

Clinical Characteristics of Determining the Connective Tissue Dysplasia Severity in Children and Adolescents Depending on Changes in the Height of the Hard Palate Vault

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ABSTRACT

Despite a wide study of connective tissue dysplasia (CTD), this problem remains unresolved, since this hereditary collagenopathy has a wide range of common and local phenotypic manifestations. The deep (Gothic) palate is most often diagnosed in the structure of CTD local manifestations, which is accompanied by narrowing of upper dentition, bite abnormalities, impaired functional activity of denture system, respiration, etc. A comprehensive craniometric study of 869 children and adolescents from 7 to 15 years old with CTD was conducted. We developed a device and method for measuring the height of hard palate vault in children and adolescents with CTD based on clinical-epidemiological results. The height of the hard palate vault is up to 18.2 ± 0.02 mm in children with mild CTD, while in patients with a moderate to severe degree of congenital dysgenesis, pronounced biometric changes in the height of the hard palate vault (19.3 ± 0.04 to 32.4 ± 0.02 mm). The relationship between CTD severity & upper dentition deformation has been revealed during our research for the first time, as well as the trend of dynamic growth of hard palate height indicators depending on CTD severity has been established for the first time.

Keywords: connective tissue dysplasia, CTD, hard palate vault, upper dentition deformation

1. INTRODUCTION

Nowadays problems of connective tissue dysplasia (CTD) are widely studied, it has been proved that its basis is hereditary collagenopathy [2, 8, 17]. It should be noted that morphological and functional peculiarity of organs and systems largely depends on "maturity" of connective tissue [30, 32]. Morphohistological features of connective tissue are universality, plasticity, polymorphism of cellular systems, which determine functionality and high level of adaptation when exposed to aggressive factors of external and internal environment [7, 31].

CTD is known to have a negative effect on the functional state of organs and tissues of the oral cavity,

as well as the maxillofacial region, because the organs and systems of the body in structural morphological components are of connective tissue origin [1, 3–6, 9–16]. The most frequently detected phenotypic signs of CTD are high Gothic palate (44.4 %), dental anomalies (64.8 %), occlusions (35.4 %), vestibular inclination of the anterior teeth of the upper jaw, deep overbite, distal occlusion (71 %), dystopia and crowding of the teeth (55.6 %), deflected septum (16.7 %) [18–29].

All mentioned above determines the relevance of our research aimed at studying the problems of CTD, which are of important theoretical, scientific and practical importance for improving the treatment and preventive care and rehabilitation of patients.

The purpose of the research is to determine the severity of connective tissue dysplasia based on anatomical and topographic height changes of the hard palate vault in children and adolescents.

2. MATERIAL AND RESEARCH METHODS

We have conducted a comprehensive clinical and craniometric survey of 869 children and adolescents from 7 to 15 years old with CTD. The survey was carried out in secondary schools № 31 and № 38 (Yakutsk), "Rehabilitation Neurological Children's Center" (Yakutsk), as well as in Allaikhovsky, Kobayasky and Churapchinsky uluses (districts) of the Republic of Sakha (Yakutia). The CTD degree was determined by T. Milkovska-Dimitrova and A. Karkashov's method (1985). The contingent of children is formed by random sampling.

Morphometric studies of teeth and jaws were performed with plaster diagnostic models. The relationship between the sizes of the permanent incisors of the upper and lower jaws was determined by Tone-index (1937). The study of the correspondence between the size of the teeth and the width of the dentitions was carried out by using the simple and sufficiently informative Pons method (1907). The palate height was determined by the value of the perpendicular from the most convex point on the drawn contour of the palate to the line connecting the vertices of the interdental papillae between the second premolars and the first molars.

The CTD degree was determined by T. Milkovska-Dimitrova and A. Karkashova's method (1985): a mild (I degree) is diagnosed in the presence of two main signs; medium (II degree) – at 3 main and 2–3 secondary or 3–4 main and 1–2 secondary signs; severe (III degree) – in the presence of 5 main and 3 secondary signs. The degree of CTD severity in each particular child was calculated by the sum of points. The sum of the points should not exceed 12 for the first degree of CTD severity (norm), for moderate – 23, for a severe degree – 24 or more points.

We have developed the device for measuring the height of the hard palate vault based on the obtained data and in order to improve the comprehensive treatment, rehabilitation and prevention of the manifestations of CTD in the organs and tissues of the oral cavity, as well as in the maxillofacial region (patent № 177476 dated from 26.02.18). The device consists of a handle with a vertical ruler up to 3-4 cm long with millimeter scale. A handle and a bar of ruler are made of metal. A bar is frictionally mounted on the bar of the ruler, which serves as a control support during measurements and is a thin and rigid metal plate. There is a through hole with a diameter on the bar in the center

providing frictional connection with the bar of the ruler. With a small force, the bar can move on a vertical plane along the rod and rotate around its axis along a horizontal plane. Friction connection is provided by fitting the bar on the rod with some tension or by means of special seals between the parts. All parts of the device have a streamlined shape and do not have sharp angles in order to avoid possible injury to the mucous membrane of the hard palate during measurement work. The upper dentition was measured by Pons method (dentition width) and Tone-index (sum of mesiodistal dimensions of the central incisors of the upper jaw). Also, we have developed the method for determining the height of the hard palate vault in children with connective tissue dysplasia (Patent № 2672369 dated from 14.11.2018). The main clinical objective of this method is to establish and confirm the presence of CTD with the determination of its severity using a device for measuring the height of hard palate vault in children and adolescents.

We carried out the analysis of the measurement data and assessment of the height of the hard palate vault by using the device we have developed (Patent № 177476 dated 26.02.2018) and method for determining the height of the hard palate vault in children with connective tissue dysplasia (Patent №. 2672369 dated 14.11.2018) for the following applications: 1) in the oral cavity – at outpatient and polyclinic reception directly in the dental chair. 2) outside the oral cavity – an impression was taken from the upper jaw with the obligatory obtaining of a clear print of the hard palate and dentition, according to which a control and diagnostic model of the upper jaw was made, then measurements of the width of the upper dentition were made according to Pons and Tone-indices (the sum of the width of the four upper incisors) using the developed device.

Observations were made in accordance with the ethical principles of scientific medical research with human participation, as defined by the Helsinki Declaration of the World Medical Association (1964, 2000), and the requirements set forth in the main regulatory documents of the Russian Federation for clinical research, as well as approved by the Ethical Committee of the Federal State Educational Institution "M.K. Ammosov North-Eastern Federal University". All children surveyed had their parents' prior voluntary consent.

Statistical processing of the research data was carried out by using standard methods of variation statistics with the calculation of the average, mean square error using the Microsoft Excel application software packages, 2003 (Microsoft Corporation). The results were grouped according to a combination of the same traits. Factorial and correlation analysis of quantitative material with determination of Pearson

coefficient (r) was carried out using the average program package "SPSS," version 22.

3. RESULTS AND DISCUSSION

Children and adolescents had signs of local ectodermal dysplastic manifestations in the form of the Gothic palate in the structure of manifestations of connective tissue dysplasia in organs and tissues of the oral cavity, there were $35.78 \pm 0.51 \%$. In addition, the dysfunctions of the temporomandibular joint ($63.36 \pm 0.19 \%$) were most often determined in their structure, then there were bite abnormalities – $31.81 \pm 0.36 \%$ in frequency, then a deep bite ($32.67 \pm 0.35 \%$) and a straight, cross, open, distal and mesial bite ($67.33 \pm 0.17 \%$). The narrowing and deformation of the dentitions were within the digital values of $25.01 \pm 0.39 \%$. It should be noted that the research results indicated the presence of some features of biometric changes in the hard palate in children with different degrees of CTD severity. So, the moderate degree ($55.12 \pm 1.05 \%$) was most often detected, followed by mild ($32.05 \pm 1.59 \%$) and less often severe degree ($12.83 \pm 2.04 \%$). At the same time, all examined children (100 %) with CTD showed high palate and narrowing of the upper jaw. Based on the study of phenotypic traits manifested in oral organs and tissues in children and adolescents with CTD, it gave us the rationale to develop a device for measuring the vault height of the hard palate. The device is used at dental consultation in outpatient-polyclinic conditions to ensure accuracy of biometric measurements of hard palate vault height in oral cavity. Pearson's correlation analysis revealed a relationship between CTD and the Gothic palate ($r = 0.49$), which confirmed the validity of the development of this device.

At the same time, a method of determining the height of a hard palate vault in children and adolescents using a device for measuring the height of a hard palate vault has been developed. The main objective of this method is to establish parameters of morphological deformations of upper dentition width and hard palate vault height in children and adolescents with different degree of connective tissue dysplasia. We measured the height of the hard palate vault by using our device according to successive technological stages. At the clinical stage, a sterile device for measuring the height of the hard palate vault is used to conduct biometric studies in a dental room. First, device is introduced into oral cavity with preliminary extension of measuring ruler to minimum level. After that, holding the device by the handle, the support control bar is fixed between the second premolars and the first molars of the upper jaw and the measuring ruler is advanced until stop towards the hard palate vault. Further, the deepest point of the hard palate vault is selected and the height of the hard palate vault is fixed on the ruler scale. In this case, the data on the ruler are taken from the outside of the bar.

The upper jaw impression is taken with the obligatory obtaining of a clear hard palate and dentition imprint, according to which a control and diagnostic model of the upper jaw is made in case of using gypsum diagnostic models. Then height of hard palate arch is measured with the help of the developed device. For this purpose, between the second premolars and the first molars of the upper jaw, a reference bar is applied to create a horizontal reference point and the measuring ruler is advanced to the stop (to the deepest [high] point of the hard palate vault).

Analysis of the results indicates some features in the height of the hard palate (Table 1). The sum of the width of the four upper incisors in the examined age groups of children and adolescents with different degrees of severity of CTD ranged from 34.03 ± 0.05 to 40.03 ± 0.41 mm ($p < 0.05$). The obtained data, interpreted as high indicators, indicate macrodentia of the upper incisors in the examined groups, which is a specific regional feature. This factor can be considered as one of the phenotypic signs manifested in the cavity in children and adolescents with CTD living in the North. It is important to emphasize that this manifestation of CTD was discovered for the first time.

Table 1. Indicators of hard palate deformation in children and adolescents with various degrees of connective tissue dysplasia

Morphological Parameters	CTD degree of manifestation		
	mild (up to 12 points)	moderate (up to 23 points)	severe (over 24 points)
Sum of width of four upper incisors (mm)	$34,03 \pm 0,05$	$36,04 \pm 0,06^*$	$40,03 \pm 0,41^{**}$
Upper dentition narrowing by Pons-premolar index	$41,42 \pm 0,02$	$34,71 \pm 0,04^*$	$33,23 \pm 0,03^{**}$
Pons index – molar index	$46,41 \pm 0,03$	$46,73 \pm 0,04$	$36,92 \pm 0,03^{**}$
The vault height of the hard palate (mm)	up to $18,23 \pm 0,02$	from $19,31 \pm 0,04$ to $27,03 \pm 0,03^*$	from $28,01 \pm 0,03$ to $32,43 \pm 0,02^{**}$

Note: P * – validity of CTD differences of mild to moderate severity; P ** – validity of CTD differences of moderate and severe severity.

Using Pons index (premolar index), the upper dentition narrowing was measured depending on CTD severity. So, the mild degree has the indicator of 41.42 ± 0.02 mm, with a heavy degree reaches 33.23 ± 0.03 mm ($p < 0.05$). According to the upper

dentition measurement according to Pons index (molar index), the narrowing indicators ranged from 46.41 ± 0.03 to 36.92 ± 0.03 mm ($p < 0.05$). The obtained data indicate a pronounced narrowing of the upper dentition, which means the presence of dentate abnormalities in the examined patients.

It is important to note that in children with mild CTD, the height of the hard palate vault was up to 18.2 ± 0.02 mm. While in patients with a moderate to severe degree of congenital dysgenesis, pronounced biometric changes in the height of the vault of the hard palate (19.3 ± 0.04 to 32.4 ± 0.02 mm) were determined. According to these facts, we came to the conclusion that we have revealed a tendency of dynamic growth of hard palate heights depending on the severity of CTD for the first time in the course of comprehensive studies.

The carried-out correlation analysis by Pearson revealed existence of interrelation between the Gothic palate and diseases of nervous system ($r=0.44$), a keeled thorax ($r=0.58$), scoliosis ($r=0.95$), hyperextensibility of skin ($r=0.81$), disturbed occlusion ($r=0.44$), anomalies of position of teeth ($r=0.49$), narrowing and deformation of tooth alignments ($r=0.79$), TMJD dysfunction ($r=0.73$), pH ($r=0.78$), viscosity ($r=0.75$). It has been proven that the relationship between CTD and the Gothic palate affects the functional state of organs and tissues of the body.

4. CONCLUSION

We have revealed the law of relationship of CTD severity and deformation of upper dentition for the first time as a result of complex analysis of phenotypic signs manifesting in organs and tissues of oral cavity in children and adolescents with CTD. For the first time, the established trend of dynamic growth of hard palate heights depending on CTD severity has theoretical, practical and scientific significance, it must be taken into account when conducting comprehensive therapeutic and rehabilitation treatment in children with this congenital pathology.

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