

Multiple impacted permanent teeth, an indicator for early detection of hypoparathyroidism: A rare case report

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ABSTRACT

Eruption is a process of continuous movement of the developing tooth bud from its developmental location to functional location. Teeth that cease to erupt before emergence to their functional position in the oral cavity are termed as impactions. In permanent dentition, third molars are the most frequently impacted teeth followed by the canines. When impaction involves few teeth, the condition is localized but when it involves multiple teeth, the condition becomes generalized and is often associated with some derangement of the normal physiological processes. Factors causing impactions may be localized, pertaining to the area or, systemic or, generalized including bone disorders such as cleidocranial dysplasia and/or some sort of endocrinological disturbance such as hypoparathyroidism. Hypoparathyroidism is a rare endocrinological disorder accompanied by anomalies of various systems including bones and teeth. The dental defects due to hypoparathyroidism may present as hypocalcemia, aplasia and/or hypoplasia, defects of mineralization, short and blunted roots, delayed eruptions, and clinically missing or impacted teeth. This report describes an interesting and unusual case where multiple impacted permanent teeth and retained primary teeth accompanied by other clinical manifestations in a 16-year-old female patient probed the clinicians for further investigations which, eventually, aided in early diagnosis of hypoparathyroidism.

Keywords: Early detection, hypoparathyroidism, multiple impacted permanent teeth, retained primary teeth

Introduction

Hypoparathyroidism, an uncommon endocrinological disorder, is identified either by the absence or abnormally low sera levels of parathyroid hormone (PTH) secondarily leading to abnormally low sera levels of calcium and/or elevated sera levels of phosphorus in the blood. It presents as a major therapeutic challenge which includes an effectual management of the balance between treating the hypocalcemia and

simultaneously avoiding hypercalciuria. The exact etiology of this disorder is attributed to hereditary (autosomal dominant, recessive, and X-linked), autoimmune, and various other acquired etiologies.^[1] Hypoparathyroidism has known adverse effects on the development of teeth and bones including their eruption manifesting as multiple impacted permanent teeth and retained primary teeth accompanied by numerous other clinical manifestations, impaired upper and lower jaw growth, alteration of facial dimensions and a shortfall of coordination in lower jaw growth which affects the overall development of the dentition and the final state of occlusion achieved.^[2] It may also present as

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brown tumors apart from eruption delays and teeth with short and blunted roots.^[3]

A tooth is termed as impacted if it fails to proceed from its developmental location inside the jawbone to its eventual, functional location in the oral cavity. Various factors at genetic, molecular, and cellular levels interplay in a successful tooth eruption.^[4] It is not a rarity to find impaction of a single tooth but a rarity to find multiple impacted teeth. Impactions can be seen secondary to physical reasons such as insufficient space, early closure of space, crowded arches, thickened overlying bone, soft tissues, or a plethora of systemic conditions including syndromes, metabolic and hormonal derangements, and rarities.^[5]

As hypoparathyroidism presents early in the form of diverse oral manifestations, an oral healthcare physician can play a major role in early diagnosis and a multidisciplinary management approach of this disorder. The present case report elaborates a rare case where multiple impacted permanent teeth and retained primary teeth accompanied by other clinical manifestations probed the clinicians for further investigations which, eventually, aided in early diagnosis of hypoparathyroidism.

Case Report

A 16-year-old female patient reported as an outpatient to the Department of Oral Medicine and Radiology with a chief complaint of multiple absent teeth. Her dental history revealed usual loss of primary teeth with subsequent failure of eruption of the permanent teeth leading to difficulty in mastication. Her family history was positive for a consanguineous marriage of her parents.

On general physical examination, the patient was found cooperative, of moderate built, and well-oriented to the time and place and the surroundings. Bilateral feet examination revealed an abnormal decrease in the size and diameter of the third, fourth, and fifth phalanges. The lateral ulnar surface of the right hand showed a mass of soft tissue along with a prominent middle phalanx of the long finger [Figure 1]. No other abnormalities were observed in relation to her physical or mental abilities. Intraoral examination revealed retained primary teeth in relation to tooth # 63, 65 with multiple clinically missing permanent teeth in relation to tooth # 17, 15, 14, 12, 11, 22, 23, 24, 25, 27, 33, 34, 35, 44, and 45 [Figure 2]. Based on the noted clinical presentation, a provisional diagnosis of oligodontia of the maxilla and mandible was made while the list of differential diagnoses included hypothyroidism, hypoparathyroidism, cleidocranial dysplasia, and Gardner's syndrome as the commoner conditions associated with such type of clinical presentation.

A series of radiographic and serological examinations were planned. Hand-wrist radiograph of the patient in relation to right hand revealed alteration in the bone architecture with mild changes in the trabecular pattern and in the thickness of the cortices, while orthopantomograph (OPG) of the patient revealed multiple impacted permanent teeth with retained

primary teeth in both the jaws at various levels of impactions in the anterior and posterior areas of the mandible, maxillary tuberosity, nasal floor, and zygomatic region. No evidence of cystic changes was observed clinically and radiographically. The maxillary and mandibular bones displayed a mild ground glass appearance with faint trabecular pattern and an altered density. Mild blunting of mandibular molar roots was also evident in the OPG of the patient [Figure 3].

Serological investigations done to evaluate serum calcium, phosphorous, serum alkaline phosphatase, PTH and thyroid



Figure 1: Clinical photograph of feet (upper) and right hand (lower left-front view and lower right- rear view of same) showing abnormal third, fourth and fifth phalanges in the feet, being almost half of the adjacent fingers, with right hand showing a soft tissue mass in the lateral ulnar surface along with prominent middle phalanx of long finger



Figure 2: Intra-oral photographs (left- maxillary arch; right- mandibular arch) showing multiple, clinically missing permanent and retained primary teeth



Figure 3: Hand-wrist radiograph of right hand (left) revealing alteration in bone architecture with mild changes in trabecular pattern and in the thickness of the cortices with orthopantomograph (OPG) (right) revealing multiple unerupted and impacted permanent teeth and retained primary teeth in both jaws

hormone levels, and vitamin D profile revealed a decrease in calcium (8.15), increase in phosphorous (6.49) and thyroid-stimulating hormone (6.79), and markedly decreased PTH (<1.2) levels. Based on clinical presentation, abnormal phalanges in the feet bilaterally, radiological, and serological investigations, a final diagnosis of hypoparathyroidism was eventually arrived at.

After counseling, the patient was advised to undergo thyroid function tests. The immediate target was to achieve normal sera calcium levels. A multidisciplinary approach was adopted for treatment plan. After basic oral prophylaxis was performed, all retained deciduous teeth were extracted. As most of the impacted teeth were vertically oriented, orthodontic procedure was drafted. Oral rehabilitation and visit to physician for an effective management of the disorder were planned. The patient is still on follow-up and ongoing treatment is uneventful.

Discussion

Tooth impactions comprise a common phenomenon; however, impaction of multiple teeth is relatively rare. The commoner reasons for impactions are often linked to an idiopathic etiology and local or systemic causes. The physical reasons of insufficient space, early closure of space, crowded arches, thickened overlying bone, or soft tissues are the common local etiologies cited for single or isolated and impactions of a group of teeth. Multiple impacted permanent and retained primary teeth are usually seen secondary to a plethora of systemic conditions including syndromes, metabolic and hormonal derangements, and rarities. Idiopathic causes can be due to abnormal eruptive forces, trauma to the developing tooth buds, and intrinsic defects in the mechanism of eruption process.^[6]

Hypoparathyroidism, an uncommon endocrinological disorder, identified either by the absence or abnormally low sera levels of PTH, has varied etiology including genetic, autoimmune, surgical/iatrogenic, etiology secondary to treatment with radioactive iodine treatment, and/or certain systemic diseases and syndromes including the more common DiGeorge, Shprintzen and velocardiofacial syndromes.^[7] A decreased level of PTH leads to calcium and phosphorus imbalances leading to hypocalcemia and hyperphosphatemia. Hypocalcemia commonly manifests as tetany, myalgias, dysphagia, irritability, anxiety and/or depression, psychosis, and convulsions. The prominent clinical signs associated with hypocalcemia are those of hyperreflexia including the well-known Chvostek's and Trousseau's signs.^[8] All these clinical features were, though, absent in the present case. The characteristic oral manifestations of the disorder include delayed eruption of permanent and multiple retained primary teeth, microdontia, hypoplastic enamel defects, poorly calcified dentin, widened pulp chambers, and pulp calcifications along with short and blunted roots that were clearly evident in the present case.

The present case revealed disturbances in the eruption pattern of the permanent teeth with multiple retained primary teeth.

The variations in formation and the eruption pattern of teeth can be critical in guiding additional investigations in the disorder.^[9] Furthermore, the present endocrinological disorder characteristically presents with abnormally short fourth and fifth metatarsal and metacarpal bones with rare involvement of the third metatarsal or metacarpal bone which might also get affected. Hand and foot radiology, thus, almost frequently, shows quite significant and nearly diagnostic changes in the third, fourth, and fifth metatarsal and metacarpal bones which can be almost half the magnitude of the adjacent, normal bones.^[10] The management of the disorder often involves oral and intravenous calcium supplementations, high doses of vitamin D supplementation as calcitriol, and a diet rich in calcium with low phosphorus. The active component of human PTH, teriparatide, developed recently can also be given subcutaneously. Dental procedures can be started as per the specific need of the patient under proper medical supervision and with periodic recalls for follow-up of the changing needs of the patient in treatment.^[11]

Hypoparathyroidism is a disorder that shows early dental manifestations and it requires a careful evaluation through clinical, radiological, and laboratory examinations. An oral healthcare physician must be vigilant enough to check for early or-dental manifestations including multiple impacted permanent and retained primary teeth and radiological observations of enamel hypoplasia, poorly calcified dentin, abnormally wide pulp chambers, and short and blunted roots. In addition, a short third, fourth, and fifth metatarsal and metacarpal bones can be suggestive of a hidden systemic disease. To conclude, an early detection of the disorder is a key to successful management, and oral healthcare physicians can play a vital role in it.

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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