

Role of Oral Physician in Diagnosis of Occult Disease of Primary Hyperparathyroidism

Abstract

Parathyroid hormone (PTH) plays a key role in the regulation of calcium homeostasis. It is secreted by a pair of parathyroid glands located behind the thyroid gland. Primary hyperparathyroidism is the disorder which is seen in 0.2%–0.3% of the population. It is the third most common endocrine disorder after Diabetes Mellitus and Thyroid disorders. There are several systemic manifestations of the disease including skeletal, Renal, abdominal, neurological ones. The following case report discusses some of the classic oral manifestations of the disease and signifies the role of an Oral physician in the diagnosis of underlying systemic disorder.

Keywords: Oral manifestations, parathyroid hormone, primary hyperparathyroidism

Introduction

Primary Hyperparathyroidism is an endocrine disorder caused by the excessive secretion of the parathyroid hormone, which is an integral component in the maintenance of normal calcium homeostasis.^[1,2] Presence of excess PTH creates an imbalance between osteoblastic deposition and osteoclastic resorption causing an increased bone modeling.^[3] PHPTH is most commonly caused by parathyroid adenoma, hyperplasia, carcinoma, multiple endocrine neoplasias, and other rare syndromic states.^[2,4] PHPTH has profound impact in the jaw bones demineralization of cortices of inferior border of mandible, Inferior alveolar nerve canal, walls of maxillary sinus, presence of radiolucencies in the bone, alteration in the bony trabeculae, presence of brown tumors.

The aim of the article is to emphasize upon definitive oral manifestations of PHPTH, which in conjunction with certain systemic findings and biochemical analysis aids the clinician to ascertain the diagnosis of a systemic disorder unknown to the patient and modify the dental management accordingly.

Case Report

A 35-year-old female patient reported to the hospital with the chief complaint of painless

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swelling in the lower front region and lower left back region of the jaw for 3 months. She also complained of generalized joint pain, abdominal pain, and lethargy. The swelling was abrupt in onset, gradually increased in size and attained the present day size. Clinical examination revealed two well-defined, bony hard, nontender swellings in mandibular anterior and left posterior regions with the obliteration of labial and buccal vestibule. The mucosa over the swellings was completely normal. On examination of the teeth, it was found that there was presence of extrinsic stains on all the mandibular teeth along with mild recession. The teeth associated with the swellings; 31, 32, 33, and 46.47 were vital in nature [Figure 1]. The provisional diagnosis of the above was stated as benign tumor of the oral cavity.

Intraoral periapical of the concerned regions revealed loss of lamina dura in all the teeth covered along with ill-defined areas of radiolucency [Figure 2a and b].

Orthopantomograph showed generalised loss of lamina dura, well defined, noncorticated radiolucent osteolytic lesion in mandibular left posterior region, reduced thickness and density of cortication at the periphery of the mandible, inconspicuous outlines of inferior alveolar nerve canal [Figure 2c].

Cone Beam Computed Tomography: Axial and Coronal sections showed

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Figure 1: Intraoral swelling on the labial aspect of 31, 32 and on the buccal aspect of 36, 37

multiple osteolytic, hypodense areas in the mandible [Figure 2d and e].

Skeletal radiographs showed osteolytic lesions in the proximal phalanx below the interphalangeal joint and metacarpophalangeal joint [Figure 2f and g].

The patient was subjected further to certain blood investigations as shown above [Table 1].

Based upon clinical, radiographical, and biochemical analysis, the diagnosis was stated as “Hyperparathyroidism” with the following differential diagnosis:

1. Paget’s Disease: It is seen in patients above the age of 60 years with generalized radiopacity, expansion of jaws with no loss of lamina dura
2. Osteomalacia
3. Osteoporosis
 - Both show a generalized radiolucency of jaw bone with thinning (no loss) of lamina dura. Serum 25(OH) D levels are reduced in patients with osteomalacia which were found to be normal in the above patient.
4. Langerhans cell disease: Presents as solitary or multiple, “scooped out” lesions in the jaw bones, periosteal reaction and localized loss of lamina dura.

The patient was referred to the endocrinologist who subjected the patient to ultrasonography (USG) and Biopsy of the parathyroid gland. USG revealed a hypoechoic structure close to the thyroid gland. Biopsy was suggestive of a parathyroid adenoma. Final diagnosis

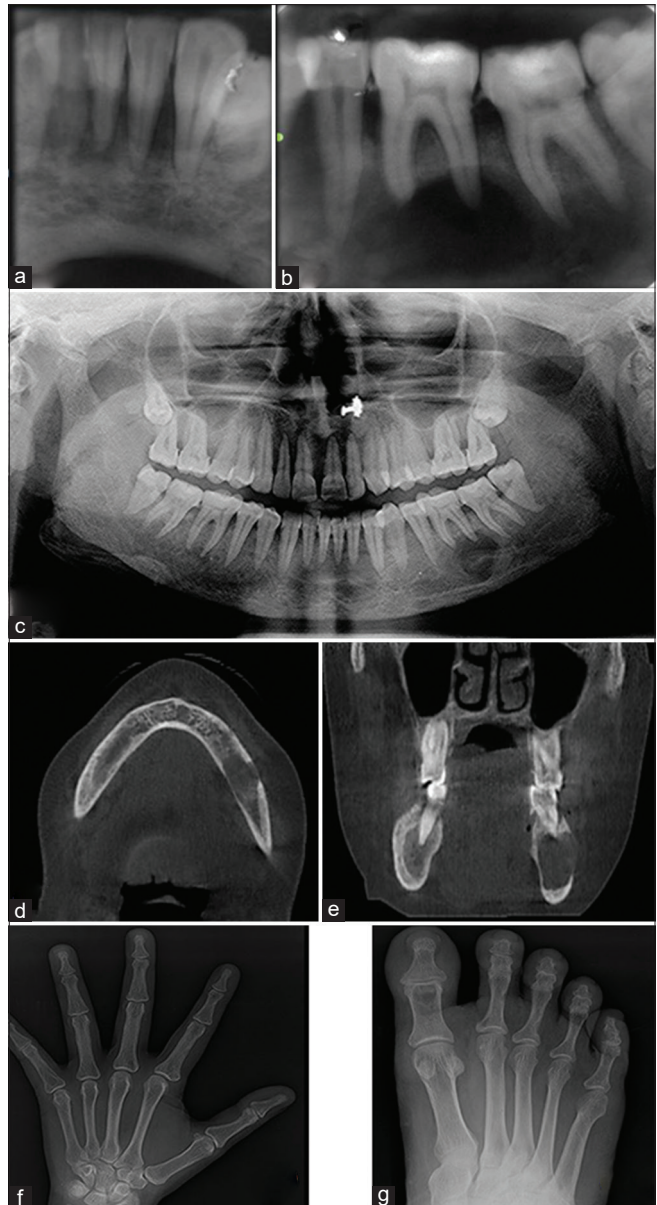


Figure 2: (a and b) Intraoral periapical showing non traceable lamina dura and an ill-defined radiolucent lesion (c) orthopantomogram showing well-defined radiolucent osteolytic lesion with generalized loss of lamina dura. (d and e) Cone beam computed tomography showing osteolytic areas in the mandible causing perforation (f and g) hand wrist and foot radiograph showing osteolytic lesion

of the above case was given as “PHPTH.” Patient was prescribed sodium alendronate 35 mg OD, cinaclet 30 mg OD, calcitriol 0.25 mg OD. The parathyroid levels did not reduce considerably within 3 months. The patient underwent the surgical procedure of Minimally invasive Parathyroidectomy followed by an uneventful recovery. The timeline of the above Case Report is as follows: [Table 2].

Discussion

It dates back to 1705 where Courtial, a French surgeon gave the complete description of a patient with HPTH. Von

Table 1: Blood Investigation Reports

Tests	Patient Values	Normal Values	Interpretation
Parathyroid hormone	1341pc/ml	10-69pc/ml	Significantly raised
Alkaline phosphatase	1508 units/L	38-126units/L	Significantly raised
Serum Calcium	10.8 mg/dL	9-11 mg/dL	Normal
Serum Phosphate	1.9 mg/dL	2.5-4.5 mg/dL	Normal
Serum 25(OH) D	45 ng/mL	25-80 ng/mL	Normal

Table 2: Timeline of the Case report

1	Intraoral swellings in the mandibular anterior and left posterior region	3 months (History)
2	Joint pain, Fatigue and Lethargy	2 months (History)
3	Increase in the size of swellings	10 days (History)
4	Non tender bony hard swellings Provisionally suspected as Benign tumor of the oral cavity.	Clinical Examination
5	IOPA, OPG and CBCT showed osteolytic lesions, generalized loss of lamina dura, thinning of cortical bone.	Radiographic Examination.
6	Biochemical analysis showed highly elevated PTH and Alkaline Phosphatase levels. Suspected case of Hyperparathyroidism.	5 days post examination.
7	Referral to an endocrinologist, USG & Biopsy of parathyroid gland revealed Parathyroid Adenoma. Final diagnosis of Primary HPTH given. Pharmacological treatment plan opted using Tab Sodium alendronate 35mg OD, Tab Cinaclet 30mg OD, Tab. Calcitriol 0.25mg OD.	10 days post examination
8	No considerable reduction in PTH levels after therapeutic intervention.	3 months after pharmacological therapy
9	Minimally invasive parathyroidectomy was performed.	3.5 months after pharmacological therapy
10	Follow up revealed considerable decrease in the PTH hormone levels from 1341pc/ml to 150pc/ml.	30 days post surgery.

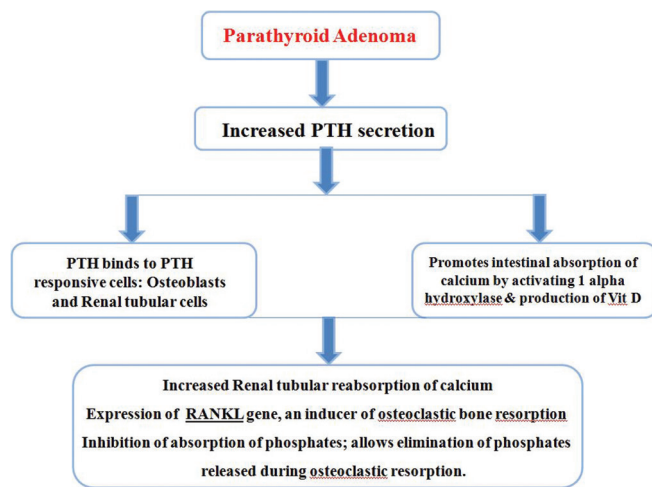


Figure 3: Pathophysiology of primary hyperparathyroidism

Recklinghausen reported three patients with Osteitis fibrosa cystica (OFC).^[5]

The pathophysiology of PHPTH has been described above^[6,7] [Figure 3].

The classic description of clinical manifestations of PHPTH is ‘Stones, Bones, Groans, Psychic Moans, Fatigue overtones.’^[8,9] In India, females are affected greater than Males with a ratio of 1.7:1 with a majority of cases <40 years of age.^[9]

An array of oral manifestations can be found in patients with PHPTH which include loss of lamina dura, bony pathologies in jaw bones, cortical destruction, changes in trabecular pattern, root resorption, tooth displacement.^[10]

OFC is an extensive osteoclastic activity causing resorption in the bone manifesting as a radiolucency. “Brown tumors” are nonneoplastic lesions that replace bone marrow with fibrous tissue and contain multinucleated giant cells, spindle cell stroma, extravasated blood cells and hemosiderin deposition thus imparting brown color. The dentist should consider the risk of bone fractures while performing the surgical procedures.^[7]

Earliest change includes subperiosteal resorption at phalanges, OFCs at multiple sites such as Skull, radius, Phalanges, femur reducing patients’ bone mineral density and making them prone to fracture. Renal complications such as Nephrolithiasis and Nephrocalcinosis, Psychiatric symptoms, Cardiac symptoms such as coronary artery disease, conduction abnormalities.^[1,10]

PHPTH is a challenging disease to treat. Cinaclet, a calcimimetic agent lowers the serum calcium and PTH levels. Bisphosphonates and Estrogen therapy act as antiresorptive drugs and increase the bone mineral density.

The above case was approached in a streamlined manner with the involvement of Oral Physician, Endocrinologist, General Surgeon leading to an accurate diagnosis and

treatment plan. The limitation of the above case was that the “Single photon emission computerized tomography” images could not be obtained as the patient did not undergo the investigation due to certain financial problems.

The patient solely came with the purpose of treating the oral lesions, completely unaware about the underlying systemic disease. Therefore, the statement, “Mouth is the mirror of the body” is justified from the above case.

Early and apt diagnosis by the dentist along with interdisciplinary approach limited the progression of the disease and saved the patient from further systemic complications. This emphasizes upon the role of oral physician in diagnosing an occult systemic disease.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient (s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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