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Диагностика скрытых расщелин нёба оториноларингологом

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АННОТАЦИЯ

Обоснование. Скрытая, или подслизистая (submucosae), расщелина нёба представляет собой редкую форму изолированных расщелин, которая характеризуется поражением речевоспроизводящих структур артикуляционного аппарата при неповрежденной слизистой оболочке нёба. Пациенты со скрытой расщелиной нёба нуждаются в особом внимании оториноларинголога, так как данный анатомический порок развития приводит к поражению среднего уха и существенно влияет на слух и качество жизни пациентов.

Цель — определить дополнительные критерии диагностики скрытой расщелины нёба и идентифицировать РКТ-маркеры скрытой расщелины нёба, оценить влияние ее на функцию среднего уха.

Материалы и методы. Проанализировано 16 пациентов со скрытой расщелиной нёба, проходивших обследование и лечение в ГАУЗ ДРКБ МЗ РТ.

Результаты. Медиана возраста установления диагноза составила 6,5 года (от 3 до 13 лет), 62,5 % случаев впервые заподозрены ЛОР-врачом. У 14 пациентов (87,5 %) при первичном осмотре уже отмечалось значительное снижение слуха, в 21,9 % случаев выявлен экссудативный средний отит в мукозной стадии, в 21,9 % случаев — адгезивный средний отит, в 18,8 % случаев — хронический отит с холестеатомой. У 56,3 % пациентов (n = 9) в анамнезе наблюдались частые гнойные средние отиты, у 87,5 % (n = 14) — частые риносинуситы. Не парциальная аденотомия, которая усугубила ринолалию, проведена по месту жительства 5 пациентам (31,3 %). У 62,5 % (n = 10) скрытая расщелина нёба компенсирована и ринолалии не наблюдалось. Выявлено три РКТ-маркера скрытой расщелины нёба: клиновидный дефект в 3D-реконструкции черепа, дефект нёбной кости и укороченный сошник в коронарной проекции, смещение задней носовой ости в сагиттальной проекции.

Заключение. Случаи скрытой расщелины нёба демонстрируют необходимость знания данной патологии оториноларингологами и педиатрами, чтобы вовремя проводить правильную комплексную реабилитацию пациентов.

Ключевые слова: скрытая расщелина нёба; экссудативный средний отит; тугоухость; холестеатома.

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Diagnosis of submucosal cleft palate by an otorhinolaryngologist

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ABSTRACT

BACKGROUND: A hidden or submucosal cleft palate is a rare form of isolated clefts characterized by damage to the speech-producing structures of the articulatory apparatus with intact mucous membrane of the palate. Patients with submucosal cleft palate require special attention from an otorhinolaryngologist because this anatomical malformation leads to middle ear damage and significantly affects the hearing and quality of life of patients.

AIM: To determine additional diagnostic criteria for submucosal cleft palate and identify CT markers of submucosal cleft palate, to assess its impact on the function of the middle ear.

MATERIALS AND METHODS: The study included 16 patients with submucosal cleft palate who underwent examination and treatment at the Children's Republican Clinical Hospital of the Ministry of Health of the Republic of Tatarstan.

RESULTS: Their median age at diagnosis was 6.5 (range, 3–13) years, and 62.5% of the cases were first suspected by an ear–nose–throat specialist. During the initial examination, a significant decrease in hearing was already noted in 14 (87.5%) patients, otitis media with effusion in the mucosal stage in 21.9%, adhesive otitis media in 21.9%, and chronic otitis with cholesteatoma in 18.8%. Frequent purulent otitis media were observed in the anamnesis in 56.3% of patients (n = 9), and frequent rhinosinusitis in 87.5% (n = 14). Non-partial adenotomy was performed at the place of residence in 5 patients (31.3%), which aggravated rhinolalia. In 62.5% (n = 10) submucosal cleft palate was compensated and rhinolalia was not observed. Three CT markers of submucosal cleft palate were revealed: a wedge-shaped defect in 3D skull reconstruction, a defect of the palatine bone and a shortened vomer in the coronal projection, and displacement of the posterior nasal spine in the sagittal projection.

CONCLUSIONS: Cases of submucosal cleft palate demonstrate the need for ear–nose–throat specialist and pediatricians to know this pathology in order to conduct timely and correct comprehensive rehabilitation of patients.

Keywords: submucosal cleft palate; otitis media with effusion; hearing loss; cholesteatoma.

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BACKGROUND

Cleft lip and palate are prevalent congenital defects that commonly impact the orofacial region. A multidisciplinary approach that incorporates the expertise of diverse specialists, including maxillofacial surgeons, otorhinolaryngologists, speech therapists, geneticists, orthodontists, pediatricians, and others, is essential for the efficacy of rehabilitative treatment for congenital cleft palate patients [1, 2].

Globally, around 250,000 infants are born each year with orofacial clefts. Over the past four decades, the number of orofacial cleft patients has increased twofold [3]. As per N.A. Kasimovskaya et al. (2020), the prevalence of orofacial cleft in different countries varies from 1 case per 673 newborns to 1 case per 1000–1200 newborns, depending on the population, geographical characteristics, and the level of population development of the countries [4–6].

Occult or submucosal cleft palate is a rare type of isolated cleft characterized by a nasal voice with an intact palate. Incomplete midline clefts of the soft and partially hard palate comprise the majority of clefts; however, unilateral forms also occur. A comprehensive examination reveals a triad of signs: a triangular bony defect along the midline of the hard palate, a uvular cleft of varying degrees, and an insufficiency of the muscles of the soft palate in closure with intact mucosa in the form of an indentation on the palate and uvula, indicating an absence of bilateral muscular connection [7]. The primary cause of speech impairment in occult cleft palate patients can be attributed to anatomical and functional deficiencies linked to damage to the speech-producing structures of the articulatory apparatus. Literature reviews, particularly of foreign and Russian literature, reveal a paucity of information regarding the diagnosis, treatment, and rehabilitation of occult cleft palate patients. The available data is primarily composed of a series of isolated observations that are presented in the context of dissertations or descriptions of syndromes [1]. A.V. Bogoroditskaya et al. examined nine patients with an average age of five years at the initial diagnosis of an occult cleft palate. The patients exhibited a history of rhinolalia (67%) and recurrent otitis media (78%) [8]. The authors present a comprehensive overview of the endoscopic diagnosis, which encompassed diaphanoscopy, as well as conservative and surgical treatment methods — including adenotomy, adenotonsillotomy, and tympanic cavity shunt.

The diagnosis of occult cleft palate with known clinical symptoms is challenging for physicians from other specialties due to a lack of knowledge about this pathology and the limitations of modern instrumental and radiological diagnostic methods. The severe health consequences of delayed diagnosis exemplify the "hidden" nature of this form of cleft palate.

The Eustachian tube constitutes a component of the functional middle ear system. Its functionality is influenced by all forms of congenital clefts, including the cleft palate. Many researchers confirm that the *m. tensor veli palatini* plays a primary role in the opening and closing of the Eustachian tube. The protective, drainage, and ventilation functions of the Eustachian tube are jointly performed by the *m. tensor tympani* and *m. tensor veli palatini*, which are both innervated by the *n. mandibularis*. The pharyngeal muscles contract during swallowing, thereby closing the pharyngeal orifices of the Eustachian tube and simultaneously dilating the lumen in the upper cartilaginous part, specifically "Rüdinger's safety canal." The pressure difference between the Eustachian tube and the tympanic cavity results in a peculiar valve mechanism [7, 9]. In a cleft palate, the aponeurosis of the *m. tensor veli palatini* attaches along the bone's edges rather than integrating into the palatine aponeurosis. This disrupts the valve opening mechanism of the Eustachian tube and contributes to the formation of exudate in the middle ear [10, 11].

In experimental studies, B. Gyanwali et al. (2016) and D.S. Heidsieck et al. (2016) demonstrated the predominant role of *m. tensor veli palatini* in the development of exudative otitis media (EOM) [12, 13]. Additional factors contributing to EOM occurrence in cleft palate patients included Eustachian tube underdevelopment, alterations in its length, anatomical features of the craniofacial skeleton, palatine-pharyngeal insufficiency and its consequences, the swallowing mechanism, dysbiotic phenomena in the nasopharyngeal mucosa and the auditory tube opening, nasopharyngeal space enlargement, and medial wing plate changes. Other factors include the wing process, increased cartilage stiffness, and an increased angle between the cartilage and the *m. tensor veli palatini*. Moreover, cleft palate patients exhibit a reduced ratio of the lateral and medial laminae in the cartilage, a small curvature of the lumen of the cartilaginous part of the auditory tube, a decrease in elastin in the cartilage, and a smaller surface of attachment of the *m. tensor veli palatini* to the cartilage [14–16].

Cleft palate, as per numerous researchers, elevates the risk of developing both EOM and chronic purulent otitis media, including cholesteatoma. B.D. Djurhuus et al. (2015), for instance, concluded that the risk of cholesteatoma incidence was 20-fold higher for isolated cleft palate and 15-fold higher for penetrating (complete) clefts than in the general population without clefts after analyzing the nationwide study results from 1936 to 2009 that included 8593 individuals with non-syndromic cleft palate [17].

In a 25-year retrospective cohort study, Canadian researchers discovered that acquired cholesteatoma was three times more common in patients with a complete cleft palate than in those with an isolated cleft palate [18].

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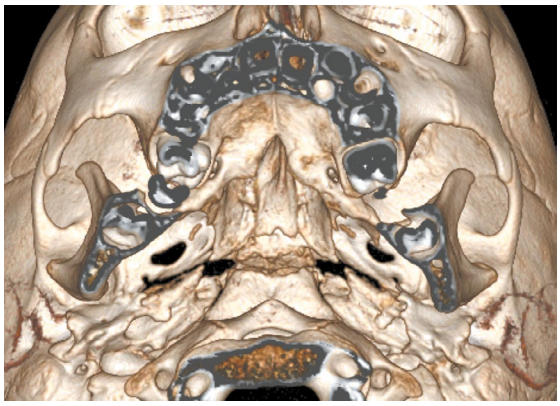

Fig. 1. Wedge-shaped defect of the hard palate on a three-dimensional computed tomography reconstructed image of the skull

Рис. 1. Клиновидный дефект твердого нёба при 3D-реконструкции черепа на рентгеновской компьютерной томограмме

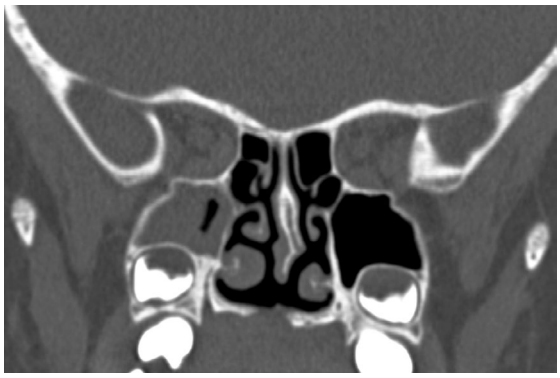

Fig. 2. Defect of the hard palate and shortening of the vomer in the coronal projection on computed tomography

Рис. 2. Дефект твердого нёба и укорочение сошника в коронарной проекции на рентгеновской компьютерной томограмме

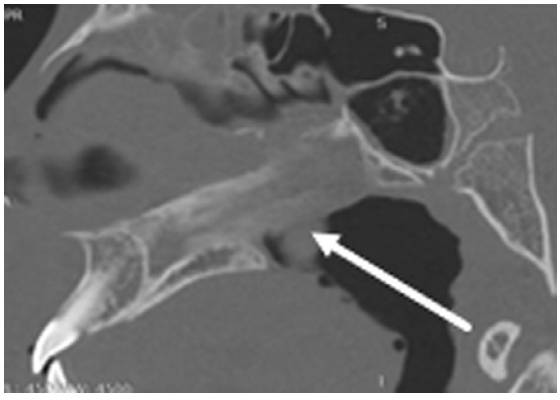

Fig. 3. Displacement of the posterior nasal spine and posterior ends of the inferior nasal conchae protruding into the lumen of the nasopharynx in the sagittal projection on RCT

Рис. 3. Смещение задней носовой ости и выступающие в просвет носоглотки задние концы нижних носовых раковин в сагиттальной проекции на рентгеновской компьютерной томограмме

The study aimed to define additional criteria for diagnosing occult cleft palate, identify occult cleft palate markers on computed tomography for otolaryngologists, and evaluate its influence on middle ear function.

MATERIALS AND METHODS

The study comprised 16 patients with occult cleft palate who were evaluated and treated in the departments of otolaryngology and maxillofacial surgery of the Children's Republican Clinical Hospital of the Ministry of Health of the Republic of Tatarstan. In 15 out of 16 patients, the diagnosis of occult cleft palate was verified through X-ray computed tomography (CT). The patients were monitored for 1 to 4 years.

RESULTS

The median age of the study participants at the time of cleft palate diagnosis was 6.5 years (ranging from 3 to 13 years), of which ten patients (62.5%) were first diagnosed with cleft palate by an otolaryngologist. A right-sided cleft palate with asymmetry of the soft palate muscles and EOM on the cleft side was identified in one patient (6.3%), while 15 patients (93.8%) exhibited isolated cleft palates. The study involved ten male (62.5%) and six female (37.5%) patients.

Following a fibrorhinoscopy, the otorhinolaryngologist observed nasopharyngeal space enlargement, shortening of the vomer, and a slight retraction of the mucous membrane on the soft palate along the midline. Additionally, the posterior ends of the lower nasal shell of the left half were visible when examining the right half of the nose.

We identified characteristic markers of occult cleft palate by analyzing the X-ray and CT data [19]. In patients without a history of palatal surgery, a palatal defect of varying severity was observed on the palatine bone in the coronal projection. The wedge-shaped defect was most evident in the 3D skull reconstruction (Fig. 1). The second feature observed on the CT scans was a shortened vomer, which is typically responsible for forming the choanas, dividing the nasal cavity into two halves, and connecting to the posterior nasal apex (Fig. 2). However, shortening of the vomer should not be confused with its hypoplasia and aplasia.

The medial sagittal projection unequivocally demonstrates the third radiologic marker of occult cleft palate. This marker involves the anterior displacement of the posterior nasal tip, which visibly enlarges the nasopharynx in the anteroposterior direction on CT. In some patients, the posterior portions of the inferior nasal shells protrude into the nasopharynx (Fig. 3).

One patient (6.3%) exhibited a developmental anomaly of the external and middle ear on the right side, classified as grade III microtia. This anomaly was accompanied by grade III

conductive hearing loss in the right ear and congenital paresis of the palate. The occurrence of these anomalies on the right side led to EOM occurrence on the left, well-hearing ear, and a pronounced asymmetry of the palate, as observed during mesopharyngoscopy. Furthermore, the diagnosis was verified by the identification of a triangular-shaped indentation on the soft palate.

The familial nature of sensorineural hearing loss (pathology in close relatives) was observed in one patient (6.3%). This evidently represents a prognostically unfavorable factor for the patient's hearing during surgical treatment for chronic purulent otitis media with cholesteatoma.

A total of 81.3% of patients (n = 13) experienced speech difficulties during their early childhood and underwent prolonged speech therapy. Additionally, 37.5% of patients (n = 6) manifested minimal speech development prior to the age of 3–5 years. Furthermore, 75% of patients (n = 12) experienced respiratory distress during breastfeeding, and 81.3% (n = 13) exhibited frequent regurgitation, including through the nose. In 56.3% of patients (n = 9), a history of frequent purulent otitis media was documented, while 87.5% (n = 14) reported frequent rhinitis, sinusitis, and nasal breathing difficulties. Five patients (31.3%) in this cohort underwent non-partial adenotomy at their residence, which resulted in an exacerbation of rhinolalia. Following mesopharyngoscopy, a slight retraction of the soft palate was observed in 50% (n = 8) of the patients. However, 62.5% (n = 10) of the patients maintained the soft palate's mobility, which compensated for the effects of occult cleft palate. The diagnosis of the occult cleft palate without the use of fiberoptic endoscopy, X-rays, and CT was made challenging by the absence of an evident bifurcation of the uvulae with compensated palatopharyngeal insufficiency during the initial examination by an otorhinolaryngologist.

During the initial examination, hearing loss was diagnosed in 14 patients (87.5%). The various grades of hearing loss detected included conductive hearing loss grade I (40.6%), conductive hearing loss grade II (3.1%), conductive hearing loss grade III (3.1%), mixed hearing loss grade I (3.1%), grade II (9.4%), and grade III (3.1%), and sensorineural hearing loss grade I (3.1%). Mucosal stage EOM was detected in 21.9% of patients, adhesive otitis media in 21.9%, and chronic purulent otitis media with cholesteatoma in 18.8%.

Two patients presented without any history of hearing loss or otitis media. One patient exhibited a significantly nasal voice with a normal otoscopic appearance. He sought treatment at our clinic for the excision of recurrent adenoids. Additionally, he was diagnosed with an occult cleft palate and a pyomucocele in the left maxillary sinus. Subsequently, the patient underwent endoscopic maxillary sinus surgery.

Furthermore, fibrorhinoscopy demonstrated the presence of a deviated nasal septum in three patients (18.75%), as well as *concha bullosa* of the middle nasal conchae and hypoplasia of the maxillary sinus (6.3%).

CONCLUSIONS

Patients with an occult cleft palate require the specialized care of an otorhinolaryngologist. In the compensated form of this pathology, inadequate clinical symptoms can impede the otorhinolaryngologist's ability to diagnose occult cleft palate. In collaboration with a pediatrician, speech therapist, maxillofacial surgeon, and surgeon, timely diagnosis enables effective treatment and rehabilitation. The examination of patients with submucosal cleft palate indicates significant alterations in the middle ear, which affect auditory function and speech development. Occult cleft palate cases underscore the necessity for otorhinolaryngologists and pediatricians to be cognizant of this pathology to expedite the timely implementation of comprehensive rehabilitative strategies.

ADDITIONAL INFORMATION

Author contribution. All the authors have made a significant contribution to the development of the concept, research, and preparation of the article as well as read and approved the final version before its publication.

Personal contribution of the authors: *I.G. Andreeva* — experimental design, collecting and preparation of samples, data analysis, writing the main part of the text, literature review, making final edits, funding acquisition; *D.I. Marapov* — statistical processing, data analysis; *P.V. Tokarev, N.A. Andreev* — making final edits.

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Competing interests. The authors declare that they have no competing interests.

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Наибольший вклад распределен следующим образом: *И.Г. Андреева* — концепция и дизайн исследования, сбор и обработка материалов, анализ полученных данных, написание текста, обзор литературы; *Д.И. Марапов* — статистическая обработка, анализ полученных данных; *П.В. Токарев, Н.А. Андреев* — внесение окончательной правки.

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