Available online at http://www.journalcra.com

International Journal of Current Research
Vol. 8, Issue, 12, pp.43821-43825, December, 2016

INTERNATIONAL JOURNAL OF CURRENT RESEARCH

ISSN: 0975-833X

RESEARCH ARTICLE

BROWN TUMOR OF THE MANDIBLE AS FIRST MANIFESTATION OF ATYPICAL PARATHYROID CARCINOMA: A CASE REPORT AND LITERATURE REVIEW

[1,*]Naji, Y., [1]MedaghriAlaoui, O., [2]Naji, A., [3]El Attar, H. and [1]BenYahya, I.

[1]Department of Oral SurgeryDental College, Casablanca Morocco
[2]Endocrinologist Private Practice
[3]Anatomopathologist Private Practice

ARTICLE INFO

Article History:
Received 23rd September, 2016
Received in revised form
16th October, 2016
Accepted 20th November, 2016
Published online 30th December, 2016

Key words:

Brown Tumor, Mandible,
Parathroid, Carcinoma.

ABSTRACT

Brown tumor is a rare clinical entity. It's an osteolyticand non neoplasic lesion due to hyperparathyroidism. This report describes the case of a 48 year old man who presentedwith a painless swelling in the mandible lasting for three months as chief complaint. The diagnosis of brown tumor was confirmed after performing the biopsy and doing the blood tests for calcium and parathyroid hormone levels which were severely high: 1962 pg/ml. those investigations led us to discover a parathyroid carcinoma. This work highlights the diagnosis challenges and pitfalls linked to this rare and poorly understood disease. It also emphasizes the interest of the early screening of such endocrinopathy.

Citation: Naji, Y., MedaghriAlaoui, O., Naji, A., El Attar, H. and BenYahya, I. 2016. "Brown tumor of the mandible as first manifestation of atypical parathyroid carcinoma: a case report and literature review", *International Journal of Current Research*, 8, (12), 43821-43825.

INTRODUCTION

Brown tumor is an outstanding feature in the practice of a dentist. It was first described in 1934 by Albright. This non-neoplastic and osteolytic lesion results from an abnormality of bone metabolism which is proved to be a late manifestation of hyperparathyroidism. Generally it can be locally aggressive and it's commonly located in the mandible, ribs, pelvis, and femur and palate (Sutbeyaz *et al*., 2009; Triantafillidou *et al*., 2006). Brown tumor resulting of primary hyperparathyroidism occurs mainly in young adult female with a male to female ratio of 1/3and mostly occurs at the five decade of life (Silverberg *et al*., 1997). Resendiz-Colosia*et al*. revealed that 91% of their patients were women (Reséndiz-Colosia *et al*., 2008). Through this work we insist on the importance of the multidisciplinary approach to reach the correct diagnoses of this uncommon disease by reporting an exceptional case of a 48 year old man with a bony lesion in the mandible which led to the diagnosis of primary HPT due to a parathyroid carcinoma.

Corresponding author: Dr.NajiYoussef,
Department of Oral SurgeryDental College, Casablanca Morocco.

CASE REPORT

A 48 year old male patient presented to oral surgery unit for evaluation of a painless swelling in the right molar area of the mandible evolving since three months.Personal history of the patient revealed no addiction to alcohol, smoked or chewed tobacco and other drugs. His medical history revealed no history of such illness or any trauma, no significant medical or surgical intervention in the past. The man claims to have suffered multiple tooth extractions in the swellings area performed by a quack doctor before because of their increased growing mobility. He also reported asthenia and weight loss. Extra oral examination showed normal facial appearance without asymmetry and revealed submandibular lymphnodes. Intraorally, there was a firm and painless swelling of the right molar mandibular area which is an old extraction seat as well as expansion of the buccal and lingual cortex extending from lower right premolar to the ramus. The swelling was around 4 cm and was filling the vestibule and invading the right side of the floor.The lesion was covered by a normal mucosa aspect except at the ridge where there is a bite mark of the opposing tooth. The adjacent teethwere vital and suffering no mobility. Radiographic examination, showed a bone radiolucency image withunclear boundaries and loss of the dental sector in the panoramic radiography.

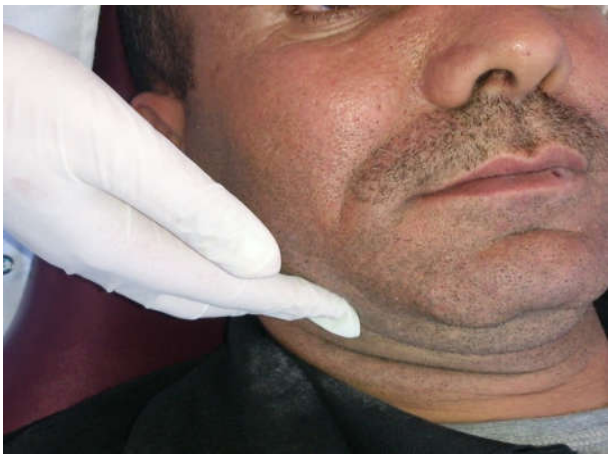

Figure 1. Extra oral palpation reveals one submandiblarlymphnode

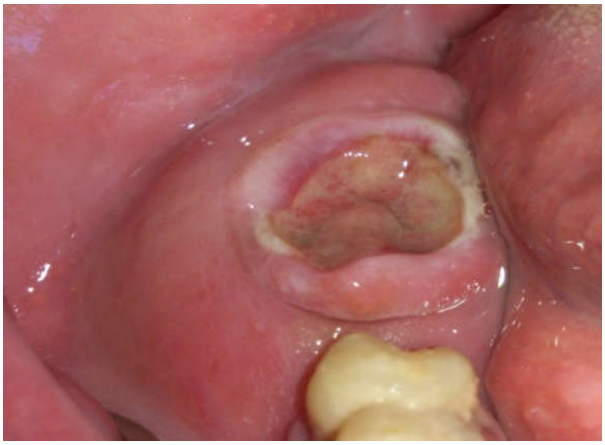

Figure 2. Intra oral view showing the swelling extending from the low right premolar to the ramus

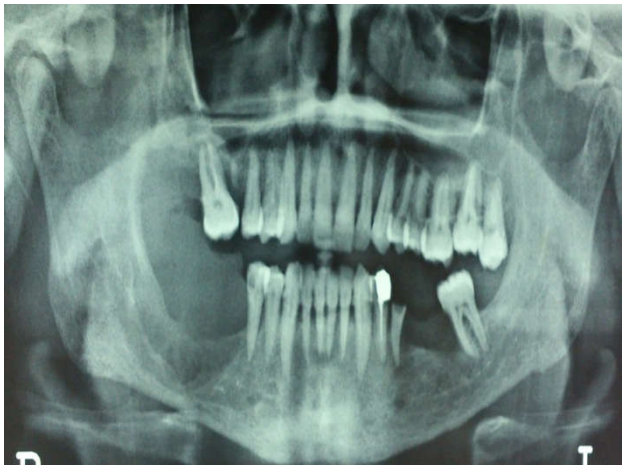

Figure 3. Panoramic showing bone radiolucency image with unclear boundaries and loss of the dental sector

We asked for a cone beam CT to better characterize the lesion and to get more specific details about the importance and the extension of the lesion by giving us cross-sectional (buccolingual), axial, sagittal, coronal and panoramic views. In the right mandibular lesion the CBCT assessed an excentric expansive osteolytic lesion blowing the cortical in the buccal side and interrupting it in the lingual side. In the left mandibular lesion it revealed an osteolytic image blowing the cortical in both buccal and lingual side without any interruption.

CBCT also established the anatomic relationship with the alveolar inferior nerve which appears to be repressed by the lesion. We evocated an aggressive tumor and a diagnostic incisional biopsy was performed. The histological exam revealed a tumor characterized by the presence of many multinucleated giant cells with cellular vascular stroma, which suggested central giant cell granuloma or giant cell tumor of the bone.Biochemical analyzes including calcium and phosphate levels and an assay of parathyroid hormone (PTH) were asked to make the differential diagnosis between giant cell granuloma and brown tumor associated to hyperparathyroidism. The parathyroid hormone level was extremely high 1962 pg/ml (15–65), blood calcium 123 mg/l (85–105) and phosphorus 19 mg/l (25–50).The patient's renal function tests were normal eliminating the brown tumor associated to secondary hyperparathyroidism.The search for other associated endocrine disease was negative.

Following the confrontation of clinical, radiological, pathological and biochemical elements, the diagnosis of brown tumor associated to primary hyperparathyroidism was confirmed to thereby eliminate the giant cell granuloma.Neck ultrasound were then performed evoking a parathyroid tumor Radiological examination including x-rays of the costal grill, tibia, fibula and hand revealed other locations than mandibular mainly in the pelvis and fingers. Technetium bone scintigraphy revealed diffuse metastatic calcifications.The therapeutic approach consisted in the removal of the parathyroid tumor that proved to be a parathyroid carcinoma in pathological examination. Macroscopic exam shows a parathyroid nodule measuring 6 cm. Microscopic exam shows an adherence to thyroid gland with capsular invasion and vascular invasion. A lobectomy were lately performed. We insisted on the importance of the monitoring of parathyroid hormone and swelling jaw.4 months later, we assessed a return to normal PTH levels and a significant reduction of the tumor; against-all indicating resorted to surgery.

DISCUSSION

Brown tumor is reported to occur in 2 to 4.5% of patients with primary HPT, and 1.5 to 1.7 of patients with secondary HPT (FassihMalika *et al*., 2013) It can be caused in 90% of cases by a parathyroid adenoma which is very difficult to palpate, 9% by a hyperplasia of the parathyroid and exceptionally in less than 1% by a parathyroid carcinoma (Holt *et al*., 2007) which account for less than 0.05% of all cancer (FassihMalika *et al*., 2013). In our case the brown tumor of the mandible was the first and only sign of a severe primary hyperparathyroidism which had led us to discoverthe parathyroid carcinoma involvement making this case unusual and exceptional. Brown tumor can be judged as a reaction rather than a true tumor. Clinically, the symptoms caused by these lesions depend on the size of the process and its location. It's generally an aggressive and painful mass growing slowly. In some cases the evolution of the brown tumor can disfigure the patient (Parisien *et al*., 1990). Radiologically, brown tumor is manifested by nonspecific osteolysis which may take many forms. The most common appearance is that of a monogéodique well-defined osteolysis or multilocularosteolysiswih no precise limits. It generally causes a blowhole or a break of the cortical. In the jaws chronically raised PTH are a rare cause of radiolucency (roughly 1%).

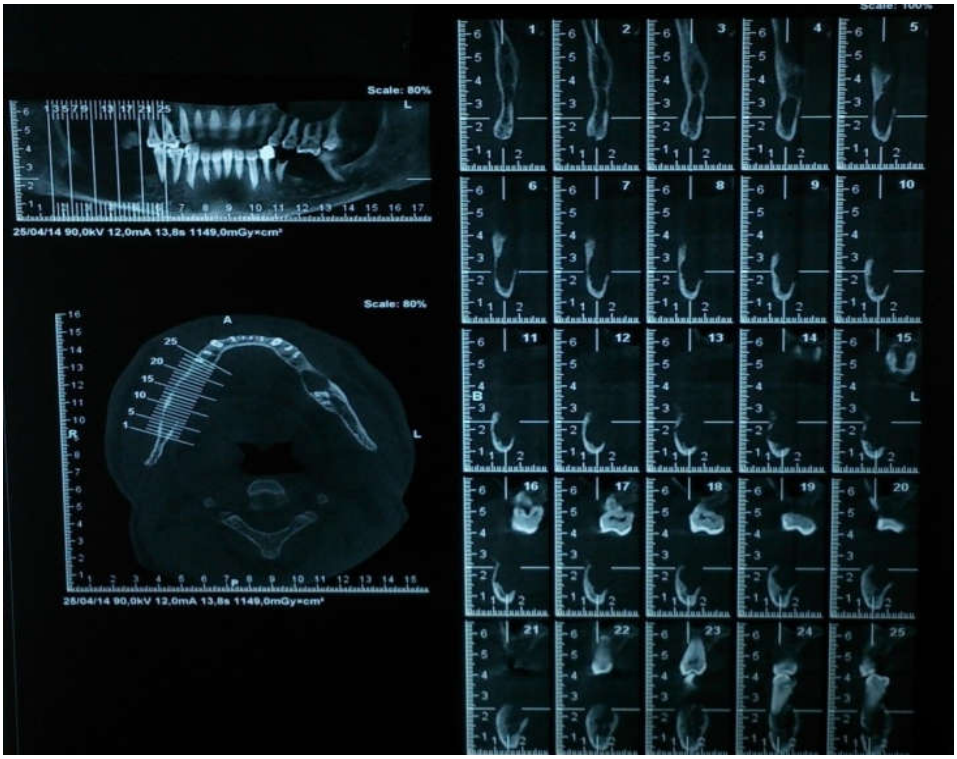

Figure 4. CBCT revealing expansive osteolytic lesion blowing the cortical in the buccal side and interrupting it in the lingual side

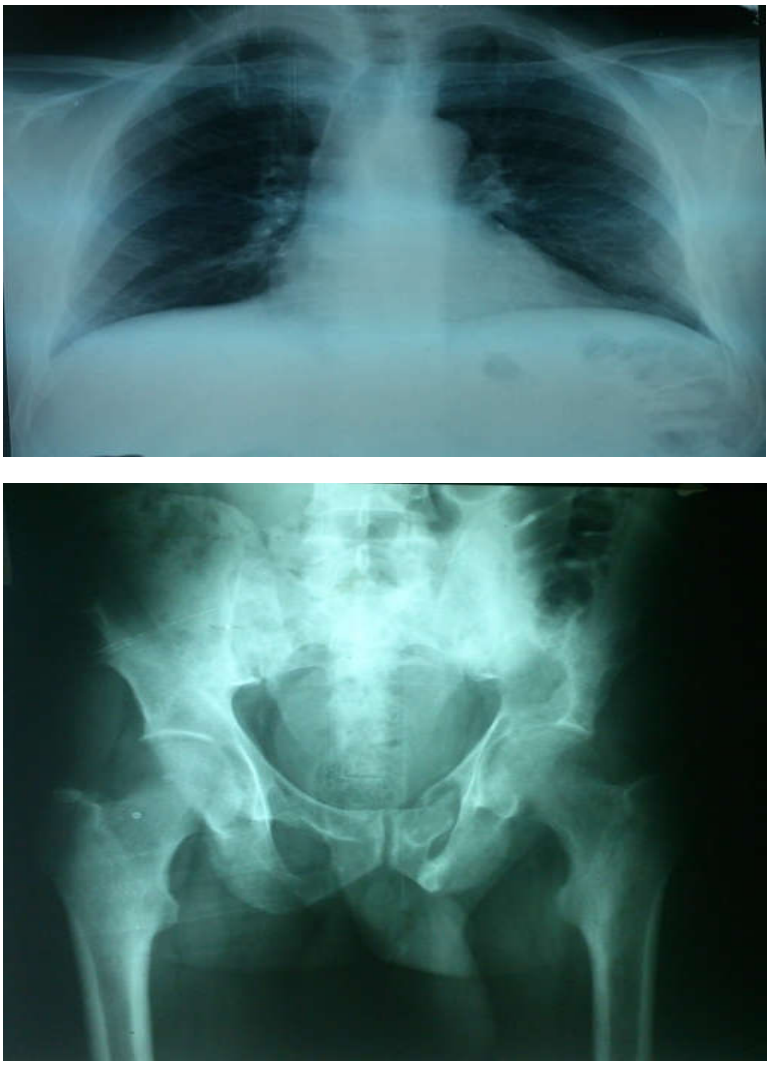

Figure 5. Radiological examination including x-rays of the costal grill, pelvis revealing other localizations of the lesion

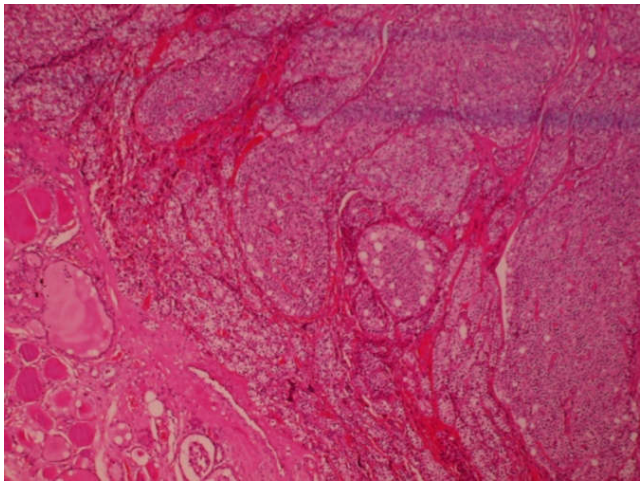

Figure 6a. Photograph of the hematoxylin and eosin *200 showing Invasion of tumoral proliferation to adjacent structure

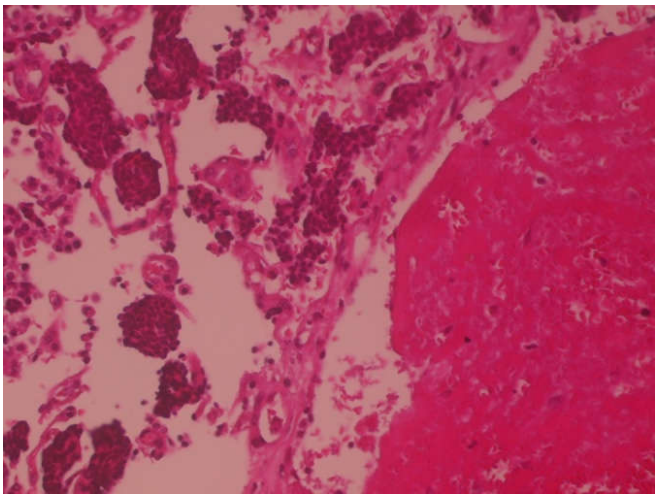

Figure 6b. Photograph of the hematoxylin and eosin *400 showing Invasion of tumoral cells to a vascular space

Mostly, osteolysis is central or eccentric. It can thin the cortical or even more interrupt it leading to a pathological fracture. These lesions can appear as large destructive ones because of the osteoclastic effect of hyperparathyroidism which may then suggest malignancy. The scan reveals a mass of tissue density, taking the contrast but do not invade the soft tissues, and no periosteal reaction is noticed (Ajmi *et al*., 2010) (Silverberg *et al*., 1997). Extra-facial bone lesions are sought by the radiological assessment of skeletal and bone scintigraphy with Technetium. Cervical imaging (ultrasound, computed tomography and magnetic resonance images) have a great contribution in the discovery of parathyroid lesion involved and are a complement to the diagnosis (Ajmi *et al*., 2010; Kim *et al*., 2012; Meydan *et al*., 2006; Hardoff *et al*., 1996; Weber *et al*., 2000). Those images modalities were a great help in revealing multiple skeletal involvement of brown tumor besides mandible locationand were essential in detecting the carcinoma.Biopsy of the suspicious bone tumor remains insufficient to ascertain the diagnosis of brown tumor. The pathological exam will just show a giant cell lesion. Only the association of clinical examination, radiographic imaging, blood tests for calcium and parathyroid hormone level with a histopathological examination is crucial and essential to reach the correct diagnosis. In our case the coexistence of multiple osteolytic lesions and the pathological exam led us to screen for PTH, blood calcium and phosphorus.

High levels of parathyroid hormone were a great help concluding the positive diagnosis of brown tumor and eliminating central giant cell lesion. The differential diagnosis arises with lesions with suspicious radiolucent images as much as giant cell tumors (GCT), central giant cell granuloma, aneurysmal cyst and rarely osteosarcoma (Murphey *et al*., 2001). Parathyroid carcinoma is a very rare malignant neoplasm with a painless evolution. It's difficult to determine it as a cause of primary HPT, as it is both rare and presents with a clinical and biochemical profile similar to benign parathyroid disorders (Chen *et al*., 2013). This case posed three major diagnostic challenges. First, identifying the brown tumor among the other giant cell tumors.Secondly, discovering the primary HPT as the cause of the tumor. Thirdly, identifying parathyroid carcinoma as the cause of the primary HPT prior to surgery.

The therapeutic approach depends on the evolution of biochemical parameters after extirpation of the parathyroid tumor. Usually, brown tumors spontaneously regress partially or totally with rapid conversion of the abnormal bony lesions to normal bone and most authors agree. Prognostic of bone turnover is mostly good. Bone reconstruction can begin 4-6 months after surgery and continue for 1-2 years it also can take too long when patient are over the sixty.Resendiz-Colosia *et al*. reported spontaneous regression of the brown tumor after the removal of the parathyroid in all their 20 cases (Reséndiz-Colosia *et al*., 2008). This study is supported by Knevezic& al and Silverman & al studies which revealed that their patients who were 15 and 23 years old with brown tumor associated to hyperparathyroidism healed six months after the parathyroidectomy (Knezevic *et al*., 1991; Silverman *et al*., 1968). Surgical treatment of brown tumor is only indicated when the tumor is wide and symptomatic leading to functional disorders, when the lesion shows an abnormal increase in its volumeleading to disfiguring patients, when the affected bone is weakened.orwhen it takes too long to regress (Pellegrino *et al*., 1977; Agarwal *et al*., 2002).

Kennet and Pollock believed that surgery of the brown tumor should be performed in association toparathyroidectomy because spontaneous regression may take too long when it comes to adenoma (Kennet *et al*., 1971). Once the indication is placed a local curettage or enucleating of the lesion should be performedassociated in some cases with a filling bone. In conclusion, giant cell lesions of the jaws must systematically seek hyperparathyroidism by the practice of calcium, phosphate and parathyroid hormone assay. The correct diagnosis of primary hyperparathyroidism is very difficult and remains a hard challenge. It avoids radical treatments with surgery because brown tumors are expected to regress and heal after the removal of the parathyroid lesions. This report highlights the important role of dentists and oral surgeons in the early screening of head and neck cancer.

Conflict of interest

None declared.

REFERENCES

Agarwal, G., Mishra, S.K., Kar, D.K. *et al*. 2002. Recovery pattern of patients with osteitisfibrosacystica in primary hyperparathyroidism after successful parathyroidectomy. *Surgery*, 132:1075–1085.

Ajmi, S., Sfar, R., Trimeche, S., Ben Ali, K., Nouira, M. 2010. Scintigraphic finding sin hungry bonesyndrome following parathyroidectomy. *Rev Esp Med Nucl.,* 29:81–3.

Chen, Q., Kaji, H., Nomura, R. *et al*. 2003. Trial to predict malignancy ofaffected parathyroid glands in primary hyperparathyroidism. *Endocr J*., 50:527-534.

Fassih Malika1, Loubna Taali1 *et al*. 2013. Tumeur brune du maxillaire révélatrice d'hyperparathyroidie primaire: à propos d'un cas et revue de la littérature *The Pan African Medical Journal,* 14:21. doi:10.11604/pamj. 2013.14.21. 1359

Hardoff, R., Frajewicki, V. 1996. Bone scintigraphy in hungry bone syndrome following parathyroidectomy.*J. Nucl. Med.,* 37:1371–3.

Holt, E.H. and Inzucchi, S.E. 2008. Physiology and pathophysiology of the parathyroid glands and preoperative evaluation. In: Oertli D, Udelsman R eds. *Surgery of the thyroid and parathyroid glands*. Heidelberg, Germany: Springer; 235–243.

Kennet, S., Pollick, H. 1971. Jaw lesions in familial hyperparathyroidism. *Oral Surg, Oral Med, Oral Pathol, Oral RadiolEndod.,* 31:502–510.

Kim, Y.I., Jung, Y.H., Hwang, K.T., Lee, H.Y. 2012. Efficacy of 99 mTc-sestamibi SPECT/CT for minimally invasive parathyroid ectomy: comparative study with 99mTc-sestamibiscintigraphy, SPECT, US and CT. *Ann, Nucl. Med.,* 26:804–10.

Knezevic, G., Uglesic, V., Kobler, P., Svajhler, T., Bagalin, M. 1991. Primary parathyroidism: evaluation of different treatments of jawlesions based on case reports. *Br J Oral MaxillofacSurg.,* 29:185-7.

Meydan, N., Barutca, S., Guney, E., Boylu, S., Savk, O., Culhaci, N. et al. 2006. Brown tumors mimicking bone metastases. *J Natl Med Assoc.,* 98:950–3.

Murphey, M.D., Nomikos, G.C., Flemming, D.J. *et al*. 2001. Imaging of giantcell tumor and giant cell reparative granuloma of bone: radiologicpathologiccorrelation. *Radiographics*, 21:1283-1309.

Parisien, M., Silverberg, S.J., Shane, E. *et al*. 1990. Bone disease in primaryhyperparathyroidism. *Endocrinol Metab Clin North Am*., 19:19–34.

Pellegrino, S.V. 1977. Primary hyperparathyroidism exacerbated by pregnancy. *Oral Surg Oral Med Oral Pathol.,* 35:915–17.

Reséndiz-Colosia, J.A., Rodríguez-Cuevas, S.A., Flores-Díaz, R. *et al*. 2008. Evolution of maxillofacial brown tumors after arathyroidectomyinprimary hyperparathyroidism. *Head Neck*, 30:1497-504.

Silverberg, S.J., Bilezikian, J.P. 1997. Primary hyperparathyroidism: stillevolving? *J Bone Miner Res.,* 12:856e62.

Silverman, S. Jr, Ware, W.H., Gillooly, C. Jr. 1968.Dental aspects of hyperparathyroidism. *Oral Surg Oral Med Oral Pathol*., 26:184-9.

Sutbeyaz, Y., Yoruk, O., Bilen, H., Gursan, N. 2009. Primary hyperparathyroidismpresenting as a palatal and mandibular browntumor. *J Craniofac Surg.,* 20:2101e4.

Triantafillidou, K., Zouloumis, L., Karakinaris, G. *et al*. 2006. Brown tumors of the jaws associated with primary or secondary hyperparathyroidism. A clinical study and review of the literature. *Am J Otolaryngol.,* 27:281-286.

Weber, A., Randolph, G., Aksoy, F. 2000. The thyroid and parathyroid glands: CT and MRimaging and correlation with pathology and clinic. *RadiolClin North Am.,* 38:1105–29.

****