Oral Manifestations established the Diagnosis of Hyperparathyroidism: A Rare Case Report

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ABSTRACT

A case of primary hyperparathyroidism is presented in a patient who reported to our OPD with the complaint of multiple central giant cell granulomas involving the upper and lower jaw. Such rare systemic conditions manifesting in the oral cavity pose a great challenge to diagnosis by the oral physician. However, a thorough clinical, radiological, histopathological and specialized imaging modalities are a key to its successful diagnosis, as was done in our case.

Keywords: Central giant cell granuloma, Jaw lesions, Parathyroid adenoma, Primary hyperparathyroidism.

INTRODUCTION

Central giant cell granuloma (CGCG) is a relatively uncommon pathological condition accounting for less than 7% of all benign lesions of the jaws. They are defined by the WHO as an intraosseous lesion consisting of cellular and hypervascular connective tissue containing multiple foci of hemorrhage, aggregations of multinucleated giant cells, and occasionally trabeculae of woven bone.

Clinically, CGCG occurs most commonly in young adults and has a female predilection of 3:1. Lesions are more commonly located in the middle of mandible and frequently cross the midline. The radiographic features of the CGCG comprise a unilocular or a multilocular radiolucency and varying degree of expansion of the cortical plates.

CGCGs can also affect extragnathic bones, mainly in the craniofacial region, and small long bones, such as those of the hands and feet. The microscopic features of CGCG also share similarities with the brown tumors of hyperparathyroidism and aneurysmal bone cysts.

Brown tumors are non-neoplastic lesions resulting from abnormal bone metabolism in hyperparathyroidism. These focal lesions found within regions of bone resorption can appear in any part of the skeleton but rarely are the first manifestations of hyperparathyroidism. Women are more commonly affected than men between 30 and 60 years of age. Clinically, most patients have renal calculi, peptic ulcers, psychiatric problems or bone and joint pain and the incidence of primary hyperparathyroidism is 0.1%.

Primary hyperparathyroidism usually results from a benign tumor (adenoma) of one of the four parathyroid glands. Secondary hyperparathyroidism results from a compensatory increase in the output of PTH in response to hypocalcemia and may be from inadequate dietary intake, poor absorption of vitamin D or from deficient metabolism of vitamin D in the liver/kidney.

Thus, reporting a case of primary hyperparathyroidism diagnosed because of suspicion of presence of multiple jaw lesions and by performing series of related radiological and biochemical investigations, in the Department of Oral Medicine and Diagnosis, Modern Dental College and Research Center, Indore, Madhya Pradesh, India.

CASE REPORT

A 50-year-old female patient reported with the complaint of a painless, progressive swelling in the right maxillary posterior region since 5 months. The swelling was gradually increasing in size and progressed to the present size without any history of trauma or neurological deficit. The patient attended for consultation at a private dental clinic three months ago for the same complaint which was associated with an occasional toothache. Considering the swelling to be an abscess secondary to an odontogenic cause by the treating dentist, the patient underwent extraction of teeth without any regression in the size of swelling. The patient was a known case of erosive esophagitis since 3 years, hypertensive since 2 years and also revealed hospitalization due to renal calculi one and half year ago.
On detailed examination, a well-defined swelling in 14, 15 region of size approximately 2 × 1.5 cm in diameter, oval in shape was noticed obliterating the right buccal vestibule. The overlying mucosa was pale in the central area and slightly erythematous at the periphery. On palpation, swelling was firm to bony hard in consistency, mildly tender, noncompressible, nonpulsatile without any bleeding tendency (Fig. 1). A similar small, well-defined, oval swelling was also noticed in the lower edentulous zone of 45, 46 region of size approximately 2 × 2 cm. The overlying mucosa in that region appeared purplish pink interspersed with brownish pigmentation.

Radiological examination (Fig. 2) revealed multiple ill-defined, osteolytic lesions ranging in size from 2 × 3 cm to 1 × 1 cm in diameter, in the region of 45, 46, 35, 36 and 14, 15. The inferior border of the mandible associated with the lesion was thinned out.

On evaluating the case history, clinical and radiographic examination, the case was provisionally diagnosed as an endocrinical disorder manifestating in the oral cavity, the most likely being, suspected as hyperparathyroidism, which induced multiple osteolytic lesions. The patient was thereafter subjected to incisional biopsy, serum chemistry and specialized imaging modalities to confirm the clinical diagnosis.

After the primary investigations, incisional biopsy of one of the lesions in the 15, 16 region (Fig. 3) was planned and histopathologically it revealed multiple osteoclastic giant cells in a spindle cell stream, confirming the diagnosis of central giant cell granuloma. The serum calcium, alkaline phosphatase and phosphorus levels were 13.5 mg/dl, 174.00 IU/ml and 2.40 mg/dl respectively. The serum parathyroid hormone levels were found to be abnormally raised to as high as 470.30 pg/ml.

The ultrasonography of the thyroid-parathyroid region (Fig. 4) revealed the presence of a solid, oval, nodular mass in the left lower parathyroid gland measuring 40 × 20 × 15 mm in size, having smooth margins and fairly homogeneous echostructure, but being a static image, its true functional nature could not be assessed in the USG scan. Hence, the patient was subjected to specialized investigative technique, the scintigraphy scan (Fig. 5). The scan revealed a ‘Hot Spot’ due to an abnormal focal tracer activity in the left lower parathyroid gland, confirming a functional parathyroid adenoma of size 4.0 × 2.0 cm.

**DISCUSSION**

Hyperparathyroidism first described in 1891 by von Recklinghausen occurs in three clinical forms: Primary, secondary and tertiary. In all instances, the disease is characterised by an increase in PTH levels and mobilization of calcium from the bones.2

Primary hyperparathyroidism results from primary parathyroid hyperplasia, a benign parathyroid adenoma or parathyroid carcinoma. Classically, increased production of PTH causes an increase in serum calcium levels by decreasing renal tubular reabsorption of phosphorus. Alkaline phosphatase levels are increased and serum phosphorus values are decreased,2 this phenomenon was appreciable in the present case. In addition, the patient suffered renal calculi too suggesting that skeletal bone and kidney are target organs of parathormone activity, which in excess secretion mediates the osteoclasts to resorb bone actively.

The clinical findings of hyperparathyroidism have been described by Jackson and Frame (1972) as the tetrad of “bones, stones, abdominal groans and psychic moans with fatigue overtones.”3,9,10 The condition occurs most commonly in
women, with a peak incidence between 40 and 50 years. The most common sequel of the disease is renal calcification which was recorded in the present case. Gastrointestinal disturbances may be associated with nausea, vomiting, anorexia, duodenal and peptic ulcers. Hypercalcemia also may result in CNS manifestations ranging from mild personality problems to severe psychiatric disorders. Hypercalcemia also results in muscle weakness and hypotonia.

The bony changes in hyperparathyroidism are identical, whether the cause is primary, secondary or tertiary. The classic radiographic features are as follows: (1) Subperiosteal bone resorption, (2) generalized demineralization, (3) localized lytic bone lesions, (4) metastatic calcification of soft tissue.

The central giant cell granulomas are classified as a true neoplasm and a reactive proliferative process because of its characteristic histologic features, dynamic biologic characteristics and variable clinical patterns. We tried to throw light on a case having multiple giant cell lesions of the oral cavity as one of the earliest manifestations of primary hyperparathyroidism. Female predominance is found for CGCG, which may be explained by recent suggestions of the association between hormonal secretion and the appearance of CGCG in females. This is in correlation with our case of a female patient diagnosed with multiple CGCGs. Radiographically, CGCG in hyperparathyroidism exhibits variable patterns, the borders of the lesion may be either ill-defined (66%), multilocular (54%), associated with displaced teeth and expansion of cortical bone (51%) and sometimes granular appearance of bone and thinning of lamina dura as seen in the present case. The microscopic features of CGCG also share similarities with the brown tumors of hyperparathyroidism.

Treatment modalities mainly include surgical removal. CGCG has also been treated with conservative methods, such as systemic daily application of calcitonin, intralesional injections of corticosteroids, and subcutaneous alpha-interferon injections, especially in case of hyperparathyroidism.

Another complication of hyperparathyroidism is a chronic renal disease (CRD), which is a multifactorial syndrome characterized by progressive and irreversible loss of renal mass and function. The most common complications of CRD are cardiovascular diseases including secondary hyperparathyroidism, secondary hypertension, immunosupression, infection, anemia, bleeding disorders and metastatic calcifications. The present case also revealed history of hypertension which further suggests a complication of hyperparathyroidism. Accurate diagnosis of the specific form of bone disease is of critical importance, as treatment modalities for each are different and frequently conflicting. Definitive diagnosis is possible only by comparative evaluation of clinical, radiological, biochemical and histological findings. It was quite apparent in the present case of a patient with multiple giant cell lesions in the orofacial complex, which on thorough examination and specialized investigation established parathyroid adenoma from the earliest oral manifestations.

CONCLUSION

Thus, the presented case of multiple giant cell lesions of the oral cavity, primarily affecting the jaw bones, in a middle-aged female patient where a thorough clinical examination, and systematic specialized investigations were the key to a successful diagnosis of an underlying rather serious medical disorder, primary hyperparathyroidism. We would like to emphasize that the oral lesions should be critically evaluated for the diagnosis of underlining any systemic disorder which often could be missed by general practitioners.

REFERENCES


