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Case of Multiple Impacted Permanent, 12 Supernuemeries and Completely Retained Primary Teeth with Transient Alkaline Phosphatemia

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Abstract

Syndromes and associated tooth agenesis and impaction are rarely encountered in clinics. Non-syndromic cases with multiple impacted and almost all retained teeth with multiple superneumeries with a transient hyper enzymatic condition is rarest. Lack of eruptive force and multiple superneumeries may cause multiple impactions and retention of primary teeth.

This case report presents a 14-year-old boy with 19 retained primary teeth, 17 impacted permanent teeth and 12 superneumeries and a high normal condition of alkaline phosphatemia. Based on the clinical appearance, case history evaluation, radiographic examination and biochemical reports, this paper discusses differential diagnosis and management of such cases.

Various tests and literature analysis lead us to a conclution that this case is a non-syndromic hyper alkaline phosphatemic condition with multiple retained deciduous, impacted multiple permanent and superneumery teeth. Additional examinations and long-term evaluation may be necessary to identify this condition genetically and exclude systemic illness.

Keywords: multiple superneumeries, multiple impaction, retained primary teeth, non-syndromic, alkaline phosphatemia.

Introduction

Eruption is a process of axial or occlusal movement of the tooth from its developmental position within the jaw to its functional position in the occlusal plane.[1]Impacted teeth is one that cease to erupt before emergence to oral cavity. Reasons causing delayed eruption commonly are insufficient space, early loss of primary teeth with eventual closure of space, crowding of arches rotation of tooth buds, excessive fibrous tissue over an erupting tooth

and eruption cysts being relatively rare[1]. Impaction of teeth can result firstly from local biomechanical impediments, and secondarily from childhood maxillofacial or dentoalveolar trauma, reconstructive surgery of the facial skeleton, malposition of an adjacent tooth, thick osseous or mucosal tissues above teeth, insufficiently developed jaw bone or the difference in the rate of development and maturation of jaw bones and teeth respectively, disturbances in eruption and direct or indirect effects of cysts or neoplasm of jaws.[2]

This case discussion is meant to share a clinical experience and spread awareness of situations of transient hyper alkaline phosphatemia and multiple retained and impacted teeth without detection of any syndrome or systemic conditions as it may be useful for early clinical diagnosis and avoidance of unnecessary concerns and investigations.

Case Report

A 14-year-old male patient presented at our outpatient department with a chief complaint of non-exfoliation of milk teeth. On physical examination patient appeared to be underweight but intelligence was subjectively normal and didn't revealed symptoms pointing to any syndromes or skeletal abnormalities. A mild malar hypoplasia was noted facially. Head hair and body hair were clinically normal. His pregnancy and delivery were normal and uneventful. There was no significant medical or dental history of the patient's family. Intraoral examination revealed the presence of 30 teeth. (Figure 1 and 2). Out of which 19 were retained deciduous. All deciduous teeth were retained expect maxillary left lateral incisor. No oral mucosal lesions were detected. Patient was sent for routine blood tests and panoramic radiographs. Routine blood tests were within normal range. Panoramic radiograph (Figure 3) and CBCT (Figures 4 and 5) revealed 29 impacted teeth (17 permanent and 12 superneumeries), developing third molars were observed in all quadrants. Maxillary impacted permanent teeth were 15, 14, 13, 12, 11, 21, 23, 24, 25 and the mandibular impacted permanent teeth were 32, 33, 34, 35 and 42, 43, 44, 45. Impacted superneumeries were observed palatally with 54,53,11 and 21, 63, 64 regions in maxillary arch and lingually with 85,84,83 and 73,74,75 region thinning of palatal cortical bone was observed with maxillary superneumeries and lingual cortical thinning with mandibular superneumeries. Buccal cortical bone plate was associated with all impacted permanent teeth. Apical third root dilacerations was noted with impacted 33, 32 42, 43. Thinning of inferior border of mandible is observed with relation to impacted 43 and 33. No cystic changes were detected and the periodontal spaces and lamina dura of most of the teeth were not visible. The jaw bones showed the normal trabecular pattern. He was sent to general physician and further referred to ophthalmologist. Further investigations were carried out for excluding DD by doing T_3, T_4, TSH and PTH tests which seemed to be normal. All his routine checkups were normal except for alkaline phosphatase which seemed to be of higher value. Kidney function tests were advised which later turned out to be normal. By these steps, chances for any syndrome or systemic illness were ruled out. Patient and parents were explained about the situation and then aware about the retained deciduous, multiple impacted permanent and superneumery teeth.

Treatment Plan

The management of non syndromic multiple impacted permanent teeth and supernumerary teeth along with multiple retained primary teeth have been planned to be done as coordinated management for guided eruption of as many teeth as possible. A series removal of primary teeth and surgical removal of superneumeries followed by a waiting period of 1month for prognosis evaluation in

eruption of permanent teeth using radiographs. If eruption isn't favored orthodontic treatment of impacted permanent teeth for proper alignment to their proper position is planned

Discussion

Literature have a good collection of multiple impacted permanent teeth and superneumeries[3-7] .Bayar et al[8] have reported three cases with multiple impacted teeth involving both jaws in which no syndrome or systemic conditions have been detected. Multiple impacted teeth may be related to syndromes like cleidocranial dysostosis[9], Gardner syndrom[10],Yunis – Varon syndrome.[11]Down syndrome, Aarskog syndrome, Zimmerman-Laband syndrome and Noonan's syndrome.[12] also metabolic disorders like mucopolysaccharidoses.[13] and hormonal disorders like hypothyroidism, , , hypothyroidism, hypopituitarism, Vitamin D-resistant rickets. The most common syndrome with impaction is cleidocranial dysplasia[14].

In molecular level analysis, numerous eruption-regulating molecules having similar and overlapping functions EGF, EGF-R, CSF-1, CSF-1R, IL-1, IL-1R, c-Fos, NFB, MCP-1, TGF-β1, PTHrP, Cbfa-l (now called Runx2), OPG, RANK/RANKL are there. But even the absence of a single factor does not interrupt the event of eruption. Defect in some genes can also be responsible for this condition. Stellate reticulum and dental follicle are the sites for these molecules[15.]

A prevalence of 0.1% and 3.8% is shown by superneumeries. A single supernumerary occurs in 76%–86% of cases, 2 supernumerary teeth are present in 12%–23% of cases and 3 or more supernumeraries are found in only 1% of cases[16]. single supernumerary tooth occurs most frequently in the maxillary midline as mesiodens, followed by the supernumeraries of maxillary molar region and mandibular premolar region. Supernumerary premolars account for 10% of all supernumeraries, and nearly 75% of them occur in the mandible.[17] Though exact etiology of the supernumerary tooth is not clearly known. Numerous theories such as hyperactivity of dental lamina theory, genetic theory, atavism theory, phylogenetic theory, dichotomy of tooth bud, progress zone, and combination of genetic and environmental factors have been proposed[12].

The literature points that similar cases were reported by Nagaveni[18] et al. and santhanu[12] both of the study observed multiple impacted permanent and supernumerary teeth in a nonsyndromic patient. Dilacerations associated with impacted teeth have been discussed in the literature. Kaban et al.[19] states that the follicle of unerupted molars near the inferior border begins to curve, resulting in "hooked" roots. in this case prognosis is worsened by the dilacerations of mandibular impacted anteriors.

Hyperphosphatemia is a condition of elevated alkaline phosphatase. It is commonly seen in kidney disorders, parathyroid hormone imbalance and bone disorders. Transient Hyperphosphatasemia (TH) is a benign condition in which serum alkaline phosphatase (ALP) is transiently elevated in the absence of other systemic diseases. It mainly occurs in infants and children and very rarely seen in adults[20]. TH has also been reported in association with viral infections such as respiratory syncytial virus, enteroviruses, Epstein-Barr virus, and human immunodeficiency virus (HIV); following liver or kidney transplant and in children on cyclosporine or chemotherapy for leukemia and lymphoma. The ALP level changes with age, it is generally higher in children than in adults, with a peak in the first six months of age and during pubertal growth spurt, because they have higher osteoblastic activity in this age. [21,22] Literature

points that the transient hyperphosphatemia are rare and those included with dental findings were not obtained. Since other syndromes and disorders cannot be connected to our patient this can be concluded as a transient high normal condition of hyperphosphatemia. Long term evaluation is required to confirming this.

By all these tests and literature analysis we concluded that this case is a non-syndromic hyper alkaline phosphatemic condition with multiple retained deciduous, impacted multiple permanent and superneumery teeth.

Relevance of this study

1. Understanding hyper phosphatemia as a transient condition in pubertal growthspurt.
2. Early diagnosis of nonsyndromic conditions.
3. Avoidance of unnecessary concerns and investigations

Conclusion

The present case was rare and unique in many ways. Along with failure of exfoliation of almost all primary teeth and multiple impacted permanent and 12 superneumeries which itself is a rare condition a transient state of high normal alkaline phosphatase was observed. Impacted teeth with dilacerations and thinning of cortical plates will be the challenge facing during interdisciplinary treatment.

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Legends Figures

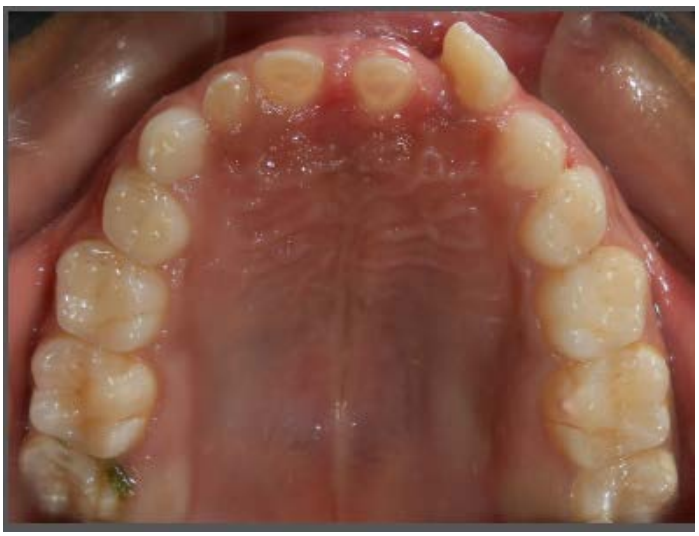

Figure 1: intraoral photograph of upper arch.

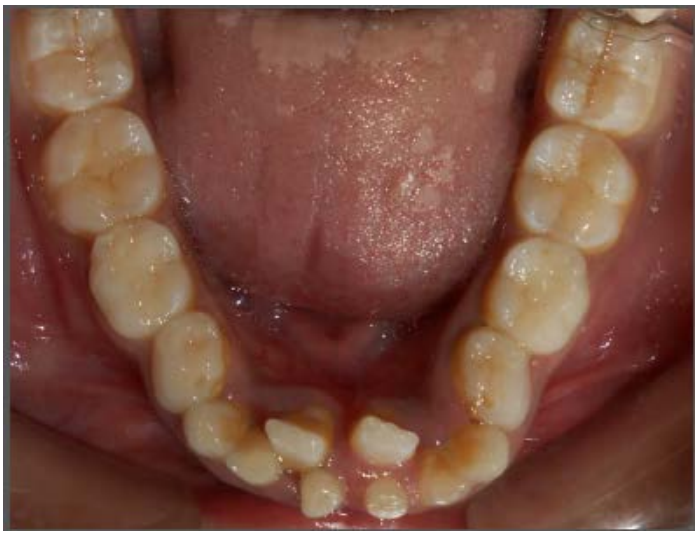

Figure 2: intraoral photograph of lower arch.

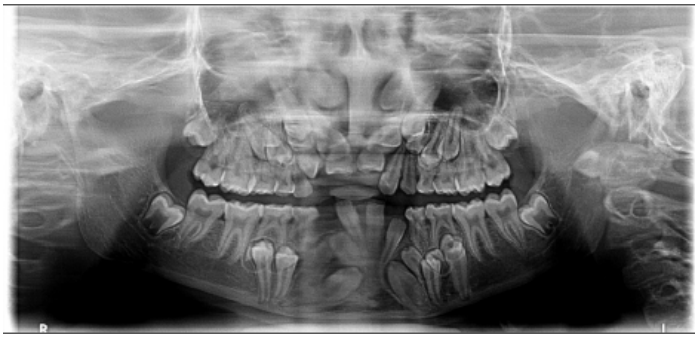

Figure 3: Panoramic radiograph after extraction of upper and lower incisors.

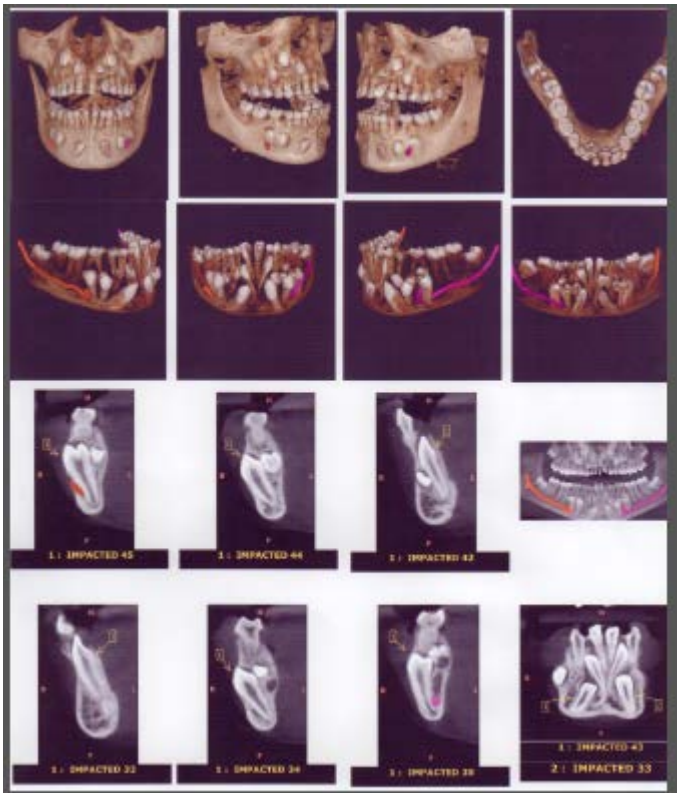

Figure 4: CBCT report 1

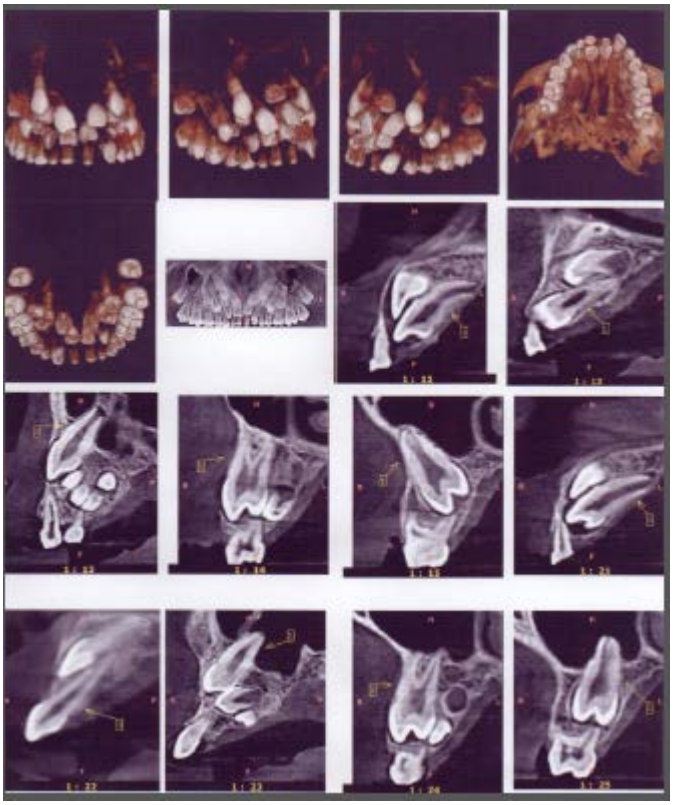

Figure 5: CBCT report 2