Pimary hyperparathyroidism as central giant cell granuloma of the jaws: Pre- and post-treatment pattern of clinical and radiographic presentation

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ABSTRACT

Introduction: The parathyroid glands regulate serum calcium and phosphorous levels by its secretion and maintenance, within physiological limits, of its hormone parathormone (PTH). Hyperparathyroidism is a metabolic disorder resulting from excessive secretion of parathyroid hormone. Hyper secretion might occur either due to a primary pathology in the glands (or) due to secondary causes. Central giant cell lesions occur in jaw bones. Case Report: A 20-year-old boy reported with complaints of swelling in left maxilla. Intra orally swelling extended from left upper canine till left upper second premolar and caused expansion of the cortical plates. Intra oral radiograph and orthopantomograph showed a poorly defined radiolucent lesion in between left upper canine and left upper 1st premolar with multiple cystic cavities in the region of the symphysis, left body and right angle of mandible. The radiographs of long bones showed osteoporosis. Serum alkaline phosphatase level was raised. Histopathology was reported as central giant cell granuloma. Nuclear scan of parathyroid showed a functioning parathyroid adenoma. The swelling in left maxilla regressed on its own after the adenoma of the parathyroid was excised. Conclusion: Timely diagnosis of parathyroid adenoma results in total regression of intra oral swelling and further progression of osteoporosis and fractures of long bones can be prevented.

Keywords: Central giant cell granuloma, Brown tumor, Hyperparathyroidism

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INTRODUCTION

The parathyroid glands regulate serum calcium and phosphorous levels by secretion and maintenance, within physiological limits, of its hormone parathormone (PTH). Secretion of PTH is mainly controlled through interaction of calcium with calcium sensitive receptors on the membrane of the parathyroid cells. Hyperparathyroidism is a syndrome of hypercalcemia resulting from excessive secretion of parathyroid hormone. A case of central giant cell granuloma in left maxilla due to adenoma of parathyroid gland is presented.

CASE REPORT

A 20-year-old boy reported with complaints of swelling in the left upper jaw (Figure 1). The swelling
was initially smaller six months ago and gradually increased in size. It was asymptomatic since onset and had not regressed in size at any time. There was no history of discharge from the swelling. No history of paresthesia or fever. The past medical history and surgical histories were unremarkable. Patient was moderately built, well nourished, calm and cooperative. His growth appeared to be retarded to his age. His vital signs were stable. A single left submandibular lymph node was enlarged, palpable, nontender, measuring 1 cm/1 cm in size, firm in consistency and mobile.

Extra oral examination revealed a solitary swelling, with well defined margins, measuring 3 cm/3 cm in diameter in the region of the left maxilla (Figure 2). It was irregular in shape, with a smooth surface extending anteriorly till nasolabial fold, posteriorly till the lateral margin of the eye, superiorly till the infra orbital margin, and inferiorly in line with angle of the mouth. It was nontender, hard in consistency. The skin over the swelling appeared normal. There was no local rise in temperature. The swelling was not mobile and it was fixed to the underlying structure.

Intra oral examination (Figure 3) revealed a swelling extending from left upper central incisor to left upper second premolar. There was expansion of the buccal and palatal cortical plates. Margins were well defined. It was firm to hard in consistency, nontender. Mucosa over the swelling was normal. It measured 1.5 cm/2 cm in diameter. The left upper canine and first premolar were decayed and the vitality test showed a delayed response.

Based on the history and clinical findings a provisional diagnosis of adenomatoid odontogenic tumor and a differential diagnosis of central giant cell granuloma, ameloblastoma, fibro-osseous lesion were given.

Intra oral radiographs (Figure 4). Showed the presence of a poorly defined radiolucent lesion in the region of left upper 1st premolar with divergence of roots of left upper 1st premolar. Orthopantomograph (Figure 5) showed the presence of a poorly defined radiolucent lesion in the region of symphysis, left body and right angle of mandible. Radiographs of long bones, chest, skull and ultrasonography of abdomen were done. Radiographs of long bones (Figure 6) were suggestive of osteoporosis. No significant findings were revealed in ultrasonography of abdomen.

Biochemical investigations revealed the following: Serum calcium: 9.4 mg%, Serum phosphorus: 3.1 mg%, serum alkaline phosphatase: -2559 mg/mL.

Incisional biopsy was done and histopathological examination showed presence of mature bundles of connective tissue with plump fibroblasts, interspersed with numerous multinucleated giant cells. Overlying epithelium appeared hyperplastic, with few bundles of blood vessels. The lesion was suggestive of central giant cell granuloma (Figure 7).

Thereafter the patient was referred to an endocrinologist to rule out hyperparathyroidism. After general examination of the patient by the endocrinologist, serum parathyroid estimation was done and was found to be elevated. Nuclear study of the parathyroid glands (Figure 8) was done with Tc-99m MIBI injected intravenously and it showed the presence of a functioning parathyroid lesion in the region of the lower pole of the left lobe of thyroid. An incisional biopsy was taken from the left. Inferior parathyroid gland which was suggestive of parathyroid adenoma. The tumor was surgically excised and post-operatively the patient was treated with IV calcium and calcium supplements, and multivitamin tablets.

The patient was periodically reviewed there-after at an interval of three months. The serum calcium, alkaline phosphatase, serum phosphorus returned to their normal limits.

The central giant cell granuloma in the region of the left maxilla regressed on its own (Figure 9, 10).

Intraoral radiographs, Occlusal X-ray (Figure 11), OPG (Figure 12), were taken which showed disappearance of the previous lesion. The multiple cystic cavities that were noticed in the mandible too disappeared.

Figure 1: Photograph of face shows swelling in the left maxilla.
DISCUSSION

Hyperparathyroidism is a metabolic bone disease. Primary Hyperparathyroidism is a disease, in which the parathyroid gland secretes excessive quantities of parathormone (PTH), due to increased activity of the gland due to (1) hyperplasia of the gland, (2) adenoma of the gland, (3) functional carcinoma of the parathyroid. Secondary hyperparathyroidism is secondary to chronic renal failure, rickets and osteomalacia [1].

Hyperparathyroidism is a relatively rare disease which is three times more common in women than men. It usually occurs in middle age, but might occur occasionally in children or in later life. Clinically, pathologic
Fractures may be the first symptom of the disease, although bone pain and joint stiffness are frequently heard early symptoms. Urinary tract stone is also a significant early finding [2].

Intraorally, the first sign of the disease may be a giant cell tumor (Brown tumor) or a cyst of the jaw. Brown tumor represents a giant cell reparative reaction. The loss of phosphorous and calcium results in generalized osteoporosis with attempts to repair the bone by new bone formation. The new bone may be resorbed and the resorption may lead to pseudocyst formation. Then the proliferation of granulation tissue from the cystic cavity occurs. As the area of bone resorption undergoes replacement by fibrous tissue, hemorrhage may occur. Small multinucleated giant cells may appear in an attempt to remove the blood. Hemosiderin in blood gives the lesion a brown color. Malocclusion caused due to sudden drifting of teeth may be the first sign of the disease [3-7, 8, 11, 12-14].

Radiographs show the presence of small or large sharply defined radiolucencies suggestive of cysts in the maxilla or mandible. Some lesions show the classical ground glass appearance. The lamina dura around the teeth may be partially lost. Multifocal involvement of other bones in the body might also be seen [8, 11, 12]. The serum calcium is raised as high as 12 to 20 mg per 100 mL, while the serum phosphorus is lowered to 2 mg or less per 100 mL. If bone lesions are present serum alkaline phosphatase and serum parathormone levels
are usually, raised, and urinary output of calcium is considerably increased [11, 12]. Histologically, multinucleated osteoclast like giant cells bone lesions show lying in hemorrhagic fibrous tissue. Deposits of hemosiderin may be seen. Multiple cystic cavities are noted [5, 6].

CONCLUSION

To conclude hyperparathyroidism is a metabolic bone disorder that might occur either due to hypertrophy (or) adenoma of the parathyroid gland, that manifests intraorally as a central giant cell granuloma. Timely diagnosis, and treatment of the parathyroid lesion, results in total regression of the intraoral lesion on its own [10]. Further progression of osteoporosis and pathological fractures can also be prevented. Therefore if any case of central giant cell granuloma involving the jaw bones is reported, it is mandatory to rule out hyperparathyroidism. There is no need to surgically excise the lesion as the lesion will regress if the pathology in the parathyroid gland is corrected.

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Author Contributions
Nalini Aswath – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Pravda Chidambaranathan – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor
The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

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REFERENCES