Brown tumor of the mandible secondary to parathyroid adenoma. A case report.

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Case Report

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Abstract: Brown tumors are one of the bony complications of hyperparathyroidism but rarely the first manifestation. The classical "brown tumor" is seen in the ends of long bones, the pelvis, and ribs. Facial involvement is rare and usually involves the mandible. A rare case of a 35-year-old male patient with a brown tumor in his mandible associated with primary hyperparathyroidism is presented. A thorough diagnostic workup was carried out, and treatment options for hyperparathyroidism and brown tumors were discussed. The importance of different radiological evaluation methods and the consultation between the oral and maxillofacial surgeons, endocrinologists, and radiologists are emphasized.

Key words: Brown tumor; mandible; primary hyperparathyroidism; case report

Introduction

Hyperparathyroidism (HPT) is a disease in which there may be complex biochemical, anatomic, and clinical abnormalities resulting from increased secretion of parathyroid hormone (PTH). It may occur in primary, secondary, and tertiary forms[1]. Primary HPT is characterized by excess secretion PTH due to an abnormality in one or more of the parathyroid glands[2].

Parathyroid adenomas were considered to be the main etiology in about 85%of primary HPT cases. The name "brown tumor" derives from the color, which is caused by the vascularity, hemorrhage, and deposits of hemosiderin[3]. This bony lesion of the HPT is caused by increased circulating levels of parathyroid hormone, resulting in increased osteoclastic bone resorption, primarily in cortical bone[4].

It is generally accepted that bone involvement is a late manifestation of primary hyperparathyroidism (PHPT). Patients presenting in this manner were once common. However, such a presentation is rare because most cases of PHPT are detected early and before symptomatic bone lesions appear due to improved blood screening techniques[5].

Case report

A 35-year-old patient presented with a swelling on the right side jaw for the past 12 months, which gradually enlarged up to the present size. The patient gives a history of generalized weakness, lethargy, and weight loss noticed for the past few months. Her family history and past medical history were non-significant.

On extra-oral examination, a diffuse swelling was noticed on the body, angle, and ramus of the right side of the jaw, measuring approximately 7.2 cm × 4.7 cm, extending from the body, angle, and ramus of the right side of the jaw (**Figure 1A**). The skin over the swelling was shiny and stretched. There was no ulceration or erythema noticed over the swelling. On palpation, the swelling was firm to hard and tender.
On intraoral examination, an ulceroproliferative growth in the vestibule on the right side, extending from the mesial aspect of the 1st lower molar to the mesial aspect of the 1st upper molar aspect. The growth was covered with a pseudomembranous slough. (**Figure 1B**).

On palpation, the swelling was soft to firm in consistency and non-tender. No associated discharge or bleeding was noticed. Lymph nodes were palpable, which was tender and firm inconsistency. Radiographic examination with orthopantomogram showed large unilocular radiolucency on the right side, extending from the body, angle, and ramus of the jaw. There was a generalized decrease in the density of the mandible (**Figure 3A**).

The patient has advised blood investigations that revealed alkaline phosphatase and PTH levels of 268 IU/L (Normal: 38-126 IU/L) and 1587 pg/ml (Normal: 12-65 pg/ml), respectively. Serum calcium level was 14.9 mg/dl (Normal: 8.4-10.2 ml/dl).

The patient underwent high-resolution sonography of the neck, which revealed a well-defined lesion in the angle of the right side of the jaw, measuring about 29 × 31 x 24 mm. The lesion had a hypoechoic solid component. A parathyroid

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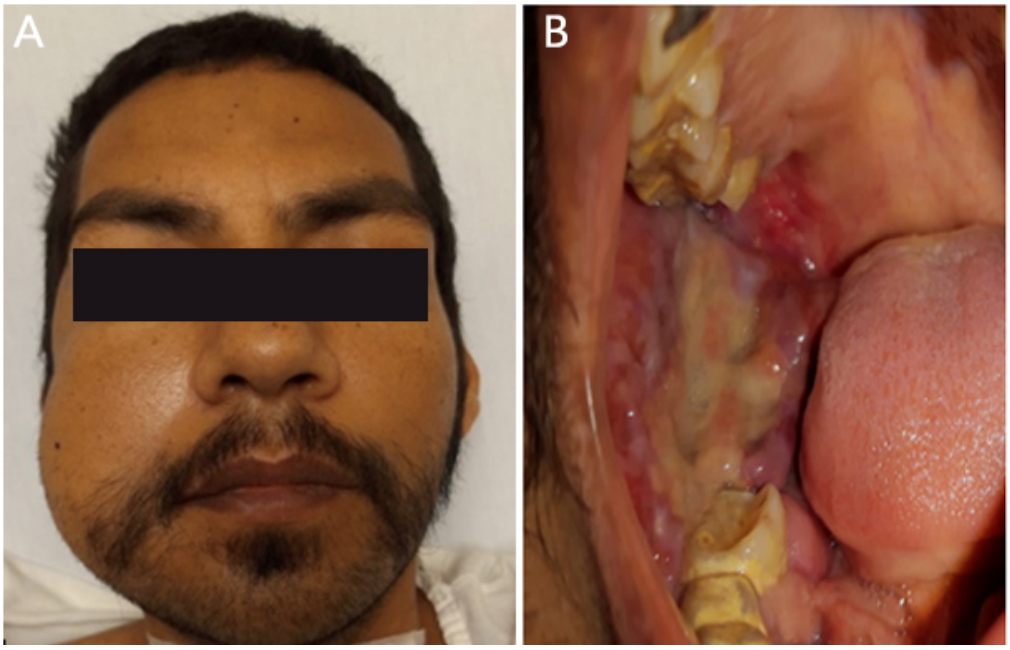

Figure 1. A. Patient presenting a diffuse swelling was on the right side of the face. **B.** An ulceroproliferative growth in the vestibule on the right side, extending from the mesial aspect of the first lower molar to mesial aspect of the first upper molar aspect.

technetium scintiscan (99Tcm SESTAMIBI; Technetium-99 MIBI; methoxy-isobutylisonitrile) findings were showing abnormally high uptake is observed at the central region of superior mediastinum interpreted as parathyroid adenoma with a possible brown tumor of the mandible (**Figure 3B**). At the level of the body, angle, and ramus of the mandible in the lower right alveolar ridge, a lytic lesion ovoid, with partially defined lobulated edges that conditions bone destruction of the mandible from the first premolar to the notch and the coronoid process; it extends into the chewing space measuring approximately 7.2 x 4.7 cm. (**Figure 3C**). An incisional biopsy of the mandibular lesion was done, which revealed hemorrhagic fibrovascular connective tissue with multinucleated giant cells consistent with a giant cell granuloma diagnosis. Parathyroid ectopic adenoma excision was done under general anesthesia. Recovery was uneventful, and the patient was discharged after one week. The resected gland was histologically suggestive of an ectopic parathyroid adenoma. The patient was given oral calcium supplementation in addition to vitamin D3 for possible postoperative hypocalcemia.

Discussion

The most significant aspect of this case report was the location of the brown tumor in the mandible, the age, and sex of the patient 35 years old male, which is below the 50 years that is commonly seen in this tumor and is the sex with less prevalence.

HPT may accompany many skeletal changes[2]. One of these changes is called the Brown tumor[2]. This tumor, also known as osteitis fibrosa, cystica generalisata, or Von Recklinghausen's disease of bone, is a metabolic bone disease that develops in primary, secondary or tertiary HPT, is caused by localized, rapid, osteoclastic removal of the bone secondary to the direct effects of PTH on the bone[3]. It should be differentiated from other true giant cell tumors of bone, and it represents reparative granuloma rather than a true neoplastic process[3,6]. Brown tumors can occur in any location but are most common in the ribs, clavicle, and pelvis. Although the reported occurrence in the mandible is 4.5% of subjects in a 220-case HPT study, it is rare to find brown tumor as the initial clinical manifestation in primary HPT[7].

Our patient exhibited none of the typical symptoms of hypercalcemia that include "bony pain/bone fractures, renal stones, abdominal groans, and psychic moans." Patients with hypercalcemia also present with associated symptoms such as paraesthesia, headaches, recent fractures, constipation, polyuria, and polydipsia. However, most of the patients are asymptomatic and are usually identified as part of routine investigations[8].

The reported prevalence of brown tumors is 0.1% and can occur in the mandible, maxilla, clavicle, ribs, and pelvic bones. Although Wood, Goaz[1] and Neville et al.[9] implied that these lesions are most common in jaws, other literatures[7,10,11,12] o not agree with them. Furthermore, the frequency of occurrence is more among persons older than 50 years of age with a male to female ratio of 1:3.[13,14]. In the present case, the patient was a 35-year-old male, mandibular brown tumor found to be the first clinical manifestation of PHPT.

Clinically, brown tumors may present as small, asymptomatic swelling in the jaw bone or a painful exophytic mass. The radiographic appearance is usually a well-demarcated monolocular or multilocular osteolytic lesion infrequently associated with root resorption and loss of the lamina dura. The

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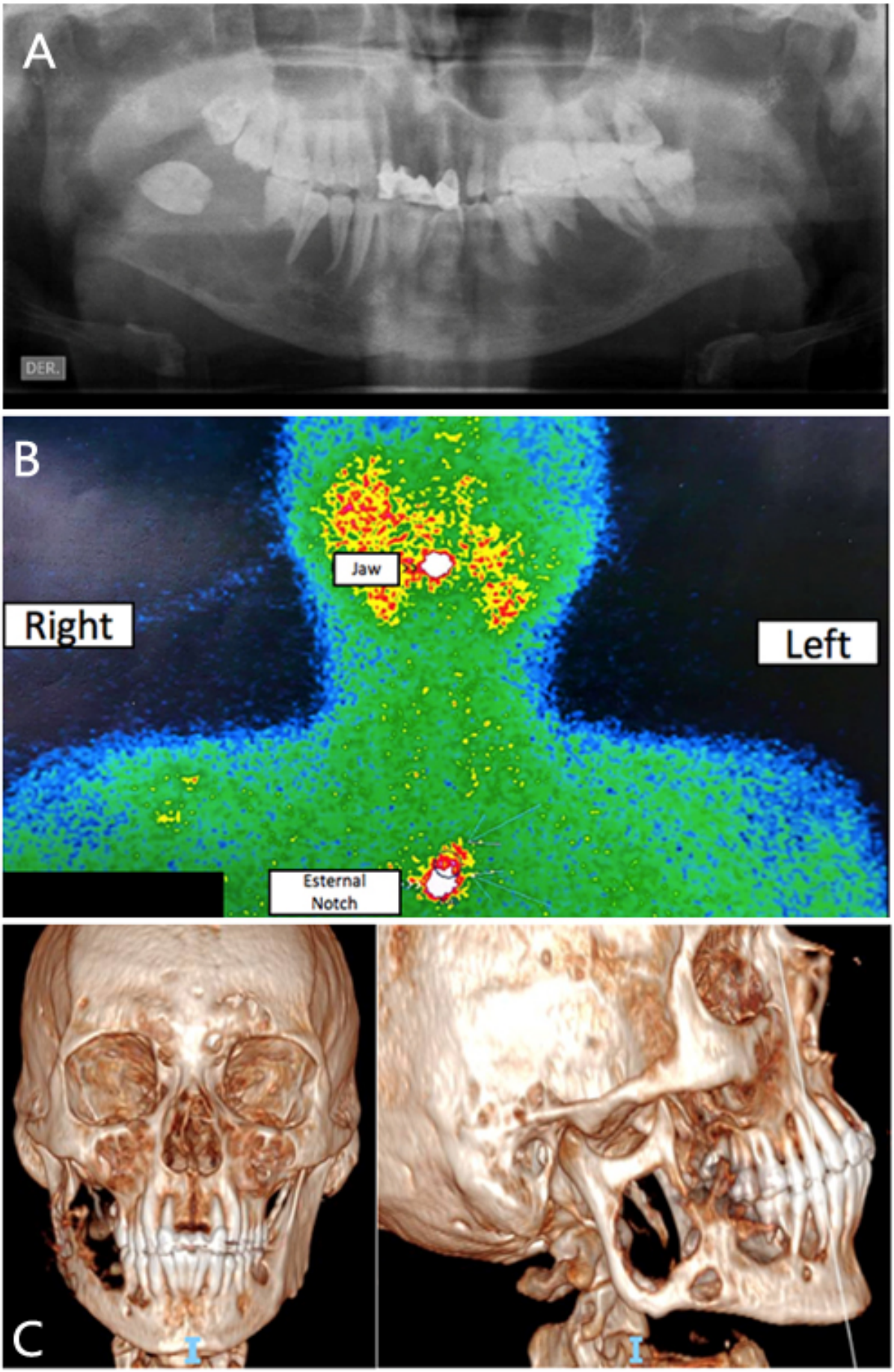

Figure 2. A. Orthopantomogram showed large unilocular radiolucency on the right side, extending from the body, angle, and ramus of the jaw. **B.** A parathyroid technetium scintiscan findings were showing abnormally high uptake is observed at the central region of superior mediastinum interpreted as parathyroid adenoma with a possible brown tumor of mandible. **C.** 3D reconstruction at the level of the body, angle, and ramus of the mandible, a lytic ovoid lesion, conditions bone destruction of the mandible from the first premolar to the notch, and the coronoid process; measuring approximately 7.2 x 4.7 cm.

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diagnosis has to be confirmed by establishing elevated serum calcium and PTH levels because histological features alone are insufficient as they may resemble any giant cell tumor. The parathyroid technetium scintiscan is one of the most preferred imaging modalities to localize diseased parathyroid glands prior to surgery.[15]

Histologically, brown tumors are characterized by vascular fibroblastic stroma, and several osteoclast-like multinucleated giant cells often interspersed with hemorrhagic infiltrates and hemosiderin deposits. The initial step in the management of primary HPT involves control of HPT, and a partial parathyroidectomy is considered effective in spontaneous regression of small osteolytic jaw lesions. However, surgical excision may be indicated in large symptomatic lesions usually done after parathyroid surgery.[7,16] Postoperative hypocalcemia may occur in patients who undergo a partial parathyroidectomy. Therefore, calcium supplements could be required, as was done with this patient.[3,17,18]

Conclusion

Even though the improvement of the various diagnostic process and biochemical tests results in the early diagnosis of HPT, the doctors should be alert of the possible occurrence of brown tumors in the jaws of the previously diagnosed patients. Also, the surgeon must be aware of oral manifestations associated with systemic diseases, hence the importance of careful physical examination and thorough investigation for the diagnosis and treatment success.

Conflicts of interests

The authors have no conflicts of interests to declare.

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References

1. Wood NK. Differential diagnosis of oral and maxillofacial lesions. 5ª ed. St. Louis: Mosby; 1997. 656 p.
2. Loushine R, Normanweller R, Kimbrough W, Liewehr F. Secondary Hyperparathyroidism: A Case Report. Journal of Endodontics. 2003; 29(4):272-4.
3. Selvi F, Cakarer S, Tanakol R, Guler S, Keskin C. Brown tumour of the maxilla and mandible: a rare complication of tertiary hyperparathyroidism. Dentomaxillofacial Radiology.2009; 38(1):53-8.
4. Daniels JS. Primary hyperparathyroidism presenting as a palatal brown tumor. Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology. 2004; 98(4):409-13.
5. Jebasingh F, Jacob JJ, Shah A, Paul TV, Seshadri MS. Bilateral maxillary brown tumours as the first presentation of primary hyperparathyroidism. Oral and Maxillofacial Surgery. 2008 [consultado el 18 de junio de 2021];12(2):97-100.
6. Soundarya N, Sharada P, Prakash N, Pradeep G. Bilateral maxillary brown tumors in a patient with primary hyperparathyroidism: Report of a rare entity and review of literature. Journal of Oral and Maxillofacial Pathology. 2011;15(1):56.
7. Suarez-Cunqueiro MM, Schoen R, Kersten A, Klisch J, Schmelzeisen R. Brown tumor of the mandible as first manifestation of atypical parathyroid adenoma. Journal of Oral and Maxillofacial Surgery. 2004;62(8):1024-8.
8. Carr ER, Contractor K, Remedios D, Burke M. Can parathyroidectomy for primary hyperparathyroidism be carried out as a day-case procedure? The Journal of Laryngology & Otology. 2006;120(11):939-41.
9. Neville BW, Neville B, Damm DD, Allen CM, Bouquot J. Oral & Maxillofacial Pathology. 2ª ed. Saunders; 2001. 768 p.
10. Tarrass F, Ayad A, Benjelloun M, Anabi A, Ramdani B, Benghanem MG, Zaid D. Cauda equina compression revealing brown tumor of the spine in a long-term hemodialysis patient. Joint Bone Spine. Diciembre de 2006; 73(6):748-50.
11. Franco M, Bendini JC, Albano L, Barrillon D, Cassuto E, Bracco J. Radiographic follow-up of a phalangeal brown tumor. Joint Bone Spine. 2002; 69(5):506-10.
12. Doğan A, Algün E, Kisli E, Harman M, Kösem M, Tosun N. Calcaneal brown tumor with primary hyperparathyroidism caused by parathyroid carcinoma: An atypical localization. The Journal of Foot and Ankle Surgery. 2004; 43(4):248-51.
13. Keyser JS, Postma GN. Brown tumor of the mandible. American Journal of Otolaryngology. 1996;17(6):407-10.
14. Queiroz SM, Vasconcelos RG, Andrade AL, Amorim AG, Gordón-Nuñes MA, Freitas Rd, Galvão HC. Maxillary brown tumor associated with chronic kidney failure: a case report. Jornal Brasileiro de Patologia e Medicina Laboratorial. 2013;49(6):424-8.
15. Triantafillidou K, Zouloumis L, Karakinaris G, Kalimeras E, Iordanidis F. Brown tumors of the jaws associated with primary or secondary hyperparathyroidism. A clinical study and review of the literature. American Journal of Otolaryngology. 2006; 27(4):281-6.
16. Alhusban M, Baqain ZH. Mandibular brown tumor as the first manifestation of primary hyperparathyroidism: A case report. The Saudi Dental Journal. 2011; 23(2):107-9.
17. Phitayakorn R, McHenry CR. Jaw Tumor: An Uncommon Presenting Manifestation of Primary Hyperparathyroidism. World Journal of Endocrine Surgery. 2010; 2(1):45-50.
18. Yildiz A, Kirdak T, Ersoy A, Ersoy C, Saraydaroglu O, Ayyildiz A, Korun N. A Rare Cause of Primary Hyperparathyroidism Presented with Giant Adenoma, Multiple Brown Tumors And End Stage Renal Failure.

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