

# REVIEWS

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## Hipodontia in Permanent Dentition in Patients with Unilateral Cleft Lip and Palate

### Braki zębowe w uzębieniu stałym u pacjentów z jednostronnym rozszczepem wargi i podniebienia

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#### Abstract

Hipodontia, also known as congenital lack of teeth or tooth agenesis, is the most common intraoral and dental anomaly. It occurs in ca. 10% of healthy people and is thought to inflict children with clefts six times more often. Occurrence of hipodontia depends on location and severity of the cleft. Unilateral complete clefts are thought to demonstrate the highest incidence of congenitally missing teeth of all the cleft groups. Mostly, in patients with cleft lip and palate, congenital lack of teeth refers to incisor region on the cleft side. A frequent lack of teeth in patients with clefts refer to premolars, usually maxillary ones. In bilateral clefts, lack of teeth usually refers to the left side of the dental arch. Even though hipodontia refers to upper lateral incisors and lower premolars mostly, it may refer to any group of teeth. Congenital lack of teeth in children with clefts needs appropriate treatment, which usually bases on orthodontic treatment followed by prosthetic reconstructions (**Dent. Med. Probl. 2009, 46, 3, 342–345**).

**Key words:** hipodontia, tooth agenesis, cleft lip and/or palate.

#### Streszczenie

Hipodoncja, nazywana także wrodzonym brakiem zębów lub agenezją zębową, jest najczęściej występującą wadą zębową i wewnątrzustną. Występuje u ok. 10% zdrowych ludzi. Uważa się, że u dzieci z rozszczepem występuje sześciokrotnie częściej. Występowanie hipodoncji zależy od umiejscowienia i nasilenia rozszczepu. Jednostronne rozszczepy całkowite są uważane za wadę, w której wrodzone braki zębowe występują najczęściej. Wśród pacjentów z rozszczepami braki zębowe występują głównie po stronie rozszczepu, w okolicy zębów siecznych. Częstym brakiem zębowym jest także brak zębów przedtrzonowych, dotyczący w większości przypadków szczęki. W rozszczepach obustronnych braki zębowe dotyczą głównie lewej strony łuków zębowych. Mimo że hipodoncja najczęściej dotyczy zębów siecznych bocznych górnych i dolnych przedtrzonowych, może dotyczyć każdej grupy zębów. Wrodzone braki zębowe u pacjentów z rozszczepem wymagają odpowiedniego leczenia, które zazwyczaj polega na leczeniu ortodontycznym, które poprzedza rekonstrukcję protetyczną uzębienia (**Dent. Med. Probl. 2009, 46, 3, 342–345**).

**Słowa kluczowe:** braki zębowe, hipodoncja, rozszczep wargi i/lub podniebienia.

Dental anomalies may be divided to anomalies in number of teeth (hiperdontia, hipodontia), variations in teeth structure and displacement of them in the tooth arch. They are thought to inflict ca. 18% of all the people. Among them, hipodontia is thought to be the most common intraoral and dental anomaly that occurs in ca. 10% of the society and its frequency is becoming higher than it was at the beginning of XXth century. Hipodontia, also known as congenital lack of teeth or tooth agenesis, is a condition of missing teeth, corresponding

to one or more of the teeth germs, excluding third molars [1–7]. Lack of tooth usually refers to second lower premolar (ca. 56%) or lateral upper incisor (ca. 46%). Dental anomalies may be the reason for malocclusion, an inappropriate arch forming and furthermore temporomandibular joint dysfunction [2]. There are many theses that explain reasons for tooth agenesis, among them the theory of phylogenic underdevelopment of dentition, development disorders of ectoderm, influence of such factors as: hormones, malnutri-

tion and environmental factors, including traumas of fetus. The influence of genetic factor is indisputable, which may be confirmed by family occurrence of hipodontia and increase its frequency in children with genetically contingent development disorders such as ectodermal dysplasia, Down Syndrome, Wolf-Hirschhorn Syndrome or cleft lip and/or palate [2]. Mutations of genes *MSX1* and *MSX2* may lead to disorders in tooth development, among them: tooth agenesis (absence of either all or some of tooth germs), disorders in shape or size of tooth crown or root [8]. Tooth agenesis inflicts girls more often and it is usually correlated with other dental anomalies such as structural variations, late eruption and tooth malposition [9].

It is the most common anomaly not only in healthy individuals, but also in children with clefts is hipodontia [2, 5–7]. There is a thesis that dental anomalies may occur more often in children with cleft lip and palate than in healthy children. Among them, hipodontia is thought to be six times more frequent in children with clefts [4–7]. It occurs in 9.3% to 68.4% of the patients with cleft, though some authors claim that hipodontia appears in 80% of patients with clefts [5, 10, 11]. Prevalence of hipodontia depends on location and severity of the cleft [10]. Most dental anomalies in patients with cleft lip and palate occur in lateral incisor region on the cleft side [4–7].

The influence of genetic and environmental factors in cleft lip and palate development is indisputable [12, 13]. Approximately, mutations of 30 genes in humans may lead to cleft lip and palate as a major or an associated malformation. The incidence of clefts is not dominated by one gene, but has its polygenic inheritance character [12]. Some genes such as *MSX1* and *AXIN2* affect early tooth development and are associated with tooth agenesis. Besides, they are also responsible for dentofacial development and its mutations may lead to facial deformations and anomalies such as cleft lip and palate [14]. *MSX1*-gene is the most common and well known gene responsible for development of facial region and fusion of medial and nasal processes with maxillary process. Theoretically, both clefts and tooth agenesis have their genetic background in *MSX1* – gene, with its expression in mesenchyme [15]. Mutation of the gene disturbs fusion of medial and nasal processes with maxillary process and, in consequence, makes it impossible to form the primary palate, as well as disturbs fusion of the secondary palatal shelves [12].

Unilateral complete clefts are thought to demonstrate the highest incidence of congenitally missing teeth of all the cleft groups [16]. In over 20% of patients, some teeth are missing, while, as in unilateral clefts, hipodontia rages at 19% [17].

At the unilateral clefts, the most commonly missing teeth are: lateral upper incisor and second premolars at the cleft side. Missing premolars may refer both to maxilla and mandible. At the non-cleft part, hipodontia usually refers to maxillary second premolars and lateral incisors [15].

There is also a thesis that the most commonly missing teeth is a maxillary lateral incisor, particularly on the cleft side of the dental arch [15, 16]. They are thought to lack in ca. 48% of patients with unilateral cleft [18]. More rarely, other missing teeth occur. Second premolars are hypodontic in ca. 21% and two times more often refer to maxilla [15]. A rare congenital lack of teeth in patients with unilateral cleft of lip and palate refers to upper central incisors [19].

In bilateral clefts, lack of teeth usually refers to the left side of the dental arch, where it occurs 33% more often than on the right. The most frequent thesis claims that missing teeth occur more often on the cleft side [15]. There is a theory that in unilateral clefts, missing teeth usually occur on the right side of the dental arch (47.6% more often), even though the incidence of clefts on the left side is ca. 27% higher in comparison to the unilateral cleft of the right side [16].

The rarest congenital lack of teeth in patients with cleft is thought to be a missing upper central incisor. Aizenbud et al. observed it in 15 out of 120 patients with clefts [20].

Besides hipodontia, also other dental anomalies are more frequent in children with clefts. There are: malposition of teeth, rotations, hipodontia microdontia, incorrect tooth shape inclinations and retention of a tooth, mostly canine [5, 17, 18, 21]. Mostly, those refer to the frontal part of upper arch. Also, the dental development in children with cleft lip and palate is delayed on the side of the cleft [5, 17]. Dental anomalies outside the cleft area are presented by over 32% of patients with complete cleft lip and palate, which mostly refer to lower premolars [22]. Also asymmetry of tooth pairs in children with clefts occur 5 times more frequently than in non-cleft individuals [23].

It has also been shown that healthy siblings of patients with clefts are two times more often inflicted with similar dental anomalies (with hipodontia among them) than non-related individuals, which also proves the influence of genetic factor in tooth development [24, 25].

The importance of taking care of children with cleft lip and palate lies on orthodontic treatment. Missing teeth and malocclusion are responsible for abnormal skeletal relationships, soft-tissue condition, presence of afunctional teeth and malfunction of some of the teeth [16].

## Treatment

A very often method of treatment of lack of teeth in patients with clefts is closure of the free space. This usually refers to premolars, but may also be useful when there is a possibility of closure of gap when a lateral incisor is missing. The correct cuspidation is required when canine stands in the place of lateral incisor. It is possible to make a prosthetic crown or change shape of canine with selective grinding, so that it could imitate incisor, though it is not obligatory [26]. In patients with hipodontia of teeth, usually referring to frontal area, also some prosthetic appliances are used. Those may be crowns, Schwarz plates with artificial teeth (usually used as temporary denture), adhesive bridges and bridges. This treatment is a continuation of orthodontic treatment and is correlated with stage of retention. Sometimes, treatment requires endodontic treatment of canine next

to cleft fissure. Orthodontic treatment concentrates on transverse extension of upper arch, steering development of frontal region with face mask and correction of malocclusion [19, 27, 28].

Moreover, there is a theory that early bone grafts, performed at the age of one, influence the severity and frequency of dental anomalies in cleft region. The studies showed that children with early bone grafts show lower incidence of hipodontia, hiperdontia and incisors deformities than children with late bone grafts or without any. Also bone grafts performed after the second permanent incisor is erupted reveal the highest number of correctly developed lateral incisors. Children with bone grafts performed after the eruption of permanent canines showed the highest rate of missing teeth outside the cleft area [29]. This shows how important it is to predict more and more malformations in this patients to form and cure patients with clefts at the appropriate age.

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