

BENIGN SOFT TISSUE TUMORS OF MAXILLOFACIAL REGION IN CHILDREN: INCIDENCE, STRUCTURE, CLINICODIAGNOSTIC FEATURES

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Received 17/06/2017; accepted for printing 08/07/2017

ABSTRACT

Tumors in children have specific characteristics, in contrast to adults, and the age changes in metabolism and physiological functions of child body specify possible clinical differences in tumor processes among children of different age groups.

The article presents data on the incidence and features of clinical manifestations of some benign tumors of maxillofacial region in children.

It has been found that in the 10-year period of our observations, the number of children with benign tumors of maxillofacial region accounted for 7.5% of the total number of patients treated in the Surgical Unit of the Poltava Children's Municipal Clinical Hospital. Among the nosological forms, dermoid cysts (32.7%) and hemangiomas (26.0%) occurred most commonly, and the peak of morbidity (25.0%) was observed in infants.

In most cases (55.4%) soft tissue benign tumors and tumor masses of maxillofacial region occurred in girls, with more frequent (62.9%) occurrence of hemangiomas, whereas dermoid cysts were more frequently found in boys (61.7%). In 22.4% of cases, the pathological focus was detected on the neck, in 14.0% on the forehead, in 10.8% in the soft oral tissue, in 7.7% on the lower lip, in 7.7% on the cheek, in 6.2% on the upper lip, and in 3.1% of patients on the chin. In 25.9% of cases, hemangiomas covered several anatomical areas at ones.

Recurrences of tumors after surgery have been recorded in 6.7% of patients: in 43.0 % of children it happened after surgical excision of the middle neck cyst and per 28.5 % of the cases of angiomas and ranulas.

The comparative analysis of the clinical diagnosis and postoperative morphological study of the removed tumors has established that in 15.3% of cases the clinical diagnosis was different from the pathohistological one. In most cases it was associated with dermoids (75.0%), fibromas (15.6%) and lymphangiomas (9.4%).

KEYWORDS: children, benign tumors, maxillofacial region.

Introduction

According to the statistical data, children with tumors of the maxillofacial region account for 12-22% of all patients with tumors, and 90% fall to the share of benign processes, where mesenchyme-related tumors or, less frequently, epithelial neoplasms, prevail. At the same time, soft tissue tumors of face and neck account for 55-62% among the tumors of the maxillofacial region in children

[Zelenskiy V, Muhoramov F, 2008; Tkachenko P et al., 2013; Topolnitsky O, 2016].

Tumors in children have specific characteristics, in contrast to adults, and the age changes in metabolism and physiological functions of child body specify possible clinical differences in tumor processes among children of different age groups [Kolesov A et al., 1989; Bernadskiy Yu, 2000].

Gender differences are specific to the clinical course of some types of tumors that, in the context of a susceptibility to the hereditary transmission, should be considered in the diagnosis and selection of treatment mode [Timofeev A, 2010; Malanchuk V et al., 2011].

Morphologists also highlight a number of features

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of child tumoral processes, regardless of their localization, reminding of the possibility of transformation of some malignant tumors into benign ones [Paltsev M, Anitchkov N, 2005; Tkachenko P, 2015].

Unfortunately, no separate classification of the maxillofacial region tumors in children exists to date; therefore, general classification for all age groups has been used, proposed by the WHO/IARC Nomenclature Committee on Tumors [Hamilton S, Aaltonen L, 2000], to standardize the classification of tumors worldwide and formed on histological principle, based on the morphological structure, the assessment of benign or malignancy and features of the tumoral structure in the specific organs. Clinical and morphological classifications adapted for the maxillofacial region are also used in practical work [Harkov L et al., 2003; Tkachenko P, 2015].

Therefore, the occurrence of certain difficulties in the diagnosis at the pre-hospital stage, the high rate of recurrence (index of recurrence of benign tumors of various localization in patients of different age groups varies, according to V.A. Semkin and A.S. Zaretskaya (2010), from 0 to 62.5% [Sheshukova Ya, 2013] and post-operative complications in the treatment of benign tumors of soft tissues in general, which is closely related to their topographic-anatomical location and long-term asymptomatic growth. Against the background of the recent tendency to the increase in the number of patients, admitted to the hospitals regarding the tumors of the head and neck, this determines the relevance of the problem for the oral and maxillofacial surgery [Timofeev A, 2010; Malanchuk V et al., 2011; Tkachenko P, 2015; Topolnitsky O, 2016]. We, therefore, want to draw the attention of the medical general public to this problem, as it has been done in case of malignant tumors of the maxillofacial region in children [Tkachenko P et al., 2016].

The study aimed at representing data on the incidence, structure and features of the clinical manifestations of some nosological forms of benign tumors of head and neck in children.

MATERIAL AND METHODS

The fundamental scientific publications on the abovementioned issues have been thoroughly analyzed by the group of authors.

The staff members of the Department of Pediatric Oral Surgery with Propedeutics at the Higher

State Educational Establishment of Ukraine “Ukrainian Medical Stomatological Academy” have carried out a retrospective analysis of the hospital archive material of the Surgical Unit at the Poltava Children’s Municipal Clinical Hospital for the period of 10 years (2782 medical histories). The age of



Figure 1. Ultrasonic image of a child with cavernous lymphangioma of the left cheek and angle of mouth (hypoechoic masses are detected)

the patients varied from 2 months to 15 years.

Children with benign tumors of maxillofacial region have been examined using the general clinical methods and ultrasonography (Fig. 1), and computed tomography (Fig. 2), MRI (Fig. 3) on indication. In all cases, puncture and excision biopsy with subsequent verification of the biopsy material has been made by the staff members of the Department of Pathologic Anatomy with Autopsy Course, using the standard methods of histological studies [Merkulov A, 1969].

RESULTS AND DISCUSSION

It has been established that in the 10-year period 208 children with soft tissue benign tumors of the maxillofacial region were treated in the Surgical Unit of Poltava Children’s Municipal Clinical Hospital, which accounted for 7.5 % of the total number of patients in the Unit.

Among the nosological forms the most frequent were dermoid cysts (Fig. 4) and hemangiomas (Fig. 5), and the whole structure of the disease, presented in table 1, is as following: dermoid cysts – 32,7%, hemangiomas – 26.0%, fibromas (including angiofibromas) – 12.5%, ranulas – 11.5%, middle neck cysts – 7.7%, atheromas – 3.8%, lymphangiomas – 2.9%, brancial cleft cysts – 2.9%.

Table 1 shows that the peak of morbidity (25.0%) was observed in infants.

In most cases (55.4%) soft tissue benign tumors

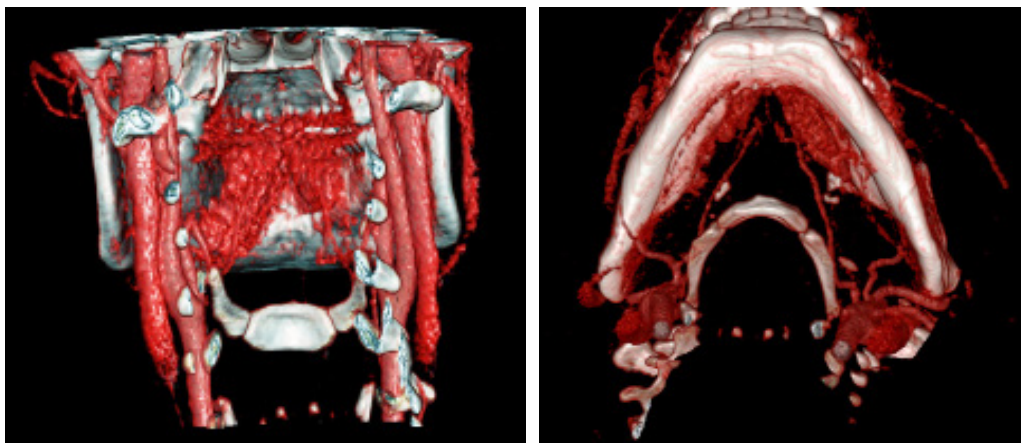


Figure 2. Fragments of computed tomography with vascular opacification of patient with recurrences of cavernous hemangioma of the mouth floor after several surgical interventions. Angioma residues of the left-sided localization are observed

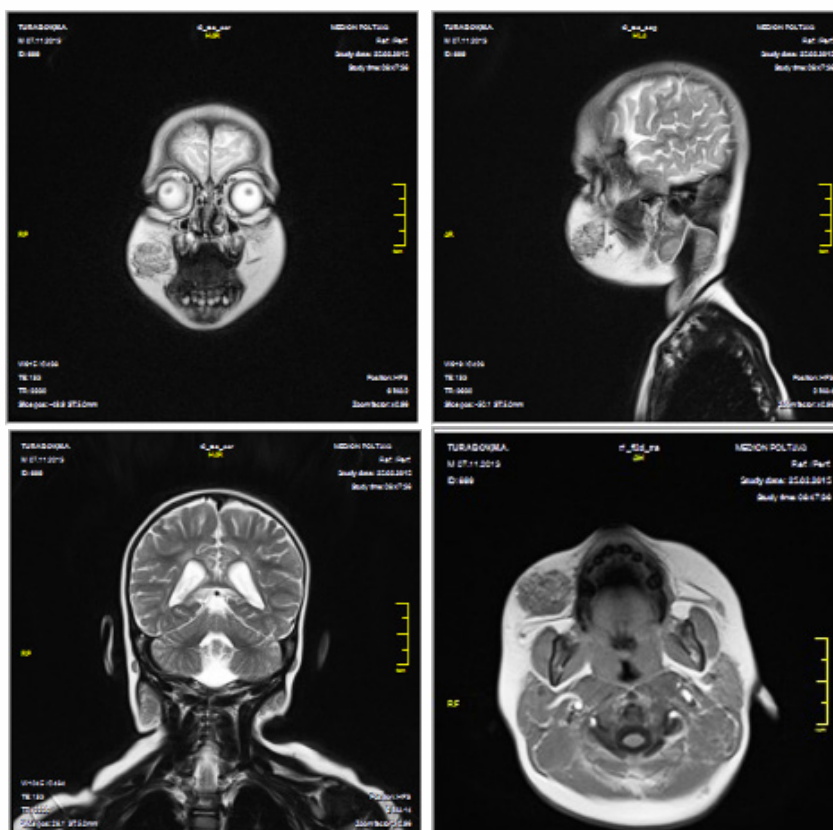


Figure 3. MRI-sections of a child with cavernous hemangioma of the left cheek

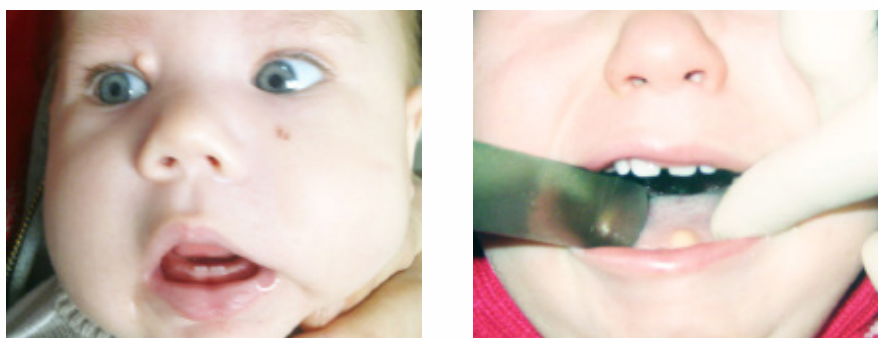


Figure 4. Appearance of children with dermoid cysts at the inner angle of the orbit (a) and on the tongue (b)

and tumor masses of maxillofacial region occurred in girls, with more frequent (62.9%) occurrence of hemangiomas, whereas dermoid cysts were more frequently found in boys (61.7%).

In 22.4% of cases, the pathological focus was detected on the neck area, in 14.0% on the forehead, in 10.8% in the soft oral tissue, in 7.7% on the lower lip, in 7.7% on the cheek, in 6.2% on the upper lip, and in 3.1% of patients on the chin. Noteworthy, in 25.9% of cases (14 children), hemangiomas covered several anatomical areas at ones (Fig. 6).

Importantly, no clear specific symptomatology has been noted in all patients and clinical signs, despite the nosological form, localization and the stages of the growth of the neoplasm, were similar: normal general condition of the patients, roundness of the shape or lobular structure of tumor, etc., and the color of the tumors, except for the majority of hemangiomas, was usually similar to the color of the surrounding tissues.

Recurrences of tumors after surgery were recorded in 14 (6.7%) patients: in 6 (43.0%) children it happened after surgical excision of the middle neck cyst and per 4 cases of angiomas (Fig. 7) and ranulas (28.5%, respectively).

Comparative analysis of the clinical diagnosis and postoperative morphological study of the removed tumor has established that in 15.4% (32 patients) of cases the clinical diagnosis differed from the patho-

histological one. In most cases it was associated with dermoids (24 patients; 75.0%), fibromas (5 patients; 15.6%) and lymphangiomas (3 patients; 9.4%).

Below is an example from the clinical practice to visualize the presented material and clarify the perception of its content.

The parents of a newborn child were referred to the clinic at the Department of Children's Oral Surgery with Propedeutics from the maternity home to receive medical advice regarding the complaints on the presence of child's pharynx a tumor mass, identified immediately after birth, which caused heavy breathing obstructing the entrance to the oral- and nasopharynx.

Examination showed symmetrical face, and coloring of the baby's skin, and visible mucous membranes were natural. Skin turgor was not changed, mouth was opened freely.

On the pharynx area an ovoid, soft elastic, painless circumscribed tumor mass was visualized; surface and color of mucosa was similar to the adjacent tissues. The mass resembled additional tongue and considering its size (9×5×3 cm) it subsequently could lead to asphyxia.

The child was hospitalized for planned surgery and after extensive preoperative examination, involving medical professionals of the allied specialties, the tumor mass was removed in intubation anaesthesia within the healthy tissues, and the wound was sutured tightly (Fig. 8).

Table 1

The structure of the soft tissue benign tumors of face and neck in children

Age (years)	Infancy (0-1)		Toddlerhood (1-3)		Early childhood (3-7)		Middle childhood (7-12)		Late childhood (12-15)		Number of cases	
	Total	%	Total	%	Total	%	Total	%	Total	%	Total	%
Nosological form												
Dermoid cyst	18	26.5	8	11.8	16	23.5	16	23.5	10	14.7	68	32.7
Hemangioma	26	48.2	18	33.3	—	—	4	7.4	6	11.1	54	26.0
Fibroma	2	7.7	4	15.4	—	—	8	30.8	12	46.1	26	12.5
Ranula	—	—	2	8.3	2	8.3	8	33.4	12	50	24	11.5
Atheroma	2	25	—	—	—	—	6	75	—	—	8	3.8
Middle neck cyst	—	—	—	—	—	—	8	50	8	50	16	7.7
Lymphangioma	4	66.7	—	—	2	33.3	—	—	—	—	6	2.9
Brancial cleft cyst	—	—	4	66.7	2	33.3	—	—	—	—	6	2.9
Total	52		36		22		50		48		208	100



Figure 5. Appearance of children with cavernous hemangiomas of the upper lip (a), tongue (b) and auriculomasseteric region (c, d)

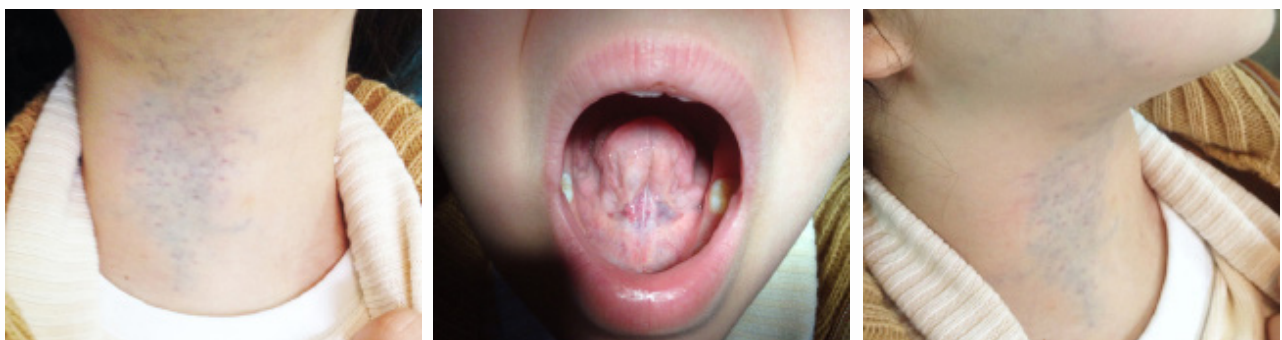


Figure 6. The child with hemangiomatosis: capillary hemangiomas of the auriculomasseteric region and anterior surface of the neck, cavernous hemangioma of the mouth floor



Figure 7. Appearance of patients with recurrences of cavernous hemangioma of the mouth floor (a) and lymphangioma of the tongue (b). Post-operative cicatricial tissues are detected



Figure 8. The appearance (a, b) and macrospecimen of tumor mass in a newborn child

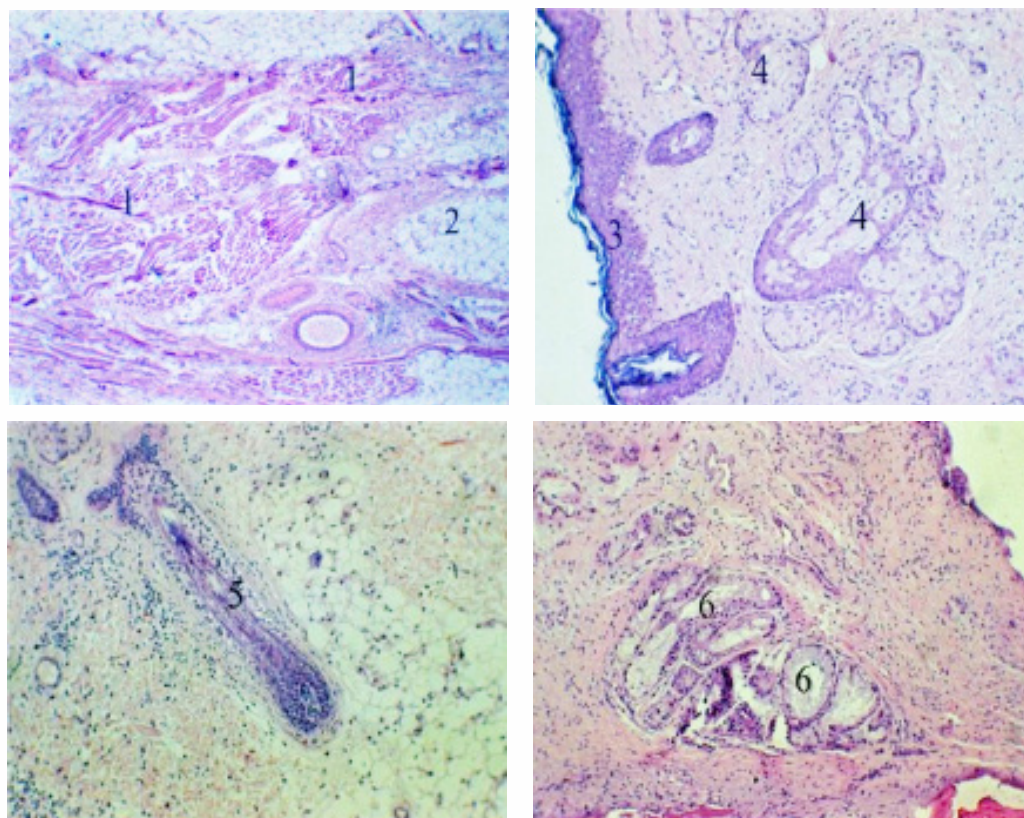


Figure 9. Microscopic structure of the tumor mass of the newborn child. Hematoxylin and eosin stain. Magnification $\times 150$ (a, b, d), $\times 350$ (c). 1 – cross-striated muscular tissue; 2 – adipose tissue; 3 – stratified squamous epithelium; 4 – acini of the sebaceous glands; 5 – hair follicle; 6 – glandular formations

Macroscopically the tumor mass had thin pedicle, and alternation of the areas of soft-and-solid elastic consistency was detected on palpation.

The course of postoperative period was without complications. Postoperative wound was antiseptized locally, and antibacterial therapy (medications per *kg/body weight*) was provided within the framework of the general drug therapy to prevent wound abscess. The suture was removed on day 10; the wound was healed by primary intension.

Surgical material was sent to histological study, the results of which showed that the tumor mass was represented primarily by a fibrous connective tissue, complexes of chaotically-oriented bundles of cross-striated muscle fibers and inclusion of the adipose tissue (Fig. 9a). The fibrous connective tissue was totally covered by the stratified squamous epithelium with the signs of keratinization. Morphologically, the epithelial cells, arranged in 6-10 layers with a clearly differentiated basal layer, were almost similar to epidermis cells. It should be noted that the thickness of the epithelial integument and, consequently, the number of its

layers in different parts of the tumor mass were somewhat different (Fig. 9b).

In addition, hair follicles, acini of the sebaceous and sudoriferous glands and separate glandular formations, structurally resembling oral minor salivary glands were found everywhere in the connective tissue (Fig. 9 c, d).

This microscopic image matched with the morphological structure of teratoma [Paltsev M, Anitchkov N, 2005].

On the basis of clinical data and findings of the morphological study of the postoperative material, we made the final clinical diagnosis: teratoma of the pharynx.

We have already mentioned this pathology in our publications, describing the examples from our own practice, both clinical and morphological [Tkachenko P, 2014; 2015].

In sum, according to our observations, children with soft tissue benign tumors of the face and neck account for 7.5% of the total number of patients with hospital surgical pathology with predominance of dysontogenetic tumors, among

which dermoid cysts (32.7%) and hemangiomas (26.0%) prevail. Benign tumors are more frequently found in children under 1 year old (25.0%), and in girls (55.0%).

Considering the fact that the prognosis for children with abovementioned pathology is determined by the option of histological structure and primary localization of the tumor, the timeliness

and adequacy of the treatment activities, implementation of the advanced methods of diagnostics with the high level of awareness is crucial in the improvement of the effectiveness of health care provided for patients of this category, contributing to the correct diagnosis, choice of the treatment procedure and determination of the extent of surgical intervention at the pre-hospital stage.

REFERENCES

1. *Bernadskiy YuI.* [Essentials of oral and maxillofacial surgery and dental surgery] [Published in Russian]. Moscow, Russia: Meditsinskaya Literatura. 2000. 404p.
2. *Hamilton SR, Aaltonen LA.* World Health Organization Classification of Tumours. Pathology and Genetics of Tumours of the Digestive System. Lyon: IARC Press. 2000. 314p.
3. *Harkov LV, Yakovenko M, Chehova IA.* [Pediatric oral surgery] [Published in Ukrainian]. Kyiv, Ukraine: Kniga-plyus. 2003. 480p.
4. *Kolesov AA, Vorobyov YuI, Kasparova NN.* [Tumors of soft tissues and facial bones in children and teenagers] [Published in Russian]. Moscow: Meditsina. 1989. 302p.
5. *Malanchuk VO, Logvinenko IP, Malanchuk TO.* [Oral and maxillofacial surgery] [Published in Ukrainian]. Kyiv, Ukraine: Logos. 2011; 2: 3-120.
6. *Merkulov AB.* [Course of pathohistological technique] [Published in Russian]. St. Petersburg: Meditsina. 1969. 237p.
7. *Paltsev MA, Anitchkov NM.* [Atlas of human tumor pathology] [Published in Russian]. Moscow, Russia: Medicine. 2005. 424p.
8. *Semkin VA, Zaretskaya AS.* [Outpatient treatment of patients with extensive bone cysts of the mandible] [Published in Russian]. Stomatologiya. 2010; 3: 34-36.
9. *Sheshukova YaP.* [Structure and incidence of benign tumors of facial soft tissues, maxillary bones in children and adults] [Published in Ukrainian]. Poltava, Ukraine: Ukrayinskiy Stomatologichniy Almanah. 2013; 3: 46-49.
10. *Timofeev AA.* [Maxillofacial surgery] [Published in Russian]. Kyiv, Ukraine: Meditsina. 2010. 576p. ISBN 978-617-505-038-5.
11. *Tkachenko PI, Starchenko II, Belokon SA, Gurzhiy YeV, Starchenko OV.* Clinical and tactical approaches in the diagnosis of malignant tumors of maxillofacial area in children. The New Armenian Medical Journal. 2016; 10(3): 27-33.
12. *Tkachenko PI, Starchenko II, Belokon SA, Gurzhiy YeV.* [Teratoma: publication data and own observations] [Published in Ukrainian]. Kharkov, Ukraine: StomatologInfo. 2014; 1: 21-25.
13. *Tkachenko PI, Starchenko II, Bilokon SO.* [Cysts of maxillofacial region (clinical and morphological aspects)] [Published in Ukrainian]. Poltava, Ukraine. 2013. 103p.
14. *Tkachenko PI.* [Soft tissue benign tumors of maxillofacial region and salivary glands in children] [Published in Ukrainian]. Poltava, Ukraine. 2015.
15. *Topolnitskiy OZ.* [Dentistry of children age] [Published in Russian]. Moscow, Russia: GEOTAR-Media; 2016. 311p.
16. *Zelenskiy VA, Muhoramov FS.* [Pediatric oral and maxillofacial surgery] [Published in Russian]. Moscow, Russia. 2008. 206p.