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Original Research Article

Brown Jaw Tumors: challenges and outcomes

Tarek Elsayed Ftohy Abdelrahman*

Maxillofacial, Head and Neck Surgery Unit, Sohag University Hospital, Egypt

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***Correspondence:**
Dr. Tarek Elsayed Ftohy Abdelrahman,
E-mail: tarekftohy2@gmail.com

ABSTRACT

Background: Brown tumors are very rare giant cell lesions that arise as a result of hyperparathyroidism, brown tumors rarely affect the mandible and maxilla and its diagnosis and treatment is very challenging and needs a high index of suspicion.
Methods: Retrospective chart review of nine cases diagnosed as brown jaw tumors treated in maxillofacial, head and neck surgery unit, department of surgery, Sohag University hospital.
Results: They were 9 patients, 3 males and 6 females with a mean age of 43.3 years. Lesions were located in the mandible (4 cases), maxilla (3 cases), both mandible and maxilla (2 cases). The main presentation was a mass lesion. Clinical examination and serum parathormone, serum calcium, serum phosphorus, neck ultrasonography, parathyroid scan was used for diagnosis. Cases did excision with parathyroidectomy. There was no recurrence during a mean follow up period of 9.3 months. All patients had satisfactory results.
Conclusions: All cases with osteolytic lesions in mandible or maxilla the possibility of brown tumors should be kept in mind especially if there is any manifestation of hyperparathyroidism. This report will aid in the recognition and treatment of the jaw brown tumors.

Keywords: Brown tumors, Hyperparathyroidism, Osteolytic lesions

INTRODUCTION

Brown Tumors are a rare result of Hyperparathyroidism, the lesion located usually in intense bone resorption areas and the bone defects filled with fibroblastic tissue. These tumors have brown or yellow colours. Brown tumors arise secondary to 1ry HPT (4.5%) and to 2ry HPT (1.5%), such rare and multiple benign lesions may resemble a malignant lesion, so diagnosis of such lesion very challenging for the maxillofacial surgeons. Brown tumors is non-odontogenic central giant cell granuloma (CGCG).[1] The pathology is rare and affects mostly young adults and more common in females with variable degree of aggressiveness. It is the terminal stage of the remodelling affecting bone which occur due to of osteoclastic activity and the fibrosis affecting the peri trabecular regions. The mandible is the site which is commonly affected in the maxillofacial region. Maxillary involvement less common.[2]

Radiologically these lesions initially appear as lytic lesion of bone classical salt and pepper appearance. open surgical biopsy is crucial for diagnosis, but radiological finding and biochemical test including serum calcium, serum parathormone, vitamin D help in diagnosis.[3] There is no consensus about the modality of management of Brown Tumor; the aim of this study was to report the outcome in most of cases to achieve the best modality of management, as curettage sometimes is done before an attempt to reach the precise diagnosis and this may lead to unnecessary bone resection. While the treatment of those condition is to correct hyperparathyroidism as the tumors may undergoes spontaneous regression after addressing the parathyroid cause. The pattern of

regression and the management will be addressed in our study.

METHODS

This is a case series descriptive study which was undertaken in the Maxillofacial, Head and Neck Surgery Unit, Department of Surgery, Sohag University Hospital, the data collected retrospectively in the periods from January 20011 to November 2016 where 9 cases were referred with Osteolytic Lesions in mandible and maxilla.

3dimentional Computed Tomography, and Biochemical tests in form of serum parathormone, serum calcium, and serum phosphorus and serum alkaline phosphatase.

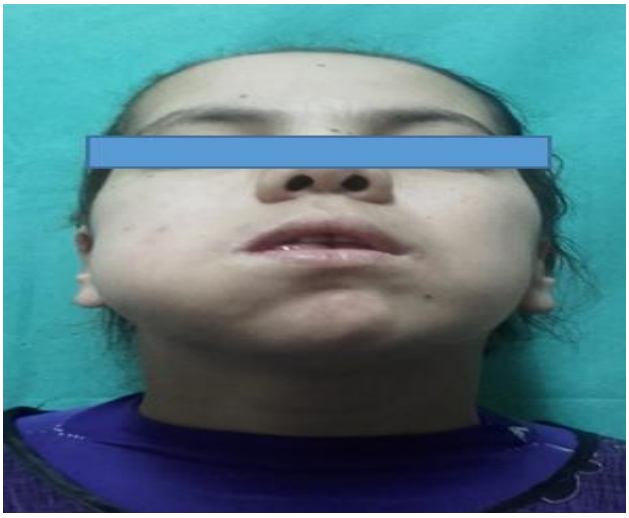

Figure 1: Brown tumor Ant. view.

Figure 2: Brown tumor lateral view.

Open surgical Biopsy is taken from all patients. Sestamibi scan done for patients with diagnosis of primary hyperparathyroidism.

Management of Brown tumor in maxilla and mandible associated with hyperparathyroidism was done either to address the hyperparathyroidism followed by follow up of the brown tumors or to do parathyroidectomy and excision of brown tumor in same session.

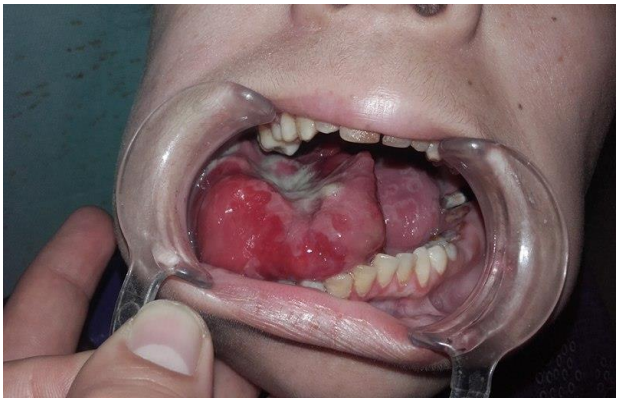

Figure 3: The oral cavity showing brown tumor.

Figure 4: Postoperative view.

The importance of different radiological evaluation methods and the consultation between the oral and maxillofacial surgeons, dentists, endocrinologists and radiologists were emphasized.

RESULTS

Case series descriptive study included 9 cases, 3 males and 6 females their ages ranged from 17 to 66 years. All patients presented with osteolytic jaw lesions affected mandible (4 cases) and Maxilla (3 cases) and mandible and maxilla (2 cases), all cases diagnosed with standard open biopsy revealed giant cell granuloma and elevated serum parathormone.

7 cases diagnosed as brown tumor secondary to 1ry hyperparathyroidism and 2 cases on top of 2ry hyperparathyroidism in renal failure patients on dialysis.

Hyperparathyroidism is addressed firstly either by excision of parathyroid adenoma in 1ry HPT patients (7 cases) or by subtotal parathyroidectomy in 2ry HPT patients (2 patients).

In four patients just follow up of the jaw swelling without curettage after parathyroidectomy three of them showed marked involution and needed no intervention while one of them showed little regression and needed curettage.

5 patients did curettage of the jaw swelling in the same session with parathyroidectomy. There is no recurrence of the jaw swelling in any patient of our study.

Table 1: Demographic criteria, clinical presentation, management outcomes of Brown tumors.

	Age (Year)	Sex	Presentation	Type of HPT	treatment	Results
1	17	Male	Multiple osteolytic lesions in mandible, previous curettage of knee lesion 3 years ago	1ry HPT	Parathyroid adenoma excision with follow up of brown tumor	Little regression after I year of surgery and needed curettage
2	55	Female	Chronic renal failure on dialysis, Rt maxillary swelling, biopsy giant cell granuloma	2ry HPT	Subtotal parathyroidectomy with follow up of brown tumor	Marked involution of the maxillary mass and needed no further curettage
3	66	Female	Left maxillary swelling, biopsy revealed RGG	1ry HPT	Excision of the adenoma with follow up of brown tumor	Marked involution and ossification of the mass and needed no further treatment
4	50	Male	Mandibular and gingival swelling	2ry HPT	Excision of the adenoma and curettage of mandibular mass	No evidence of recurrence in 1 year follow up
5	40	Female	Right Maxillary swelling	1ry HPT	Excision of the adenoma and curettage of the maxillary swelling	No evidence of recurrence in 1 year follow up
6	16	Male	Recurrent Left Mandibular Swelling	1ry HPT	Excision of the parathyroid adenoma and curettage of the mandibular lesion	No further recurrence
7	66	Female	Mandibular and maxillary swelling	1ry HPT	Excision of parathyroid adenoma and curettage of mandibular and maxillary swelling.	No evidence of recurrence in 1 year follow up
8	46	Female	Left Swelling Mandibular	1ry HPT	Excision of the parathyroid adenoma and curettage of the mandibular swelling	No evidence of recurrence in 1 year follow up
9	43	Female	Swelling Mandibular	2ry HPT	Subtotal parathyroidectomy and follow up of brown tumor	Marked involution of the mandibular mass and needed no further curettage

DISCUSSION

osteolytic lesions in facial bones either benign or malignant are very variable, in those lesions the most common diagnosis is either dental cyst, radicular cyst, ameloblastoma, localized osteomyelitis metabolic bone tumors, bone cyst i.e. simple cyst.

Osteolytic giant cell lesions that may occur in the maxillofacial region include giant cell tumors, brown tumors and giant cell reparative granuloma. It is difficult to differentiate brown tumors from other giant cell lesions depending on radiological and histological evaluation. Diagnosis is made depending on the presence of hyperparathyroidism.[4]

Excessive secretion of parathyroid hormone cause bone resorption accompanied with fibrovascular marrow replacement and increased osteoblastic activity, the imbalance between osteoclastic and osteoblastic activity manifest as an enlarging painful bony mass. The histopathology shows diffuse proliferation of the osteoclastic multinucleated giant cells mixed with fibro

cellular proliferation and hemorrhagic foci, with release of hemosiderin so appears as reddish-brown mass.[5,6]

However, the histopathological diagnosis feature alone cannot confirm the diagnosis because other giant cell lesions such as giant cell granuloma is similar to the lesions therefore, patients with giant cell tumors should be included in investigation for the presence or absence of hyperparathyroidism in order to differentiate these types of granuloma from brown tumors.

Awareness of the condition is mandatory for early diagnosis. In patients presenting with multiple osteolytic lesions brown tumors should be excluded.

Surgical biopsy is considered the most important modality for diagnosis of brown tumors, it is likely that a biopsy can be inconclusive in many cases. An accurate diagnosis difficult to be relied on a single diagnostic modality and a comprehensive image should be considered. Serum parathormone levels with a bone scan provide a highly accurate method to reach an accurate diagnosis in such cases.[7]

Classically, the clinical manifestation of hyperparathyroidism has been described as "bone, stone, abdominal groans, psychic moan".[8,9] These symptoms due to hypercalcemia, excessive bone resorption, and mental troubles that comes along.

There is a consensus that the treatment of choice for primary hyperparathyroidism being parathyroidectomy either excision of adenoma in 1ry HPT or subtotal parathyroidectomy, in 3ry HPT but there is no consensus regarding the treatment options of the brown tumors. To our knowledge this the 1st literature describing the different modalities of management of brown tumor.

Scott et al believe that bone lesions resorb spontaneously following addressing of the diseased parathyroid, in our study only 3 cases the lesions resolute spontaneously after addressing of parathyroid disease and needed no further surgery.[10] In the case of large osteolytic cysts, the amount of tissue damaged may be so great that the chance for remodelling after correction of normocalecemia be very unlikely in these conditions and or in patients where the lesion continue for more than 6 months, or if there is affection of the function of the affected organ, or continuous increase in size despite adequate metabolic control, Yamazaki et al, recommend curettage and enucleation, in our study 6 cases did curettage in the same session with parathyroidectomy with very good result as there is no recurrence for 1 year postoperative.[11]

CONCLUSION

Brown tumor is very rare lesion and the incidence is decreasing due to the advances in in the investigation and diagnosis of the hyperparathyroidism.

The treatment of choice for brown tumors is parathyroidectomy and follow up, however in cases of large, destructive lesions or deforming lesions or lesions with persistent growth in spite of addressing parathyroid disease curettage and excisions should be conducted.

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