

Case Report

Single-rooted primary molar in a little girl with Ectodermal dysplasia: A rare caseArghavan Kamali¹, Zahra Alipuriani^{2*}¹Assistant Professor, Department of Pediatric Dentistry, Dental Research Center, Faculty of Dentistry, Hamadan University of Medical Sciences, Hamadan, Iran.²Resident of Pediatric Dentistry, Department of Prosthodontics, Dental Research Center, Faculty of Dentistry, Hamadan University of Medical Sciences, Hamadan, Iran.***Corresponding author**

Zahra Alipuriani

Email: z.alipuriani@gmail.com

Abstract: The aim of this study was to present a rare case of a single-rooted primary molar that is associated with syndromichypodontia. Dental anomalies, especially permanent single-rooted molars are fairly common between the second and third molar teeth; but the single-rooted primary molar is very rare and few cases have been reported. Information about the tooth root morphology and anatomical differences between single-rooted primary molars will enable a pediatric dentist to achieve a successful root canal treatment. Also, because there is relationship between a single-rooted primary molar and a specific gene, suspect can arise regarding the presence of the underlying disorder related to that gene after seeing single-rooted primary molar.

Keywords: syndromichypodontia, Dental anomalies, pediatric

INTRODUCTION

Dental development is affected by genetic and environmental factors [1] that in fact this process is a complex conversion between epithelial and mesenchymal cells and organize all odontogenesis stages, including initiation, morphogenesis, differentiation, which affect position, number and shape of the teeth [2]. There have been significant anatomical differences and various anomalies in human permanent molar teeth and many of these differences include shape and number of roots and root canal [3-6]. Single-rooted molars are frequently observed in the second and third permanent molars; but it has been reported rarely in primary molars [7-9]. Single-rooted molar is described as root fusion or a severe taurodontism and the roots are called pyramidal, cuneiform, tubular, cylindrical, prismatic, conical [10]. Today, the widespread belief is that the differences between the root forms are the results of the failure in Her twig epithelial root sheath (HERS) and this failure may occur in squeeze off stage; i.e. the exact time of roots separation.

Reduction in mesiodistal crown dimension has been reported in patients with hypodontia and one group of them is suffering from ectodermic dysplasia [11]. Ectodermal dysplasia heredity is a vast group of conditions, in which two or more anatomical structures

originating from the ectoderm are deficient. Patients are identified with hypoplasia or aplasia of structures such as the skin, nails, hair, teeth, nerve cells, sweat glands, parts of the ears, eyes and other organs. The number of teeth is particularly low. They often suffer from abnormal evolution. For example, they may have anterior, conical or pointed teeth molars declined in size are also observed [12].

CASE PRESENTATION

8-year-old girl was referred to the pediatric ward of Hamadan Dentistry School in Iran after parents complained of the lack of eruption of permanent teeth of their children. Very thin and sparse hair, sparse eyebrows, dry skin, protruding chin (Diagram1) were observed in extra-oral observation and reduced height of the lower third of the face and the relation between class 3 was confirmed based on the orthodontic specialist's diagnosis and lateral cephalometric radiograph. There was no permanent tooth in intraoral observation and incisor teeth between the two sides were also present.

And the patient had no history of smoking or exfoliation. Anterior teeth have an anatomic conical appearance with intra-dental diastema and posterior teeth had reduced mesio-distal dimension. Radiography analysis showed maxillary central incisor teeth missing

and missing of all permanent teeth except and and..... (Fig. 2)Deciduous maxillary first molar caries in both sides and the right incisor was detected.

First primary molars right anatomical appearance was of single-rooted type and patients complained of pain in these teeth. Based on clinical observations and radiographs of the patient, the diagnosis recommendation led to the anhidrotic ectodermal dysplasia.

The patient was later referred to a pediatrician and the anhidrotic ectodermal dysplasia was diagnosed after a skin biopsy. According to the periapical X-ray results, first primary molars and right maxillary incisor caries developed beyond the dentin and preferably we needed to carry out CBCT X-ray for the definitive diagnosis of single-channel single-root nature of the first molar, but parents were not inclined to do so. Therefore, after careful evaluation using radiographic and clinical evaluation, single-channel single-root nature of the first molar was diagnosed. Irreversible pulpitis was diagnosed in the treatment plan and pulp therapy for primary teeth and right incisor first molar. Endodontic treatment was obturated by metapex under rubber dam isolation after complete instrumentation, drying the channels with paper points. First primary molars of the both sides underwent plated stainless steel treatment and composite restoration treatment was carried out for the incisor (Figure 3 and Figure 4)



Fig-1:



Fig-2:



Fig-3



Fig-4:

DISCUSSION

The presence of single-rooted primary molar has been reported very rarely. The first case was reported by Ackerman for a 10-year-old child [11].When we see the single-rooted tooth in the permanent and deciduous teeth, there may be several modes: One root and a broad channel, two channels that may be connected or not or a C-shape channel. CT-Scan or in vitro analyses through teeth and histological sections should be applied in order to achieve better and more accurate diagnosis [13]. However, we realized single-channel tooth in two angles, because of the reluctance of parents to do CBCT in this case, through careful clinical evaluation and periapical x-ray [14].The role of genetics in the dental development and direct effect of genes and their signals on the evolution has been exactly proved and any mutation in them will lead to various malformations in teeth [15]. Hypodontia may be seen in an isolated form (non-syndromic) or syndromic, for example, ectodermal dysplasia.

One of the main reasons is lacks of proliferation of the tooth germination from the dental lamina [1]. The syndromic or non-syndromichypodontia should be distinguished after seeing any dental malformation based on clinical study of hair and nails and sweat glands [15].

In this case of the syndromic type was diagnosed. A small number of genes associated with root formation have been identified. Most genes

associated with tooth agenesis, MSX1 and PAX 9 have been identified [16]. Although root agenesis has been in recent research in mice lacking NFI-C gene [17], some studies reported autosomal dominant inheritance pattern and autosomal recessive inheritance pattern for the single-rooted primary molar [18, 19]. There are two broad categories for ectodermal dysplasia based on function and the number of sweat glands, including 1: Xlinked anhidrotic or hypohidrotic where there is no sweat glands or their number has been reduced (Chirst Siemens Tourain syndrome) and 2: hydrotic type of sweat glands was normal and is inherited in a autosomal dominant form(Clouston's syndrome) [20-22]. Teeth and hair are similarly involved in both of them, but the inheritance pattern of nails and sweat glands distinguish these two types [23].

In this case single-rooted molars diagnosis is very important clinically endodontically.

Special attention should be paid either in terms of access cavity preparation its outline and or its channel size and type. Diagnosis and treatment of the case of hypodontia has led is to further clinical and syndrome diagnosis studies. Perhaps, treating these teeth in these patients may affect the aesthetic and functional problems. On the other hand, the diagnosis and treatment of primary single-rooted molar due to its rarity is a great help in the treatment of these teeth.

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