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Non-Syndromic Oligodontia In A Child - A Rare Case

Case Report

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Abstract

Oligodontia(OD) is a rare numerical dental anomaly characterized by agenesis of six or more permanent teeth, excluding the third molars. It may show severe manifestation in relation to function and psychosocial wellbeing. Literature indicates children with OD are associated with taurodontism in one or more mandibular molars. To the best of our knowledge this is the first case of a non-syndromic OD in a child with taurodontism in both permanent maxillary and mandibular first molars. Due to the sporadicity of this anomaly, every individual case of OD should be reported to improve understanding of the condition.

Keywords: Oligodontia; Taurodontism; Retained Primary Tooth.

Introduction

Congenital absence of permanent teeth is the most common developmental anomaly in children [1, 2]. The prevalence of congenitally missing permanent teeth has been reported to vary from 2.2 to 9.7% in children of saudia arabia [1-3]. In literature, this has been classified according to the number of congenitally missing permanent teeth, excluding the third molars. Anodontia refers to a condition with complete absence of teeth. Oligodontia (OD) is the term given to the condition with six or more missing teeth. Patients with one to five missing teeth are categorized as having hypodontia [4].

OD is a rare condition with a reported prevalence of 0.084%.5 It has been studied as syndromic and non-syndromic forms, since this developmental disturbance was found in patients with defined congenital anomalies or syndromes [4, 6, 7]. The causative mechanism of oligodontia has been associated with multiple factors [8, 9]. It is proposed that complex interactions between environmental, genetic and epigenetic factors during odontogenesis play a role in the development of numerical dental anomalies [10]. The non-syndromic variant has been linked to mutations of genes PAX9, EDA, MSX1, AXIN2, EDARADD, NEMO, KRT17 and WNT10A [11-13].

This numerical dental anomaly is associated with anomalies of tooth size and form (microdontia, conical shaped teeth, taurodontism and dens invaginatus) [14-19] and disturbances in tooth eruption (delayed eruption of permanent teeth and retention of primary teeth) [16, 19-23]. The association of taurodontism in children with oligodontia has been reported in the literature. In a study, children with OD have shown 28.9% occurrence of taurodontism in one or more first mandibular molars.15 A few cases with taurodontism in mandibular molars in patients with OD have been reported [16, 17, 24]. However, to the best of our knowledge the association of OD with taurodontism in permanent maxillary molars has not been reported. Based on the available data, this developmental anomaly has shown to have severe manifestations relating to function and psychosocial well being [6, 25]. Moreover quality of life outcomes in children affected with OD appears to be adversely impacted in their daily life in terms of function and emotional well being [25-27]. To the best of our knowledge, this is the first reported case of a non-syndromic OD

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patient with taurodontism in both permanent maxillary and mandibular first molars.

Case Report

An eight year old boy of saudi descent, reported to the pediatric dental clinic for a routine dental check-up. Medical history and family history of the child were non-significant. Father revealed frequent incidents of child embarrassment because of being teased for dissimilar teeth appearance by children at school. Difficulty in chewing food was also observed by the parents. Patient had a normal built; no physical and developmental abnormalities were observed. Extraoral examination revealed a symmetrical face having a convex profile with no skeletal abnormalities. Intraoral assessment (Figure A-C) showed that the patient was in the first transitional phase of mixed dentition period. Permanent first molars were in early mesial shift relationship with bilateral spacing in the lateral incisor region of maxillary and mandibular arch. The permanent mandibular central incisors were shown to have rotation. Permanent maxillary left central incisor (#21) and primary maxillary left canine (#63) were having reverse overjet. Dental caries was present involving primary maxillary right central incisor(#51) and mandibular left first molar (#74). Calculus was evident over permanent mandibular central incisors. Radiographic evaluation revealed (Figure 4) multiple bilateral congenitally missing permanent teeth and an unilateral congenitally missing permanent mandibular left lateral incisor. (Table 1) Over-retained primary anterior teeth were observed (Table 1). Taurodontism was apparent in both right and left maxillary and mandibular permanent first molars. (Figure D) Based on the history, clinical and radiographic assessment, a diagnosis of nonsyndromic oligodontia was determined. The treatment plan advised to the patient was oral prophylaxis, pit and fissure sealants, restoration of decayed primary mandibular molar, orthodontic therapy and prosthetic rehabilitation. Although the child and the father were concerned about appearance and chewing difficulty, they hesitated to take treatment. Because of lack of cooperation, treatment was not carried out.

Discussion

Congenital absence of permanent teeth is the most prevalent developmental anomaly in children [1, 2]. OD is a rare numerical dental anomaly that refers to the agenesis of six or more teeth, excluding the third molars [4, 5]. It has multifactorial etiology, which involves environmental factors and genetic regulation [8, 9]. This anomaly has shown to have adverse impact on oral health related quality of life in the affected children [25-27]. In the present paper, we report a child patient who showed functional and psychosocial impact due to non-syndromic oligodontia characterized by nine congenitally missing permanent teeth in association with over retained primary teeth and taurodontism in all four permanent first molars.

The development of dentition is a complex process which is primarily determined by genetics [28]. Mutation in the genes PAX9, EDA, MSX1, AXIN2, EDARADD, NEMO, KRT17 and WNT10A have been associated with nonsyndromic OD [11-13].

Dental agenesis has been reported in association with taurodontism. The literature indicated that the association is more common in cases of severe dental agenesis or oligodontia [14, 29-32]. A few reports among children with OD having taurodontism in one or more first mandibular molars exist [15-17, 24]. The present case highlights the association of OD with taurodontism in both permanent maxillary and mandibular first molars.

The orthodontic implications of OD includes midline diastema, crossbite, intra-arch excess spacing, rotation, over retained primary tooth deflecting the erupting successor, midline deviation [33-35]. In the present case, all the above mentioned attributes were evident indicating the need of orthodontic correction. Moreover, the patient may later require prosthetic rehabilitation at an appropriate age.

Figure 1. a. Maxillary Arch, b. Mandibular Arch, c. Front View of teeth at occlusion, d. Orthopantomogram.

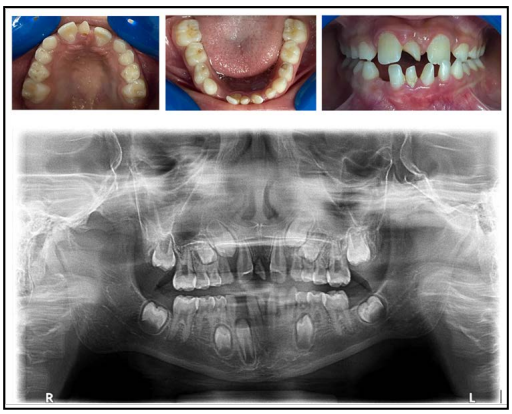

Table 1. Oligodontia and their associated features: Tooth numbers are according to the FDI* system of tooth numbering.

	Maxilla	Mandible
Congenitally Missing Permanent teeth	12, 15, 22, 25	32, 33, 35, 43, 45
Over retained Primary teeth	51	81
Teeth with Taurodontism	16, 26	36, 46

*Federation Dentaire Internationale

There are few case reports which show the association of OD with retained primary teeth [16, 36]. These teeth can maintain space and preserve bone and function in case of agenesis of successor. However they may delay or deflect the erupting permanent tooth if these primary teeth over retain. Hence clear understanding of the pathology, early diagnosis, timely intervention and appropriate treatment are necessary to manage the OD patients; thereby improving their oral health, esthetics and quality of life.

Conclusion

This is the first case to the best of our knowledge which reports a non-syndromic OD patient with taurodontism in both permanent maxillary and mandibular first molars. Due to the sporadicity of this anomaly, every individual case of OD should be reported to improve our understanding of the condition.

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