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TUMORS AND JAW CYSTS FORMED FROM THE RESIDUAL GLANDULAR EPITHELIUM

ПУХЛИНИ ТА КІСТИ ЩЕЛЕП ІЗ ЗАЛИШКОВОГО ЗАЛОЗИСТОГО ЕПІТЕЛІЮ

Summary. The article deals with the clinical cases of the glandular-odontogenic cyst of the front part of the lower jaw and adenocarcinoma of the upper jaw which is slightly differentiated due to it. The sources and reasons of the glandular-odontogenic cysts' development, adenocarcinoma, and central mucoepidermoid carcinoma of the jaw have been clarified in the research; their realtion to the residual glandular epithelium has been also described.

Key words: the glandular odontogenic cyst, adenocarcinoma of jaw, the residual glandular epithelium.

Резюме. В статті представлено клінічні випадки залозисто-одонтогенної кісти фронтального відділу нижньої щелепи та помірно-диференційованої аденокарциноми верхньої щелепи. Встановлено джерела та причини розвитку залозисто-одонтогенних кіст, аденокарцином та центральних мукоепідермоїдних карцином щелеп, описано зв'язок їх із залишковим залозистим епітелієм.

Ключові слова: залозисто-одонтогенна кіста, аденокарцинома щелеп, залишковий залозистий епітелій.

Introduction. Jaw cyst is one of the most common pathologies among the diseases of the jaw-facial area. Among the surgeries, performed by the dental surgeons on an outpatient basis, operations on the odontogenic jaw cysts take one of the first places after the tooth surgical removal, and among the patients who need inpatient treatment in the stomatolagy those with odontogenic cysts make 8%[1, p.4-7, 2, p.385, 3, p.320]. The clinical course of these diseases is fully investigated, and does not present any special diagnostic difficulties. Though, sometimes after the cystectomy operation and pathological-anatomical examination the dental surgeon receives quite unexpected results, when the inner bone tumors and cysts contain salivary gland tissue.

For the first time this bone was called "sialo odontogenic" by Padayachee and Van Wyk in 1987. Later Gardner together with the co-authors (1988) described it as "glandular odontogenic cyst" (GOC). This term was accepted in 1992 by the International Classification of Diseases of the World Health Organization (WHO) and is

widely used in the scientific publications. One of the early cases of the cyst observations is the report of Waldron and Koh (1990), who discussed it in the possible relation to the central mucoepidermoid jaw tumor; Ficarra and others (1990) who also described the case in which they diagnosed the given type of the cyst as an inferior mucoepidermoid carcinoma [4, p.228]. In 2008 H. Geha and others described the case of the central adenocarcinoma in the area of the odontogenic follicular cyst of the upper jaw [5, p.126].

GOC is a rare disease. Nowadays more than 100 cases of the given pathology have been described. From the general number of 112 patients, 62 were men (55,4 %), and 50 – women (44,6 %). 66,3% of cysts located in the lower jaw, mostly in the front area. Roentgenologically cysts had distinct boundary lines and multicellular structure; more seldom they had unicellular structure. Clinical characteristics are the following: aggressive course of disease with heightened relapse susceptibility (from 21 to 55%). Besides, many authors mention that unicellular cysts have lower relapse susceptibility than the multicellular ones [4, p.226].

Though, in modern medical literature we didn't find any description of reasons and sources of such cysts' development.

The objective of our research is describing of two clinical cases of the given pathology and investigating of the possible development reasons.

Tools and methods.

The case of the glandular-odontogenic cyst of the front part of the lower jaw was observed in a 43-year-old patient on the basis of Chernivtsi RCH in 2013. Sightly differentiated adnocarcinoma of the upper right jaw was diagnosed in a 55-year-old patient on the basis of Vinnytsia Regional Clinical M.I. Pyrohov Hospital in 2008. In both cases the preliminary diagnosis was "odontogenic cyst" and surgery "cystectomy" under local anesthesia was performed.

Results and discussion.

Patient R. 43, asked for help in the dental surgery department of Chernivtsi RCH, he complained about an intermittent pain in the front part of the lower jaw and recurrent lower lip swelling. Some confusion appeared 8 months ago, the patient didn't see any connection to it.

In the front part of the lower jaw the transitional fold of the mouth is slightly swollen, painful to touch. Percussion of intact 31, 32, 41, 42 teeth is quite painful. On the orthopantomogram there is a distinct thinning of the bone tissue in the front part of the lower jaw which has a multicellular structure with distinct boundary lines with the diameter 35 mm. Teeth crowns 31, 32, 41, 42 are intact, but 1/3 of their roots immerse into the cyst cavity.

Preliminary diagnosis: "Radicular cyst of the upper jaw in the area of 31, 32, 41, 42 teeth". In the presurgical period nerves in teeth 31, 32, 41, 42 were removed and teeth were endodontically treated. Cystectomy on the lower jaw in the front part with the roots' resection of 31, 32, 41, 42 teeth was performed.

Results of the pathohistological investigation: fibrous connective tissue with areas of salivary glands' clusters, hemorrhage.

The final diagnosis: "Glandular-odontogenic cyst of the front part of the lower jaw".

Patient O, 55, asked for medical help in the Jaw-Facial Department of Vinnytsia Regional Clinical M.I. Pyrohov Hospital with complaints about unpleasant feelings in the

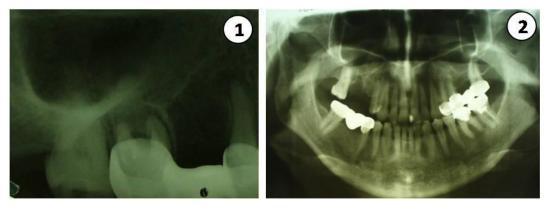
area of the first molar tooth on the right which she was having during 3 years. Unpleasant feelings appeared periodically, the patient didn't ask for medical help before. One month ago she had a toothache when biting.

The general state of the patient is satisfactory. There are no extraoral changes. It has been defined that in the mouth cavity tooth 16 is under the stamped crown and used as a bridge-like prosthesis. Percussion is painful but accompanied by aggravation of unpleasant feelings. On the alveolar shoot in the tooth area there is a swelling of the cortical plate of the oval form with relatively distinct boundaries, diameter about 1 cm, with the softening in the center (absence of the cortical plate).

On the intraoral target roentgenogram of the alveolar shoot of the right upper jaw in the area of tooth 16 the multicellular thinning of the bone tissue with diameter about 1,3 cm is indicated. The tooth wasn't treated endodontically.

Bundle of the right upper jaw of the sinus was observed distinctly without any pathological changes (Pic.1).

Preliminary diagnosis: "Radicular cyst of the upper jaw in the area of tooth 16". Tooth 16 was removed and cystectomy was performed under the local anesthesia. While removing the pathological locus it was indicated that it didn't have any distinct membrane bone and looked like a soft white-color formation, which is soldered with the mucous gland of the upper-jaw sinus. While inspecting the surgical wound there were no pathological changes in the area of the upper-jaw sinus indicated. The wound was irrigated, filled with blood and sutured with the means of plastic surgery of the oroantral connection with the mucous oxygen pane taken from the vestibular surface of the alveolar outgrowth. The removed tissue was sent for a histological investigation. The wound healed by first intention without complications.



Pic.1 The roentgenogram of the upper jaw of the patient O, 55: 1) target intraoral (before surgery), 2) orthopanogram (after surgery)

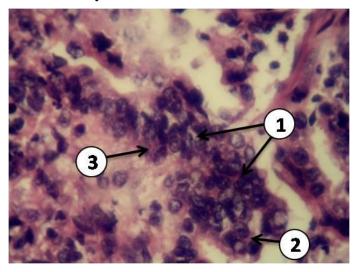
Numeral glandular structures with different number of tumor cells' layers were indicated while making a histological investigation. Cell elements had big polychromatic nuclei and eosinophilic cytoplasm. Among the chaotically allocated cell elements also there were solid complexes (cells that join together).

Some cells had secretion marks. Screen and folliculus like structures, and also mucous filling between the cells were observed. Space between the glandular structures was filled with the chaotically located cells without distinct boundary lines with solid dark nuclei. The tissue tumor infiltrated by inflammatory cells.

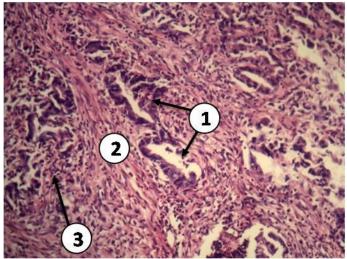
It was indicated that in all sections there was cellular and nuclear polymorphism, nuclei polychromy of the cancer cells, secretion marks of the tumor epithelium with the glands' filling of the eosinophil secretion, epithelium desquamation (pic.2).

Tumor areas with differentiating marks were observed. Salivary glands of the tumor swelling placed among the soft fibrous tissue, infiltrated by the inflammatory cell elements, where the segmented-nucleic leukocytes, one at a time lymphocytes, and plasmocytes with the displaced nuclei were indicated (pic.3).

Results of the histological investigation: adenocarcinoma with the slight extent of differentiating and inflammatory infiltration.



Pic. 2 Adenocarcinoma fragment with the slight extent of differentiating and inflammatory infiltration. Lens x 40. Ocular x10. Color: hematoxylin-eosin. 1) nucleic polymorphism; 2) solid structures; 3) eosinophil secretion.



Pic.3 Adenocarcinoma fragment with the slight extent of differentiating and inflammatory infiltration. Lens x10. Ocular x10. Color: hematoxylin-eosin. 1) salivary glands with hyperchromic epithelium; 2) fibrous tissue; 3) inflammatory cell elements.

The final diagnosis: "Slightly differentiated adenocarcinoma of the right upper jaw".

Discussing results. The indicated glandular epithelium in the jaw cysts and intra bone jaw tumors, which develop from the salivary gland tissues, appears to be quite rare. These findings in the range of cases are stated to be casuistic, as a fact that cannot be discussed, and are dropped away as a mistaken notion.

Report on the occurrence of the glandular epithelium in the jaw embryo tissues was found in the research of Nikanorow (1990). Due to the investigation results on 96 embryos the author determined that laying of the glandular epithelium takes place in the first part of the prenatal ontogenesis at the age of 11-12 weeks in the "tooth gums" (author's term) of the embryo.

In our opinion these formations cannot be called "tooth gums" as there are no formed alveolar outgrowths and teeth. Possibly glandular elements occur under the ectodermal layer. They are also called the submucous glands. Starting from the 17-week age embryo the number of the glandular elements in "toothgums" quickly decreases, which is followed by their development breaking: glycogen disappears in them and their function breaks. By the end of the prenatal ontogenesis the "toothgum" mucous is free from the glandular tissue, though seldom its remnants stay in it. To the author's point of view, the research allows to make an assumption that in the postnatal period these remnants can be a reason for many periodontal diseases [7]. In his article the author mentions of the found single researches on the glandular epithelium in the embryonic period which had quite controversial character. Thus, I.S. Kudrin (1968) thought that the glandular structures are absent in the alveolar outgrowth tissues. Though, Hirtle (1874) in the last century accentuated on their presence [7, p.44-45].

Reasons for the jaw glandular epithelium appearance are usually not discussed. Thus, professor S.M. Shuvalov assumed that in the period of the embryonic jaw development in the ontogenesis the embryo copies the development stages of phylogenesis of some animals (reptiles, mammals). Thus, preservation of glandular epithelium in the teeth development is observed by some species of reptiles and mammals [6]. In the process of the head phylogenesis the mouth cavity formation happened with the following special-purpose organs' development like teeth, tongue and salivary glands. Their development is closely connected. Fish and amphibians have very primitive mouth glands. In the poisonous snakes, lizards and mammals, like viper, duckbill, solenodon, some mouth glands of the serous type turned into a complicated tubular poison gland, which is connected with the teeth (pic. 4) [6, p.144].







Pic. 4 Animals with poisonous secretion, connected with teeth. 1) the Hispaniolan solonodon; 2) poisoneous second low cutting tooth of the solenodon; 3) snake's poisonous teeth.

In a human also with the fold of the salivary gland tissues, big and small, glandular cells are sometimes located behind their capsules' boundaries. It is known that the glandular epithelium fold in a human develops together with the dental plate during first three months of the embryonic development after which its involution happens. Though, sometimes cells of the residual epithelium are preserved and can act as a basis for adenocarcinoma development and appear in the evolutional cysts' structure.

Conclusions.

- 1. Teeth development is directly connected with the salivary glands' development not only in ontogenic but also in phylogenetic aspects.
- 2. In the postnatal period remnants of the glandular epithelium are the reason for glandular odontogenic cysts.
- 3. The cells of the residual epithelium can be a reason for the jaw adenocarcinoma and central mucoepidermoid carcinoma development.

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