Single Family Members with Multiple Impacted teeth: A Rare Case Report

Nilesh Patil¹, Aruna Tambuwala², Deepak Kaul³, Rakesh Oswal³, K Rahul Kumar¹
¹Post-graduate Student, Department of Oral and Maxillofacial Surgery, M.A. Rangoonwala College of Dental Sciences and Research Centre, Pune, Maharashtra, India, ²Professor, Department of Oral and Maxillofacial Surgery, M.A. Rangoonwala College of Dental Sciences and Research Centre, Pune, Maharashtra, India, ³Reader, Department of Oral and Maxillofacial Surgery, M.A. Rangoonwala College of Dental Sciences and Research Centre, Pune, Maharashtra, India

Multiple impacted teeth of idiopathic origin are a rare dental anomaly. Various local and systemic causative factors have been implicated in the literature; however, it is unknown at present about the localization of the genetic defect in the phenotype of failure of eruption. Retained primary teeth is a well-known process, but multiple permanent and supernumerary teeth that too asymptomatic is surely a rare possibility. This article aims in to consolidate and organize the available information regarding the tooth eruption failure and to collaborate the current evidence with the report of three adult sibling’s cases of failure of eruption of multiple permanent teeth in a family without a known cause.

Keywords: Idiopathic, Multiple impacted teeth, Siblings, Surgery, Syndromes

INTRODUCTION

Tooth eruption is the axial or occlusal movement from its site of development within the alveolus to its functional position in occlusion.¹ Teeth eruption and shedding are continuous process, which replaces the exfoliated primary teeth with permanent teeth. Sometimes Impaired tooth eruption takes place in the form of delayed or complete absence of eruption, giving rise to impacted, embedded permanent teeth or retained deciduous teeth.²

Early loss of primary teeth with the eventual closure of space can be a result of impaction of teeth from local biomechanical impediments. Crowding and rotation of tooth buds and maxillofacial or dentoalveolar trauma in childhood, ectopic eruption of an adjacent tooth, thickened overlying osseous or mucosal tissues, discrepancy in maxillofacial skeletal development or lack of correlation between maxillofacial skeletal development and tooth maturation, disturbances in eruption pattern, and consequences related to direct or indirect effects of cysts or neoplasm are the causes of tooth eruption failure.³ Systemic causes such as Syndromes, metabolic, and hormonal disorders can also contribute to multiple impacted permanent teeth.⁴

Our aim and objective of this study is to increase awareness among surgeons that cases of multiple impacted teeth always have to be discussed under multiple etiologies and investigations.

CASE REPORT

A 40-year-old patient has been presented with extra oral draining fistula to the Department of Oral and Maxillofacial Surgery; his radiological investigations revealed multiple impacted teeth in the maxillary and mandibular arch. On further examination, the family history of the patient revealed that two of his siblings and one paternal aunt have similar conditions.

Case 1

A 40-year-old male patient reported to the Department of Oral and Maxillofacial Surgery with compliant of extra oral draining fistula in the submental region. Patient was well built and nourished with no relevant medical history. On extra oral examination, draining fistula was present in the submental region. Intra-oral examination showed a complete edentulous maxillary arch and only nine teeth were present in the mandibular arch. He had lower complete denture and fixed partial denture with an upper arch since 14 years. On radiological examination, 12 impacted teeth were seen. Patient’s past dental history revealed improper
exfoliation of deciduous teeth. The extra-oral fistula was surgically removed with extraction of impacted lower left canine (Figure 1a-c).

Case 2
A 42-year-old male patient came to the Department of Oral and Maxillofacial Surgery, with a complaint of pain and swelling in lower left front region of the jaw since 15 days. Patient was well built and nourished with no relevant medical history. Patient’s past dental history revealed improper exfoliation of deciduous teeth permanent teeth failed to erupt after exfoliation of deciduous teeth, except lower molars that were extracted 10 years back for denture prosthesis. On extraoral examination, there was a swelling present in lower left border in the submental region. On intra-oral examination, eight teeth were present. Intra-oral draining sinus was present with lower left premolar. Radiological examination shows 10 teeth impacted. Patient underwent surgical extraction of impacted lower right lateral incisor and canine, which were clinically exposed to the oral cavity (Figure 2a-c).

Case 3
A 45-year-old female patient came to the Department of Oral and Maxillofacial Surgery with a complaint of pain in lower anterior region of the jaw since 6 months. Patient was well built and nourished. Patient was hypertensive since 5 years and was on medication, amlodipine 5 mg. Past dental history revealed that he underwent extraction of over retained deciduous teeth 20 years back. On extraoral examination, there was no abnormality detected. On intra-oral examination, only one tooth was present in the oral cavity. Patient was denture wearer since 20 years. On radiological examination, there were fifteen teeth impacted. In the anterior region, an impacted tooth was exposed to the oral cavity causing pain that was surgically removed (Figure 3a-d).

Patients were referred to the physician, tests which included thyroid function test, growth hormone assays, serum calcium and phosphorus levels were carried out to rule out patient’s condition with any other associated syndrome either metabolic or hormonal disorder. After the clinical and radiographic evaluations, a treatment plan was made under which all the teeth were been extracted.

DISCUSSION

Epidemiologically 25-50% of the population shows multiple impactions.\(^5\)

Although impaction of teeth is widespread, multiple impacted succedaneum teeth along with numerous retained primary teeth by itself is a seldom condition. An abnormal eruption pattern creates a clinical situation that is challenging to diagnose and treat. The clinical sequence of abnormal tooth eruption includes both syndromic and non-syndromic problems.\(^6,9\)

Various local factors like, mechanical obstruction from soft tissue overgrowth, supernumerary teeth, gingival fibromatoses, crowding, rotation of tooth buds, retained primary teeth and pathologic lesions are most common reasons behind impactions.\(^6,10\)

However, according to numerous reports and literature suggests various syndromes and metabolic conditions to be associated with multiple impacted permanent or supernumerary teeth. Few of them are cleidocranial dysostosis and Gardner’s syndrome, Down syndrome, Aarskog syndrome, Yunis–Varon syndrome, Zimmerman–Laband syndrome and Noonan’s syndrome along with hormonal disturbances such as hypothyroidism, hypopituitarism and hypoparathyroidism.\(^11\)
In the case of cleidocranial dysplasia, the patients usually present with high-arched palate (Figure 3b) with prolonged retention of the deciduous teeth, which leads to subsequent delay in eruption of the permanent teeth. Though both the intra-oral features were present in our case the abnormalities of skull and shoulder girdle were not seen. Gardner’s syndrome is also presents with multiple impacted teeth was ruled out as associated signs like multiple epidermoid cysts and other lesions were not present. Patients suffering from Gorlin–Sedano syndrome shows features like short hands, foot bones with short, straight collar bone along with multiple impacted teeth. In Yunis–Varon syndrome; there present agenesis or hypoplasia of clavicle, micrognathia, digital anomalies, hypodontia, spinal defects, and impacted teeth. Both the syndromes presented with multiple impacted teeth but were ruled out from our case as other symptoms were not present.

GAPO is the acronymic designation for a syndrome of growth retardation, alopecia, pseudo-anodontia (failure of tooth eruption) and optic atrophy. Patients who suffer from GAPO syndrome may have several abnormalities including broad anterior fontanel, bossed forehead, micrognathia, pouting lips and auricles, saddle nasal bridge, reduced perspiration.

Sickle cell anemia is a genetic disease. This anemia is due to a homozygous state of the abnormal hemoglobin-S. In the case of sickle cell anemia in dental finding mucosal pallor, delayed eruption, dental hypoplasia and radiographic changes are common oral findings associated with the disease.

In the cases of hormonal disorders, hypothyroidism, hypoparathyroidism, and pseudohypoparathyroidism are to be considered as a causative factor for disturbed normal physiology of eruption of teeth. T3, T4, and thyroid stimulating hormone serum estimation is needed for diagnosing of hypothyroidism. Total serum calcium concentration and serum parathyroid hormone estimation are needed for hypoparathyroidism and pseudohypoparathyroidism. There is a decrease of serum calcium in both hypoparathyroidism and pseudohypoparathyroidism. Elevated levels of parathyroid hormone can be seen in pseudohypoparathyroidism and decrease levels of parathyroid hormone in hypoparathyroidism can be seen. Metabolic disorders like vitamin-D deficiency rickets are also associated with impacted teeth. Failure of eruption can occur due to crowding of supernumerary teeth but in our case, that was not been observed.

The timing and sequence of emergence of teeth differ to some extent between populations of various geographic areas and even within the homogenous groups; environmental factors exert to some extent the rate of emergence of dentition.

Only a few cases of non-syndrome multiple impacted teeth were reported. In those studies, the predominant factor was, that some physical barrier led to impaction and non-eruption of teeth.

**CONCLUSION**

Multiple unerupted teeth without a known cause are a rare anomaly and need multi-disciplinary approach toward diagnosis and management of the patient. For the diagnosis, further investigation are needed to determine the requirement of expression (or inhibition) of putative eruption genes, as well as to identify where in the follicle they are expressed. In our cases, a definite diagnosis of disease or disorders or syndrome was not present. As financial status of the patient was poor hence investigation like gene mapping, was not been able to study. However, overall symptoms are suggestive of the syndrome, genetic or idiopathic etiology.

**REFERENCES**