical behavior, radiological and histological behavior. This article aims to report a case of osteoblastoma of an aggressive variety appearing in the maxilla. It aims to gain a better understanding of the clinical, histological, radiological, and differential diagnosis of osteoblastoma. A review of English language literature in 2006 revealed 43 previously reported cases of osteoblastoma appearing in the maxilla and mandible since 1967. 16 cases including this one are added to the previous well-documented review of literature by Jones where he included additional 24 cases to the previously reported 43 cases. Brief discussion and review on treatment modalities are analyzed.

INTRODUCTION:

Osteoblastoma is a rare bone tumor traversing varied nomenclature and constant ambiguity historadiologically, accounting for 1% of all primary bone tumors. It was first mentioned in 1932 in a case report by Jaffe and Meyer titled 'Osteoblastic osteoid tissue-forming tumor of a metacarpal bone'[1.] Then various reports under the terminologies like "giant osteoid osteoma"[2] and "osteogenic fibroma of the bone" [3] have appeared in the literature. Eventually, the proposal of "benign osteoblastoma" nomenclature is achieved by Jaffe and Lichtenstein independently in 1956.[4]

It is a benign, slow-growing bony neoplasm characterized by the appearance of plump osteoblasts rimming the newly formed osteoid and bony trabeculae, rapidly increasing in size and number, in a well-vascularized fibrous stroma.

Demographic evidence supports its appearance more commonly in males with an average age being 20.4 years. Various reports claim osteoblastoma's inclination in males,[5,6,7]

but Jones additional cases in his highly illustrated review find female predilection. Lucas in his clinicopathologic study of 306 cases stated "virtually every bone in the body is affected" but the vertebral column bears the major brunt along with the sacrum accounting for 32% of the cases.[5] Its occurrence in the skull and jawbones are relatively rare and represent only 15% of all osteoblastoma.[6] El lofty in his review of the literature finds slight predilection for the lesion to occur in the mandible.[7.]

A constant uncertainty regarding the tumor's diagnosis lies in its similar yet not identical characteristics to osteoid osteoma. Osteoblastoma's propensity to occur surrounding the roots of the teeth raises uncertainty in its diagnosis with cementoblastoma as well. Lucas's retrospective study mentions the challenge of distinguishing between a few histological varieties of osteoblastoma and osteosarcoma. Such variation led to the adoption of "aggressive osteoblastoma" terminology for tumors mimicking a few characteristics of malignancy.

The clinical picture encompasses swelling, pain, and expansion of the involved bone. The radiographic appearance is extremely fluctuating depending on the degree of calcification in the lesion, may appear radiolucent or semi radiolucency with radiopaque mottling and well-demarcated margins.[8]

The radiologic assessment highlights mixed radiolucent/radiopaque lesions with more or less defined borders, and these usually lack sclerotic borders, periosteal reactions, or perpendicular bone speculations. In the presence of such findings, osteosarcoma shouldn't be ignored as one of the differential diagnoses. [9]

Histologically the tumor is characterized by enlarging the number and size of osteoblasts (plump osteoblasts) generating and rimming the haphazardly arranged trabeculae of osteoid and woven bone. Well vascularised stroma comprising scattered trabeculae and "new blue bone", plump osteoblasts, few multinucleated cells, and chronic inflammatory cells adjacent vascular spaces.[8]

This article aims to report a case of osteoblastoma of an aggressive variety appearing in the maxilla. It aims to gain a better understanding of the clinical, histological, radiological, and differential diagnosis of osteoblastoma. A review of English language literature 2006 revealed 43 previously reported cases of osteoblastoma appearing in maxilla and mandible since 1967, the year of the first reporting of osteoblastoma arising in jaws by Borello and Sedano.[10,11] 16 cases including this one is added to the previous well-documented review of literature by Jones where he included additional 24 cases to the previously reported 43 cases.

CASE REPORT:

A 60-year-old woman presented to our facility of the oral and maxillofacial surgery department, suffering from 'bearable pain' and swelling on the right side of her face. She stated that she has been aware of a gradually progressive swelling on the right side of her maxilla for over six months. No contributory medical history was recorded. Family history and past anamnesis are noncontributory too.

Clinical workup revealed a moderate built and well-nourished woman with fairly well-demarcated, mildly tender swelling of approximately 4.5 cm in diameter involving the molar region of the maxilla. The skin overlying the tumor presents with normal color and texture. Intraoral examination revealed poor periodontal status and relatively demarcated mildly tender swelling involving right maxillary canine, premolars, and molars extending till the maxillary tuberosity. On palpation, the swelling was of bone-like hardness and the cortical bone in the area was expanded both buccally and palatally with teeth involved in the lesion were mobile.

On conventional radiographs, it presents as a well-defined, elliptical radiolucent lesion comprising dense irregular radiopaque foci located at the periapical region of the right maxilla. The lesion was well depicted, with a faint sclerotic margin. A computed tomographic scan helped us with hypoechoic focus involving the right maxilla marking the dimensions of 3.5cm and 4.5 cm anteroposteriorly and mediolaterally. A speckled pattern of calcification is appreciated through varied densities on the CT scan. Some degree of sclerotic change was appreciated adjacent to the lesion. A provisional diagnosis of a benign bone tumor was considered.

The patient was admitted to the hospital, and surgical intervention involving complete en-bloc resection of the lesion was performed under general anesthesia. Surgical specimen included irregular pieces of hard and soft tissues which were submitted for histopathological examination.

Histopathological examination revealed a circumscribed lesion showing lamellar bone and trabeculae of mature bone enclosing the connective tissue. The bone appears mature with well-formed resting lines with lacunae containing osteocytes and lined by osteoblastic rimming. The soft tissue appears fibrous and cellular at a few races showing spindle and stellate shaped cells with mild inflammatory infiltrate showing predominantly lymphocytes and plasma cells. Engorged and dilated endothelial lined blood capillaries with giant cells were evident too. The surface epithelium is parakeratinized stratified squamous epithelium.

Our patient made an uneventful recovery without any recurrence postoperatively with a follow up of one year.

DISCUSSION.

Benign osteoblastoma is a rare variant of bone tumor which poses challenges to the attainment of a stable diagnosis clinically and histologically. Histologically it presents with the proliferation of plump osteoblastic cells rimming the trabeculae of osteoid and immature bone in a vascularized stroma. There is a mild predilection for males, young age as well as mandible. Though our case report stays antonymous to the predilections. The first detailed case report of an osteoblastoma of the jaw was published by Sedano and Borello in 1967.[10] Though Farman credited Greiner for the first mention of 'fibro-osteoblastoma' of the maxilla in a five-year-old.[1112] El mofty et al published a report reviewing 26 cases of osteoblastoma with the addition of their case where he stated the clinical picture stands nonspecific where only 20 of 27 cases presented with painful tender swelling. Benign osteoblastoma may be classified into cortical, medullary, and periosteal types. Gnathic osteoblastoma is either medullary or periosteal, but not cortical as in retrognathic sites.[7] Asada in 1991 surveyed 44 cases from English and Japanese literature in his article where he illustrated his case report showing the first multicentric occurrence.[13]He adopted the terminology of 'desmo-osteoblastoma' from Makek's classification for tumor's histological variations.[14] The radiographic features of benign osteoblastoma are not diagnostic, ranging from a well-defined radiolucency or radiopacity or a combination of both depending on the degree of calcification. Computed tomography can be used as an adjunct in attaining diagnosis to conventional radiography.

Osteoblastoma tends to be aggressive and predisposed for recurrence. These features make it difficult to distinguish these lesions from low-grade osteosarcoma. Various clinical behavior led to the origin of inconsistent terminologies in the past like 'malignant osteoblastoma'[15], 'pseudomalignant osteoblastoma'[16] and osteoblastoma like osteosarcoma.[17]

Mayer in 1967 described aggressive osteoblastoma as a different entity from conventional osteoblastoma with an inclination

towards recurrence. It is histologically characterized by the presence of epithelioid osteoblasts which are larger than their conventional counterparts.[18] Dorfman et al. further defined this variant as having features resembling those of osteoblastoma and low-grade osteosarcoma, as well as a low mitotic activity but no atypical mitosis. It lies on the continuum between conventional osteoblastoma and low grade osteosarcoma.[19]

The close relationship between variants of osteoblastoma and different bone tumors cannot be ignored. Its close clinical and histological association with osteoid osteoma needs to be mentioned as one of the differential diagnoses. Old literature used both the tumors synonymously, but subsequently, authors understood the unwise attempts of blurring the whole different clinical entity. Differences between the two can be stated based on tumor size and the presence of tumor nidus. Osteoid osteoma hardly attains a larger size than 1.5 cm. Similarly, association with cementoblastoma needs to be elucidated as well. The emergence of osteoblastoma around roots of teeth blurs the diagnosis and confuses the examiner with cementoblastoma. Most of the hard tissues produced by the tumor cells were osteoid and osseous tissue therefore, excluding cementoblastoma from our differential diagnosis. The lesion was easily differentiated from osteosarcoma because of the lack of malignant features such as nuclear atypia and abnormal mitoses and the 'non-aggressive' lifespan of the tumor. Histologically, the sclerosing form of osteosarcoma in which osteoid and new bone production are prominent and cellular atypia is not pronounced may create an ambiguity towards a diagnosis of osteoblastoma. The presence of cartilage favors a diagnosis of osteosarcoma since it is only found in osteoblastoma if there is a pathologic fracture, another rare event.[17]

Bertoni believed that the feature which separates osteosarcoma from osteoblastoma is the permeation of tumor margins into adjacent tissues which is never seen in osteoblastoma or any variants.[15]

Harrington commented on the microscopic differential diagnosis of osteoblastoma where he deliberately differentiated between conventional, aggressive, and osteoblastoma like osteosarcoma variety based on histological features.[20]

Radiographically lesions fluctuate between completely radiolucent to a mixed pattern with varying degrees of calcification to a complete radiopaque pattern. These variations can be allotted to the 'age of tumor' or could be the possibility of 'already-programmed lesions to produce more calcified products'. This review attempts to ponder on different clinical symptoms and rare sites of occurrence of osteoblastoma. The patient presented with a lesion located on the articular tubercle misdiagnosed as the temporomandibular joint disorder is a rare event reported by Emanuelsson.[21] An aggressive case of an osteoblastoma of the nasal cavity that invaded the anterior skull base in a 3-year-old girl, requiring resection via an intracranial approach probably represents another case of the aggressive variety.[22]

Eisenbud reported an incidence of spontaneous regression of osteoblastoma after the biopsy.[23]Smith described an osteoblastoma in which, despite an incomplete removal of the tumor tissue, the area of bone with definite evidence of tumor involvement was replaced by normal-appearing bone.[24]

Recurrence of osteoblastoma of jaws accounting for 9.8% in a review of 181 cases of osteoblastoma affecting the whole body by Jackson puts conservative treatment as a suboptimal option for the management of this tumor. Complete resection of the tumor followed by long term follow up provides the exemplary treatment option.

Table 1: Reviewed cases of osteoblastoma in the jaws

Borello and Sedano[10]	21	Male	Left posterior maxilla	Pain, swelling	Radiopaque
Kramer[25]	6	Female	Left posterior mandible	Pain, swelling	Expansion, osteolytic areas, periosteal layering
Kent et al[26]	13	Female	Right posterior maxilla	Discomfort, swelling	Well circumscribed radiopaque lesion, loss of lamina dura
Kopp[27]	19	Male	Left coronoid process	Asymptomatic	Radiolucent
Brady and Browne[28]	19	Male	Mandibular symphysis	Pain, swelling	Radiolucent with speckled radiopacities
Smith[24]	7	Male	Left mandible	Pain, facial asymmetry	Spherical mass, irregularly calcified
Yip and Lee[29]	22	Female	Left maxilla	Pain, swelling	The circumscribed radiolucent area with central radiopacity
Remagen and Prein[30]	15	Male	Left posterior mandible	Pain, swelling	Well delineated radiopacity
Farman et al[11]	9	Male	Right mandible	Pain, swelling	Radiolucent with radiopaque material
Greer and Berman [31]	30	F	Anterior mandible	Tender swelling	Poorly circumscribed shadow
Chatterji et al[32]	30	F	Left maxilla	Pain, swelling	Well circumscribed soft-tissue shadow
Hatakeyama and Suzuki[33]	14	M	Left anterior maxilla	Pain, swelling	Radiolucency with central radiopacity—“sunburst” appearance
Nowparast et al[34]	14	F	Left mandible	Pain, swelling	Well-demarcated, osteolysis, and osteogenesis
Sidhu et al[35]	13	F	Left mandible	Asymptomatic, swelling	Diffuse radiolucency
Danielidis et al[36]	15	F	Right ramus	Pain, swelling	Circumscribed, translucent lesion
Miller et al[37]	37	F	Right ramus	Pain	Circumscribed radiopaque/radiolucent lesion
	6	M	Left mandible	Pain, swelling	Circumscribed radiopacity with central radiopaque nidus
Monks et al[38]	19	F	Left posterior mandible	Pain, swelling	Well circumscribed radiolucency

Smith et al[39]	21	M	Left posterior maxilla	Pain	A radiolucent-radiopaque lesion, moth-eaten appearance
Van Der Waal et al[40]	20	F	Right posterior maxilla	Pain, swelling	Well circumscribed radiopacity
Shatz et al[41]	17	F	Right posterior mandible	Pain, swelling	A radiopaque lesion with central radiolucent zone
Uma et al[42]	13	F	Left posterior mandible	Pain, swelling	A radiopaque lesion with the radiolucent band and sclerotic rim
Eisenbud et al[23]	11	F	Anterior mandible	Tender, swelling	Diffuse mottling thickened periodontal ligament
Weinberg et al[43]	19	M	Right mandibular condyle	Pain, tenderness	Enlarged condylar head
Colm et al.[44]	35	M	Right posterior mandible	Pain, swelling	A radiolucent lesion, margins well-demarcated and sclerotic
Ohkubo et al[45]	6	M	Left posterior maxilla	Pain, swelling	Ill-defined radiopacity
El-Mofty and Refai[7]	11	M	Left maxilla	Tender, swelling	Well demarcated mottled opacity
Strand-Pettinen et al[6]	20	F	Left posterior mandible	Asymptomatic, swelling	Well circumscribed radiolucency; root resorption
Haug et al[46]	35	F	Left ramus, condyle, and coronoid process	Pain, swelling	Well demarcated radiolucent lesion
Asada et al[13]	38	F	Right posterior mandible	Asymptomatic, swelling	Well defined radiolucency with radiopaque foci
Guest and Juniper[47]	26	M	Left posterior maxilla	Pain	Radiopaque
Svensson and Isacsson[48]	14	M	Left ramus and condyle	Pain, swelling	Radiolucent
Ataoglu et al[49]	23	M	Left posterior mandible	Pain, swelling	Well circumscribed radiolucency with irregular radiopacities
Peters et al[50]	16	M	Left anterior mandible	Discomfort	Multilocular radiolucent/radiopaque lesion
Rasse et al[51]	20	M	Left condyle	Pain, swelling	Mottled radiolucent/radiopaque

					lesion
Ribera MJ[52]	69	M	Right anterior maxilla	Pain	Radiolucent
Ahmed and Nwoku[8]	17	M	Right ramus and condyle	Pain, swelling	Well defined semiradiolucent lesion
Gordon et al[53]	19	F	Left posterior maxilla	Pain, swelling	Radiopaque
Öztürk et al[54]	21	F	Left posterior mandible	Pain, swelling	Well circumscribed opacity with a lucent rim, root resorption
Ufuk et al[55]	23	F	Right temporomandibular region	Pain, swelling	Radiolucent
JONES[9]	25	F	Right posterior mandible	NS	Radiopaque
JONES[9]	30	M	Left maxilla	Swelling, bone expansion	Radiolucent
JONES[9]	25	F	Anterior mandible	Lingual bone perforation	Radiolucent
JONES[9]	27	F	Right posterior mandible	Asymptomatic	Radiopaque
JONES[9]	34	F	Left posterior maxilla	Tender	Ground glass opacification
JONES[9]	36	M	Anterior mandible	Asymptomatic, bone growth—lingual cortex	Normal
JONES[9]	14	F	Left posterior maxilla	Pain, expansion	Normal
JONES[9]	61	M	Right posterior mandible	Bone growth—buccal cortex	Radiolucent/radiopaque
JONES[9]	22	F	Anterior mandible	Tender, swelling, bone expansion	Radiolucent/radiopaque, ill-defined
JONES[9]	37	F	Left posterior mandible	Lingual expansion	NS
JONES[9]	25	F	Left posterior mandible	Tender, expansion	Radiopaque
JONES[9]	15	M	Left posterior mandible	Buccal expansion	Multilocular, radiolucent
JONES[9]	21	F	Right posterior mandible	NS	NS
JONES[9]	24	F	Left posterior mandible	NS	Well circumscribed, radiolucent
JONES[9]	3	F	Right posterior	Asymptomatic,	Radiolucent/radiopaque

			mandible	expansion	
JONES[9]	23	F	Left posterior mandible	Pain	NS
JONES[9]	37	F	Left posterior mandible	NS	Radiolucent
JONES[9]	24	F	Left posterior maxilla	Palatal swelling, expansion	Radiolucent/radiopaque
JONES[9]	53	F	Left posterior mandible	Asymptomatic, swelling	Radiopaque
JONES[9]	14	M	Right mandible	Asymptomatic	Radiolucent/radiopaque
JONES[9]	11	F	Right posterior mandible	Pain	Radiolucent, focal opacifications, destruction of lingual cortex
JONES[9]	25	F	Anterior mandible	Pain, swelling	Perforation of the buccal cortex, calcifications
JONES[9]	25	F	Left maxilla	Pain, swelling	Radiolucent/radiopaque, well-demarcated
JONES[9]	26	F	Right posterior mandible	Asymptomatic	Radiolucent
Adair and Kashtwari[56]	14	M	Left side posterior mandible	Asymptomatic	Radiopaque
Akram and Bashel[57]	25	M	Right hemimandibular swelling	Asymptomatic	MIXED
Kashikar[58]	18	M			
Tiago and Silva[59]	27	F	Left Posterior mandible	Swelling	Radiopaque
Harrington and Accurso[20]	25	M	Left Palatal mass	Asymptomatic	A radiolucent lesion with radiodensities
Lypka[61]	10	M	Left mandible	Pain and swelling	Mixed lesion
Patel[62]	19	M	Right maxilla	Asymptomatic	Mixed lesion

CONCLUSION

Intricate similitude in histology, radiology, and clinical picture puts the diagnosis of a tumor in the hazy background. With unpredictability in etiology of the relatively benign course of the tumor puts the examiner through a difficult course. Literature suggests trauma or trauma after extraction or infection could be an etiology of change of course of the normal physiology of bone leading to the formation of a tumor. To conclude, our case with a judicious surgical plan and follow up didn't present with any recurrence. In a race to achieve a swift and correct diagnosis of osteoblastoma, close cooperation of an oral and maxillofacial surgeon, radiologist and pathologist is required for an adequate surgical treatment of this benign tumor.

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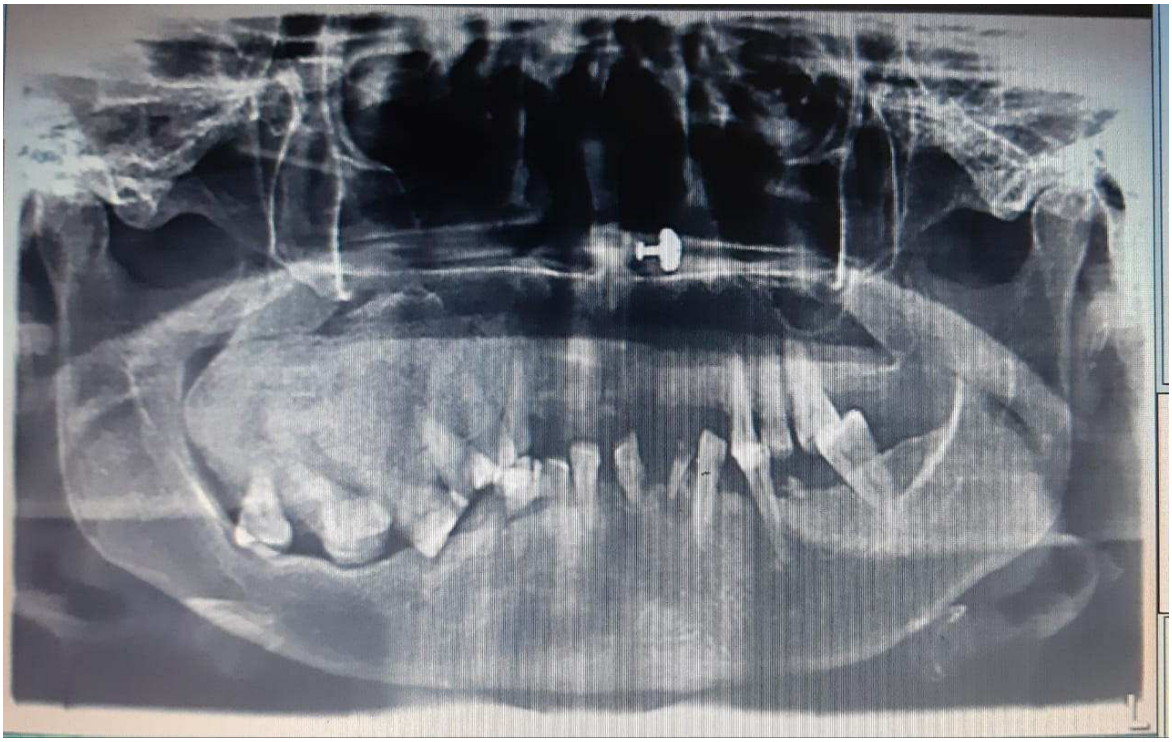

Figure 1: An orthopantomogram showing well defined radiopaque lesion on the right maxilla posterior segment causing extreme deformity of right maxilla and mobility of left maxillary posteriors.

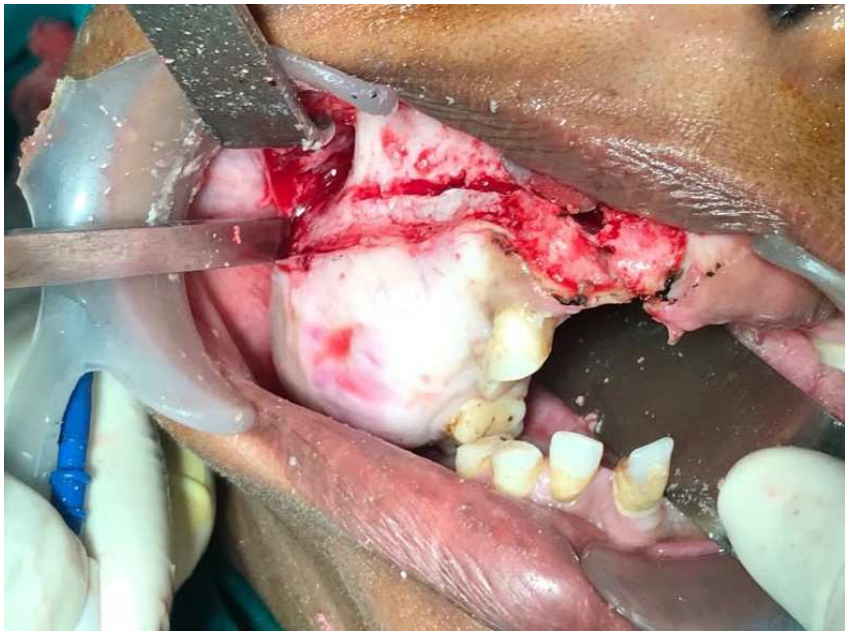

Figure 2: Intraoperative image showing segmental resection done for osteoblastoma.

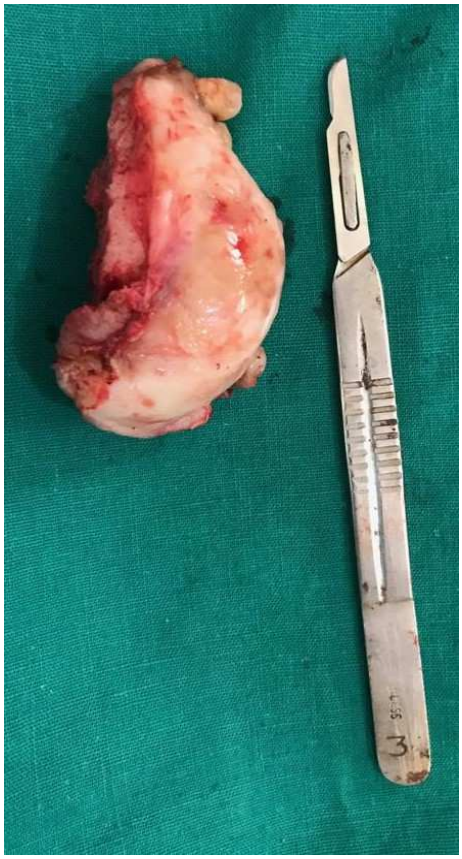

Figure 3: Excised specimen after segmental resection of the tumor.

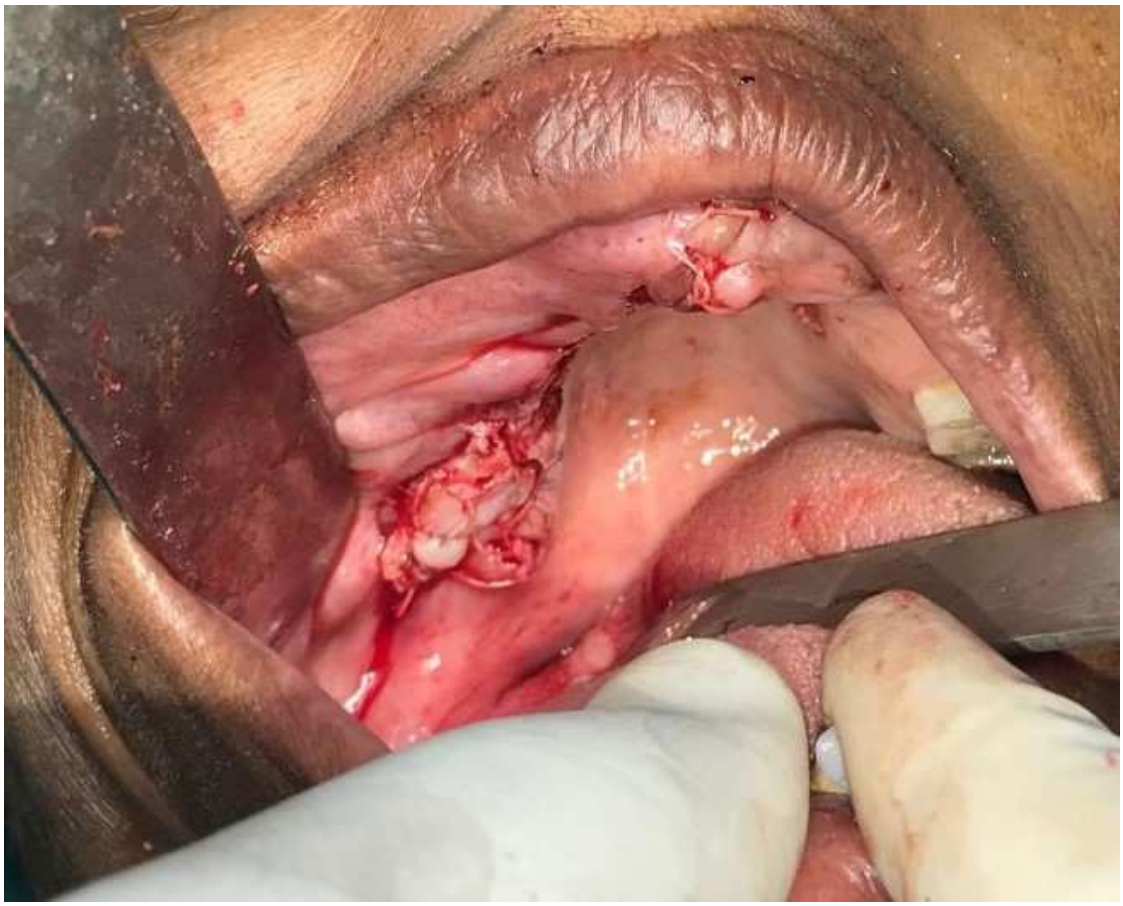

Figure 4: Primary closure achieved.