CASE REPORT

Craniofacial brown tumor as a result of secondary hyperparathyroidism in chronic renal disease patient: A rare entity

Pradhuman Verma, Kanika Gupta Verma[1], Dinesh Verma[2], Nitin Patwardhan[3]

Department of Oral Medicine and Radiology, [1]Pedodontics and Preventive Dentistry, [2]Oral and Maxillofacial Surgery, Surendra Dental College and Hospital, Sriganganagar, Rajasthan, [3]Department of Preventive and Community Dentistry, Vaidik Dental College and Hospital, Daman, Daman and Diu, India

Address for correspondence:
Dr. Kanika Gupta Verma,
Department of Pedodontics and Preventive Dentistry, Surendera Dental College and Research Institute, H.H Gardens, Power House Road, Sriganganagar - 335 001, Rajasthan, India.
E-mail: drguptakanika@yahoo.co.in

Received: 11-11-2013
Accepted: 25-07-2014

ABSTRACT

Brown tumors are erosive bony lesions caused by rapid osteoclastic activity and peritrabecular fibrosis due to primary or secondary hyperparathyroidism resulting in a local destructive phenomenon. The differential diagnosis based on histological examination is only presumptive. Clinical, radiological and laboratory data are necessary for definitive diagnosis. Here, we report a very rare case of brown tumor involving maxilla and mandible, which is the result of secondary hyperparathyroidism in 30-year-old female patient with chronic renal disease.

Key words: Brown tumor, chronic renal disease, giant cell, hyperparathyroidism, renal diseases, secondary hyperparathyroidism

INTRODUCTION

Brown tumor is an osseous lesion that develops in bones affected by primary or secondary hyperparathyroidism (SHPT) as a component of a metabolic bone disease known as osteitis fibrosa cystica or Recklinghausen's disease of bone.[1] At present, brown tumors are considered as a reparative cellular process rather than a true neoplasm. This phenomenon is considered as pathognomonic of hyperparathyroidism (HPT) secondary to chronic renal disease.[2] SHPT is a frequent complication of chronic renal disease because of renal osteodystrophy. Brown tumors, as unifocal or multifocal bone lesions, represent a serious complication of advanced HPT with a frequency of 1.5-1.75% in SHPT. It usually affects young people, especially females, with varying degrees of aggressiveness and risk of recurrence. It affects the base of skull, orbits, paranasal sinuses and spinal column. The mandible is more commonly affected than the maxilla and is usually asymptomatic except when large in size. Radiographically, the brown tumors in the jaws presents as a well-defined radiolucent osteolytic lesion, making it hard to differentiate from other expansive lesions. The diagnosis based on histological examination is only presumptive. Clinical, radiological and laboratory data are necessary for definitive diagnosis.[3]

Access this article online

Quick Response Code:

Website: www.jomfp.in

DOI: 10.4103/0973-029X.140779

CASE REPORT

A 31-year-old female visited the Department of Oral Medicine and Radiology with the chief complaint of pain and swelling over the left side of face since 13 months. History of presenting illness revealed that similar type of swelling was present on the right side of jaw since 6 months, which resolvoed spontaneously. There was difficulty in chewing food because of presence of multiple root stumps. There was no history of recent weight loss, facial paraesthesia and trauma in past. Past medical history revealed the presence of bilateral renal calculi since 1.5 years. The patient was known hypertensive for which she was on medication since 2 years. Past dental history revealed that patient underwent extraction of 36 and 37 four months back from a local dentist. The patient was married, with two children.

On general physical examination, patient was anemic with paleness of skin and palpebral conjunctiva. Except systemic temperature of 100°F, all vitals were within normal limits. Extraoral examination showed solitary diffuse swelling over left side of face causing gross facial asymmetry [Figure 1]. It extends from ala tragus line superiorly up to 4 cm below the inferior border of body of mandible inferiorly. It extends from midline anteriorly upto posterior border of ramus of mandible posteriorly. The overlying skin was intact and of

normal color. On palpation, the swelling was tender; overlying skin was pinchable with no localized increase in temperature. The regional lymph nodes were not palpable. Intraoral examination showed large round, oval-shaped solitary diffuse swelling measuring 6.5 cm in maximum dimensions over the left mandibular alveolar ridge, extending anteriorly from canine region to retro molar regions posteriorly. The overlying mucosa was intact without any discharge. [Figure 2] On palpation, it was hard in consistency, non-fluctuant, and tender presenting with egg shell crackling. Root stumps 13, 14, 15, 16, 26, 37, 38, 43, 44 and 45 were noted.

Based upon history and clinical examination, aprovisional diagnosis of giant cell lesion of left body mandible was made and differential diagnosis of central giant cell granuloma, ameloblastoma, aneurysmal bone cyst, cherubism and brown tumor SHPT due to renal disease were considered.

Hematological investigation showed an RBC count of $3.02 \times 10^6/\mu L$, Hb of 6.6 g/dL, erythrocyte sedimentation rate (ESR) of 15 mm/1st hour and blood urea nitrogen of 29.9 mg/dl (*N* = 5-21 mg/dl).

Serological investigations revealed raised serum calcium 14.3 mg%, alkaline phosphatase level of 1963 U/L (*N* = 108-306 U/L) and serum creatinine of 1.6 mg% (*N* = 0.5-1.2 mg%). Serum sodium and potassium were within normal limits.

Urine examination showed clumped pus cells of about 80-90 cells/HPF.

Thyroid profile by fully automatic chemiluminescent immunoassay showed raised intact parathyroid hormone (PTH) level of 234.1 pg/mL (*N* = 15-68.30). Total T3 and total T4 were within normal limits.

Radiological investigations

Panoramic radiograph showed multilocular, well-defined radiolucencies with corticated margins involving body of mandible crossing midline and ascending ramus of mandible of left side. There is generalized loss of lamina dura around all the teeth. The trabecular pattern showed ground glass appearance of numerous, small, randomly oriented trabeculae [Figure 3]. Mandibular cross-sectional occlusal radiograph showed expansion of both buccal and lingual cortical shapes.

The ultrasound of kidney showed 2.4 cm renal calculus in the right calyx and large staghorn calculus in the left sided kidney suggestive of SHPT [Figure 4]. The ultrasound of thyroid and parathyroid glands showed no abnormality except increased vascularity.

CT scan showed 62 × 48 mm large expansile bony lesion with cortical expansion of bone and eccentric location at anterior and left lateral aspect of mandible. Bony lesion was causing displacement of teeth with loculated cystic lesion extending to left lateral side and approaching orbit [Figure 5].

Complete skeletal scan was performed showing no abnormality.

Incisional biopsy was performed, which showed multiple spindle to oval shaped scattered multinucleated osteoclast-like

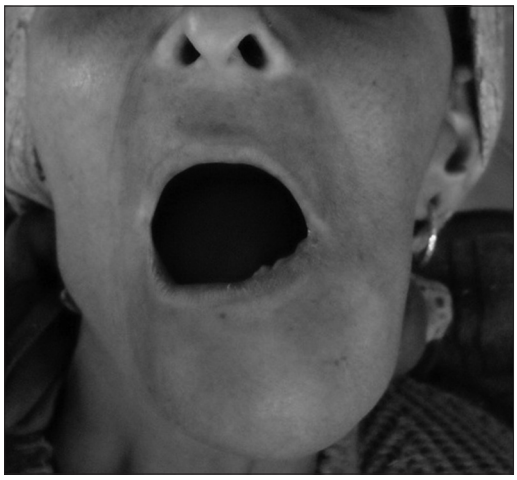

Figure 1: Photograph showing extra oral swelling on left side of face

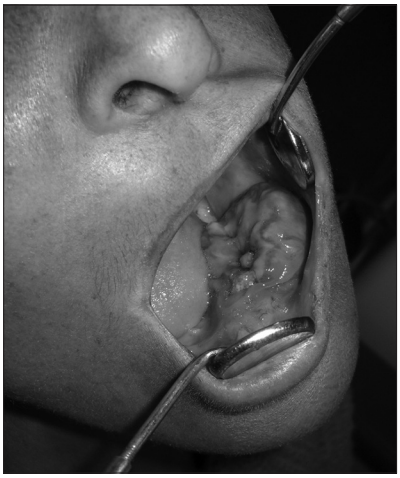

Figure 2: Intraoral photograph of patient

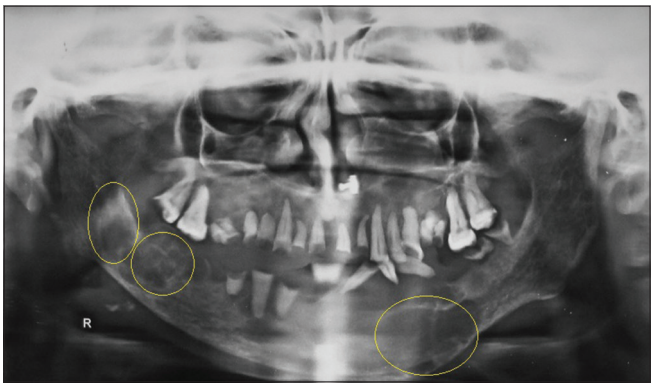

Figure 3: Panoramic radiograph showing multilocular radiolucencies

giant cells along with areas of hemorrhage and scattered lymphocytes in the stroma and loose fibrillar matrix [Figures 6 and 7].

Based upon the history, clinical, laboratorial, imaging and histopathology investigations the final diagnosis of craniofacial brown tumor because of SHPT due to chronic renal disease was made and the patient was referred for treatment in higher medical centre where a multidisciplinary approach was planned including nephrologist, endocrinologist and oral and maxillofacial surgeon.

DISCUSSION

Sylvanus (1743) was the first to diagnose HPT. Recklinghausen (1891) is credited with the first description of the associated bone changes known as osteitis fibrosa cystica.[4] The brown tumors reported to occur in 4.5% of patients with primary HPT and in 1.5-1.7% of those with SHPT. The disease can manifest at any age and is three times more common in women than in men. Brown tumors commonly affects the mandible, clavicle, ribs, pelvis and femur. In craniofacial bones, the involvement of maxilla, palate, temporal bone, nasal cavity, orbital bone and paranasal sinuses has been reported.[5] SHPT is considered when there is hyperplasia of parathyroid gland with increased secretion in an attempt to compensate for prolonged hypocalcaemia caused by chronic renal failure. SHPT may lead to renal calculi formation as seen in the present case. Chronic renal diseases cause deterioration of nephrons with decreased glomerular filtration, which results in decreased vitamin D synthesis by kidney leading to decreased calcium absorption. Consequently, an increased level of serum phosphate is also seen. Phosphate is the driving force of bone mineralization; excess phosphate tends to cause serum calcium to be deposited in bone, leading to decreased serum calcium level and structurally deficient bones. In response to low serum calcium, parathyroid glands are stimulated to secrete PTH, which results in SHPT. The best diagnostic method for SHPT is parathyroid immunoassay and confirmation of chronic renal disease.[6] Clinically, the lesion appears as smooth expansile masses causing bone destruction.[7] Radiographically, lesions are characterized as well defined radiolucent areas, unilocular or multilocular with cortical expansion. Dental changes reported in association with HPT include abnormally narrow pulp chambers, generalized loss of lamina dura around the roots of teeth and generalized demineralization of medullary bones of jaws causing characteristic "ground glass appearance",[8] all these were seen in the present case. Microscopically, brown tumors are characterized by intensely vascular fibroblastic stroma with numerous osteoclast-like multinucleated giant cells. The presence of hemorrhage, hemosiderin and hypervascularity leads to the brownish color,

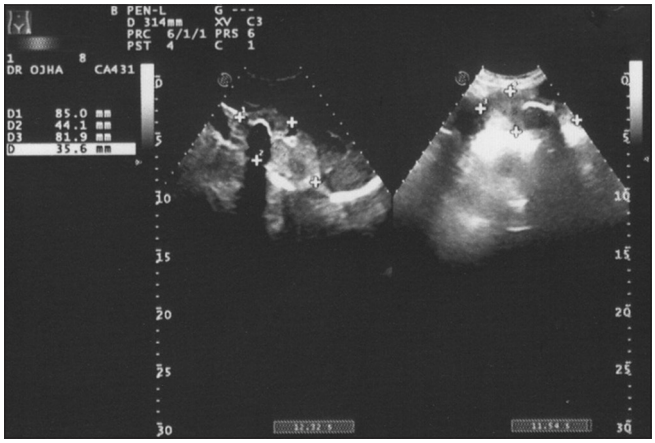

Figure 4: Ultrasound of right and left kidneys

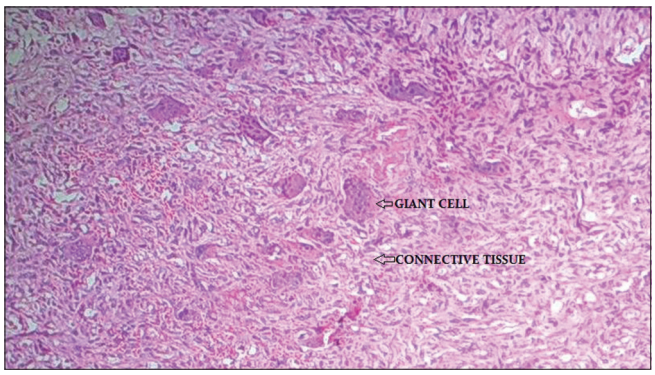

Figure 6: Photomicrograph showing multinucleated giant cells and fibro-vascular connective tissue (H&E stain, x100)

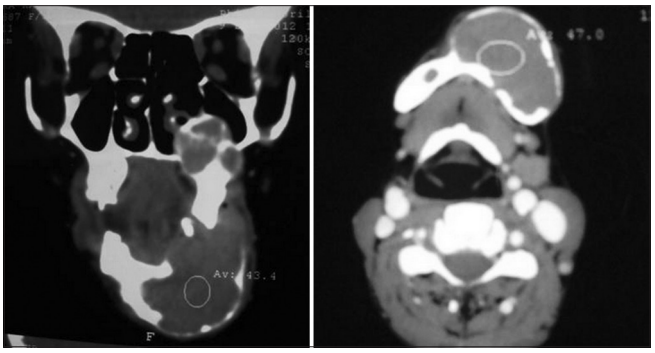

Figure 5: CT scan showing expansile bony lesion

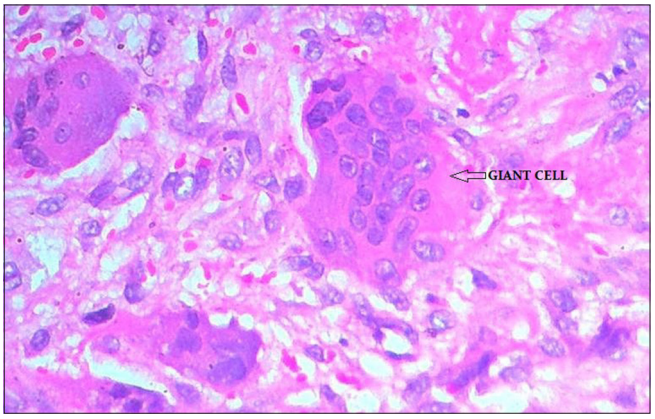

Figure 7: Photomicrograph showing multinucleated osteoclast like giant cells (H&E stain, x400)

thus, the name.[9] These findings are not pathognomic, making it necessary to perform a differential diagnosis with other lesions such as aneurysmal bone cyst, cherubism, reparative granuloma, fibrous dysplasia, giant cell tumors and central giant cell granuloma.[10] A true giant cell tumor is more infiltrative than brown tumors and shows some degree of cellular atypia. A reparative granuloma can be differentiated from brown tumor by the absence of HPT. Fibrous dysplasia is common among young women and histology reveals trabecular bone with a stroma rich in fibrous tissue. The treatment of a brown tumor is mainly pharmalogical by treating the underlying chronic renal disease and HPT; however, surgical excision is sometimes necessary.[11,12] The bone healing in these patients is compromised, HPT must be controlled prior to surgical bone reconstruction.[13] Here, a similar type of multidisciplinary approach was planned for the patient involving nephrologist, endocrinologist and oral and maxillofacial surgeon. Hence, the early diagnosis of jaw lesions of HPT is very important to demonstrate the relevance of interdisciplinary activity and the inclusion of dentist in such team.

CONCLUSION

Brown tumors of jaws develop in chronic renal disease patient; these tumors may simulate bone neoplasms and should be included in the differential diagnosis of multilocular lesions of the jaws. The oral physician may play a vital role in early diagnosis of such lesions.

REFERENCES

1. 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. J Oral Maxillofac Pathol 2011;15:56-9.
2. Fineman I, Johnson JP, Di-Patre PL, Sandhu H. Chronic renal failure causing brown tumors and myelopathy. Case report and review of pathophysiology and treatment. J Neurosurg 1999;90:242-6.
3. Kaugars GE, Niamtu J 3rd, Svirsky JA. Cherubism: Diagnosis, treatment and comparison with central giant cell granulomas and giant cell tumors. Oral Surg Oral Med Oral Pathol 1992;73:369-74.
4. Auclair PL, Avendt DM, Hellstein JW. Giant cell lesions of the jaws. Oral Maxillofac Surg Clin North Am 1997;9:655-80.
5. Som PM, Lawson W, Cohen BA. Giant-cell lesions of the facial bones. Radiology 1983;147:129-34.
6. Prado FO, Rosales AC, Rodrigues CI, Coletta RD, Lopes MA. Brown tumor of the mandible associated with secondary hyperparathyroidism: A case report and review of the literature. Gen Dent 2006;54:341-3.
7. Krause I, Eisenstein B, Davidovits M, Cleper R, Tobar A, Calderon S. Maxillomandibular brown tumor-a rare complication of chronic renal failure. Pediatr Nephrol 2000;14:499-501.
8. Langlias RP, Langland OE, Nortje CJ. Diagnostic imaging of the jaws. In: Cooke D, Zinner S, DiRienzi D, editors. Generalized Rarefactions. 1st ed. Baltimore: Williams and Wilkins; 1995p. 463-4.
9. Jebasingh F, Jacob JJ, Shah A, Paul TV, Seshadri MS. Bilateral maxillary brown tumors as the first presentation of primary hyperparathyroidism. Oral Maxillofac Surg 2008;12:97-100.
10. Fernandez-Bustillo AJ, Martino R, Murillo J, Garatea J, Palomero R. Fernandez-Bustillo AJ, Martino-Gorvea R, Murillo-Cortes J, Garatea-Crelgo J, Palomero-Rodríguez R. Primary hyperparathyroidism: Presenting as brown tumors in the maxilla. Med Oral 2000;5:208-13.
11. Pinto MC, Sass SM, Sampaio CP, Campos DS. Brown tumour in a patient with hyperparathyroidism secondary to chronic renal failure. Braz J Otorhinolaryngol 2010;76:404.
12. Magalhaes DP, Osterne RL, Alves AP, Santos PS, Lima RB, Sousa FB. Multiple brown tumours of tertiary hyperparathyroidism in a renal transplant recipient: A case report. Med Oral Patol Oral Cir Bucal 2010;15:e10-3.
13. Rosenberg EH, Guralnick WC. Hyperparathyroidism, a review of 220 proved cases with special emphasis on finding in the jaws. Oral Surg Oral Med Oral Pathol 1962;15:84-94.

How to cite this article: Verma P, Verma KG, Verma D, Patwardhan N. Craniofacial brown tumor as a result of secondary hyperparathyroidism in chronic renal disease patient: A rare entity. J Oral Maxillofac Pathol 2014;18:267-70.

Source of Support: Nil. **Conflict of Interest:** None declared.