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FEATURES OF THE COURSE, CLINICAL AND MORPHOLOGICAL CHARACTERISTICS OF AMELOBLASTOMA AND FIBROUS DYSPLASIA IN CHILDREN

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The article highlights the features of the clinical course and morphological characteristics, diagnostic measures and treatment of children with ameloblastoma and fibrous dysplasia of the jaw bones. Absence of clear clinical manifestations, similarity of symptoms and nature of the course of ameloblastoma and fibrous dysplasia is inherent in all benign tumors of the maxillofacial area in children. They cause a significant percentage of diagnostic errors, making the issue of their differential diagnosis quite relevant for surgical dentistry and maxillofacial surgery.

Key words: adamantinoma, fibrous dysplasia, benign tumors, children.

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ОСОБЛИВОСТІ ПЕРЕБІГУ І КЛІНІКО-МОРФОЛОГІЧНА ХАРАКТЕРИСТИКА АМЕЛОБЛАСТОМИ ТА ФІБРОЗНОЇ ДИСПЛАЗІЇ У ДІТЕЙ

У статті висвітлені особливості клінічного перебігу та морфологічних характеристик, діагностичних заходів і лікування дітей з амелобластомою та фіброзною дисплазією щелепних кісток. Відсутність чітких клінічних проявів, подібність симптоматики і характер перебігу амелобластоми та фіброзної дисплазії, що є притаманним для усіх доброякісних пухлин щелепно-лицевої ділянки у дітей, обумовлюють значний відсоток діагностичних помилок, роблячи питання їх диференційної діагностики доволі актуальним для хірургічної стоматології і щелепно-лицевої хірургії.

Ключові слова: адамантинома, фіброзна дисплазія, доброякісні пухлини, діти.

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It is well known that some researchers under the term "ameloblastoma" (AB) combine a group of odontogenic tumors of epithelial origin located in the jaw thickness. These include both true AB (adamantinoma, adamantinomous epithelioma, adamantinolastoma, etc.) and ameloblastic fibroma (ameloblastic fibroodontoma), adenoameloblastoma (adenomatoid odontogenic tumor), ameloblastic fibro-odontoma, and odontoameloblastoma. They tend to infiltrative growth with significant aggressiveness, the probability of jawbone destruction and further invasion into the soft tissue or the maxillary sinus [1, 2, 5].

In turn, fibrous dysplasia (FD), which was isolated in a separate nosological form only in 1940, also has its "varieties" ("leontiasis ossea", "hemicraniosis", McCune–Albright syndrome, Paget's disease). Each of them has its own clinical features, and they occur in different age groups of different genders [5, 8, 9].

Against the background of the lack of clear clinical manifestations that is generally inherent in all benign tumors (BT) of the maxillofacial area (MFA) [1–3, 9], the similarity of symptoms and the nature of the course of AB and FD cause a significant percentage of diagnostic errors, which makes quite topical issues of their differential diagnosis for surgical dentistry and maxillofacial surgery.

The purpose of the study was to summarize the results of scientific research on the clinical features of the course, morphological characteristics, diagnostic measures and treatment of children with ameloblastoma and fibrous dysplasia of the jaw bones.

Materials and methods. For this purpose we carried out a generalized analysis of fundamental scientific works and publications on these issues with a comparison of the results of our own scientific and practical research on these nosological forms.

Clinical section of the study concerns 11 children with fibrous dysplasia and 9 children with ameloblastoma who were treated in the Clinic of the Department of Pediatric Surgical Dentistry on the basis of the surgical department of Poltava Children's City Clinical Hospital for 10 years. There were 6 girls (54.5 %) and 5 boys (45.5 %) with fibrous dysplasia, and 6 girls (66.7 %), and 3 boys (33.3 %) with ameloblastoma.

General clinical (examination, palpation), laboratory (general blood and urine tests, biochemical blood test and blood sugar test) and additional examination methods were used to establish the final clinical

diagnosis. In particular, puncture, incisional biopsy, radiography and, if necessary, depending on the location of the formation and its size, computed tomography (CT) or magnetic resonance imaging (MRI).

All patients underwent comprehensive treatment according to the nosological form of the disease in each case, which included surgery to fill the bone defect with osteoplastic material, close it with titanium plates or using a bone suture [6] (taking into account areas of bone growth) as well as further management of postoperative bone wounds and soft tissue wounds according to the general principles of surgery with appropriate pharmacotherapeutic support (use in age-appropriate dosages of antibacterial (if necessary), anti-inflammatory, hyposensitizing and tonic drugs).

Employees of the Department of Pathological Anatomy with a sectional course on preparations manufactured according to conventional methods [5], studied the microscopic structure of postoperative material, which allowed to verify the histological structure of tumors and establish a final diagnosis.

Results of the study and their discussion. Asymptomatic, slow nature of the AB growth in the absence of visual clinical manifestations did not allow sick children and their relatives to state the beginning of tumor development in all our observations. Therefore, they usually sought help for a long time after the tumor formation, complaining mainly about the pronounced asymmetry of the face due to the tumor contour in the angle (6 cases – 66.7 %) and the ramus (3 cases – 33.3 %) of the lower jaw (LJ) and aching pain in the jaw and teeth. In 1 child (11.1 %) with AB, who had previously had the 46th tooth extracted at the place of residence, the alveolar socket did not heal for 3 months.

If the tumor reached a large size, then due to the difficulty of opening the mouth was disturbed chewing and speech, which was observed in 3 cases (33.3 %) with its contour in the ramus area.

In 1 follow-up (11.1 %), the child's relatives focused on the occurrence of periodic soft tissue swellings in the projection of the mandibular affected area. This clinical picture was joined by symptoms of "acute odontogenic inflammatory process", which was accompanied by the formation of fistulas with purulent or bloody exudate on the mucous membrane (MM) around this focus.

Objective examination in all cases determined the facial asymmetry due to painless, spindle-shaped, dense, smooth or hilly thickening of the mandible, over which the skin is not changed in color and gathered in a wrinkle. 5 children (55.6 %) had enlarged regional lymph nodes, but this was true for long-standing cases.

In the oral cavity there was swelling of the mandibular body or ramus with smoothing of the transitional and pterygomandibular folds with signs of expansion of the vascular system over the pathological focus, and the MM generally not changed in the color. Teeth in the area of the pathological focus were always loose, in 3 cases (33.3 %) the convergence of dental crowns was visually determined, and in 4 – (44.4 %) X-ray images revealed divergence of their roots.

A significant bone defect in 5 patients (55.6 %) caused a symptom of “parchment crunch”, and in 2 cases (22.2 %), the tumor sprouted the mandibular cortical plate, spreading to the adjacent soft tissues.

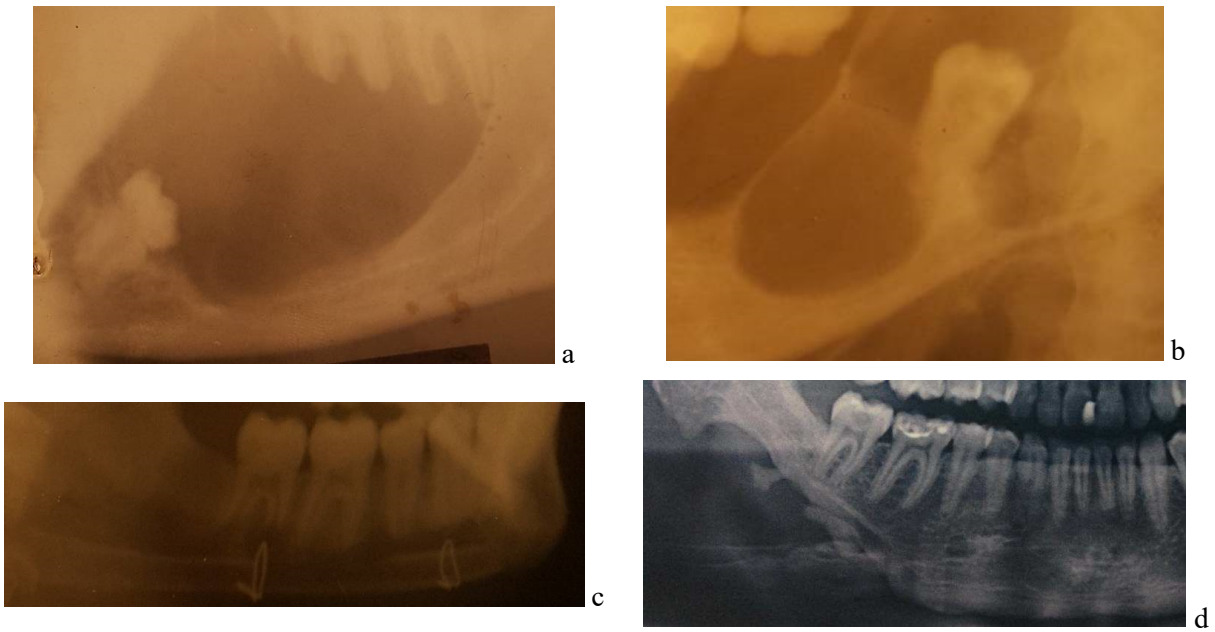

Fig. 1. X–ray picture of mandibular AB (a – violation of bone integrity near the retained 47th tooth; B – retained 38th tooth in the bone cavity) and fragments of orthopantomograms of patients with monostotic form of FD (C – bone grafting and bone suture after FD treatment in the mandibular body and angle; D – 7 years after surgery for the monostotic form of FD of the mandibular body on the right).

Given the lack of uniformity in the clinical picture of AB, X-ray of the jaws was of great importance in the diagnosis, which in the vast majority of cases (7-77.8 %) determined rounded cavities of various sizes with clear uneven borders, separated by bone membranes, around which small foci were located. The cavities came into contact, overlapped and even merged. Bone structures thinned or disappeared with tumor growth. In some cases (3–33.3 %), AB was represented by a single cystic cavity. The tumor could have retained or dystopic tooth or preserved dental roots (fig. 1 A, B).

To clarify the diagnosis, a puncture of the formation was performed, revealing yellow or brown fluid, and CT or MRI was performed to clarify the location of the tumor and to plan the volume of surgical intervention.

The clinical course of fibrous dysplasia (Braitsev-Lichtenstein disease) generally resembled the symptoms of AB.

In 3 cases (27.3 %) the disease was hereditary.

In all our observations, the monostotic form of FD was determined (fig. 1 c, d).

All patients with AB and FD underwent radical removal of tumors within healthy tissues (Fig. 2). Surgical interventions in all patients with FD were performed at 14–15 years of age due to the fact that recurrence is possible before the end of puberty.

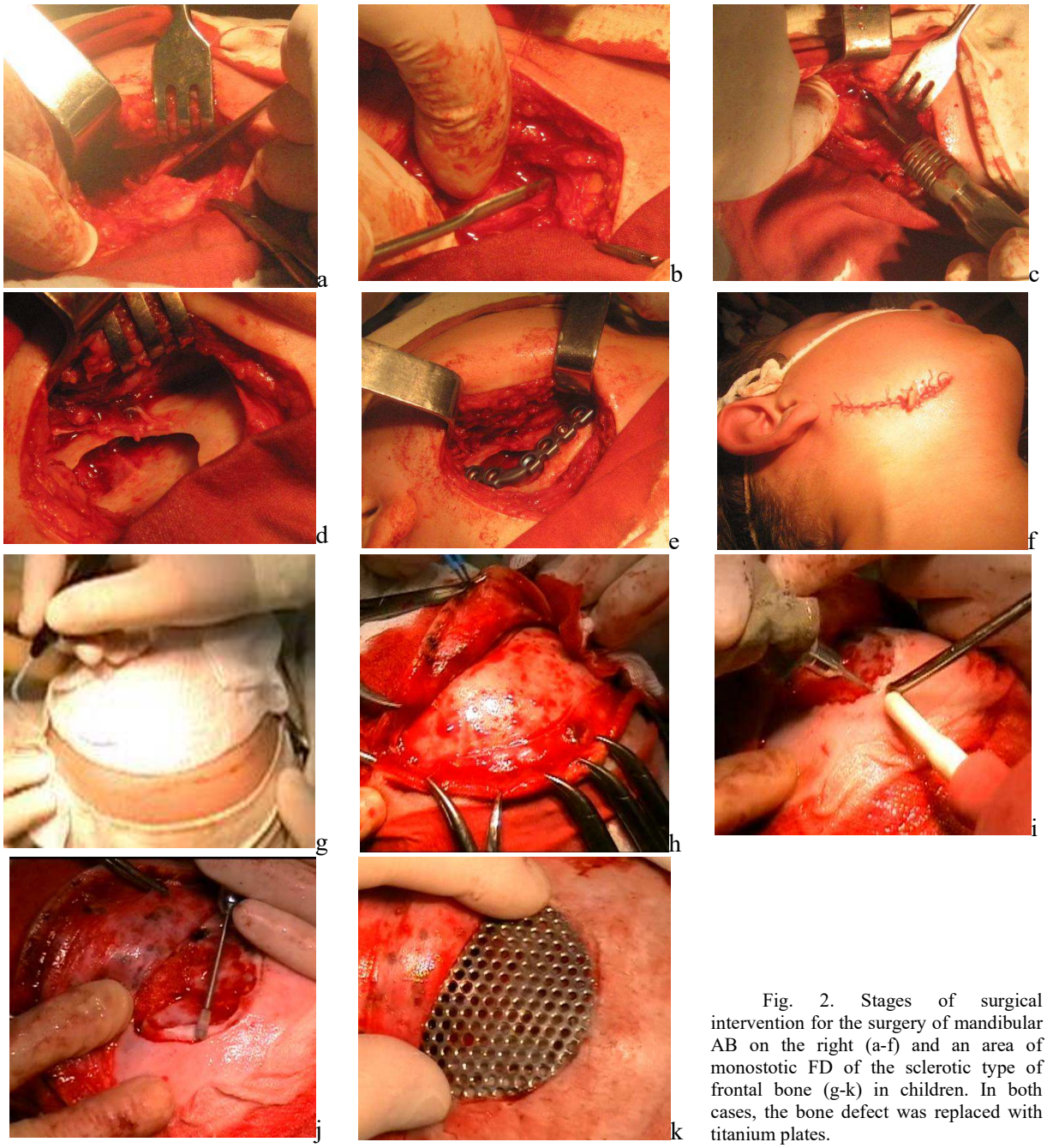

Fig. 2. Stages of surgical intervention for the surgery of mandibular AB on the right (a-f) and an area of monostotic FD of the sclerotic type of frontal bone (g-k) in children. In both cases, the bone defect was replaced with titanium plates.

At the same time, patients with AB located within the body or the inner edge of the mandibular ramus underwent gentle resection of the jaw while maintaining bone continuity, and during surgery they retreated 2 cm from the radiologically visible borders of the tumor towards healthy bone.

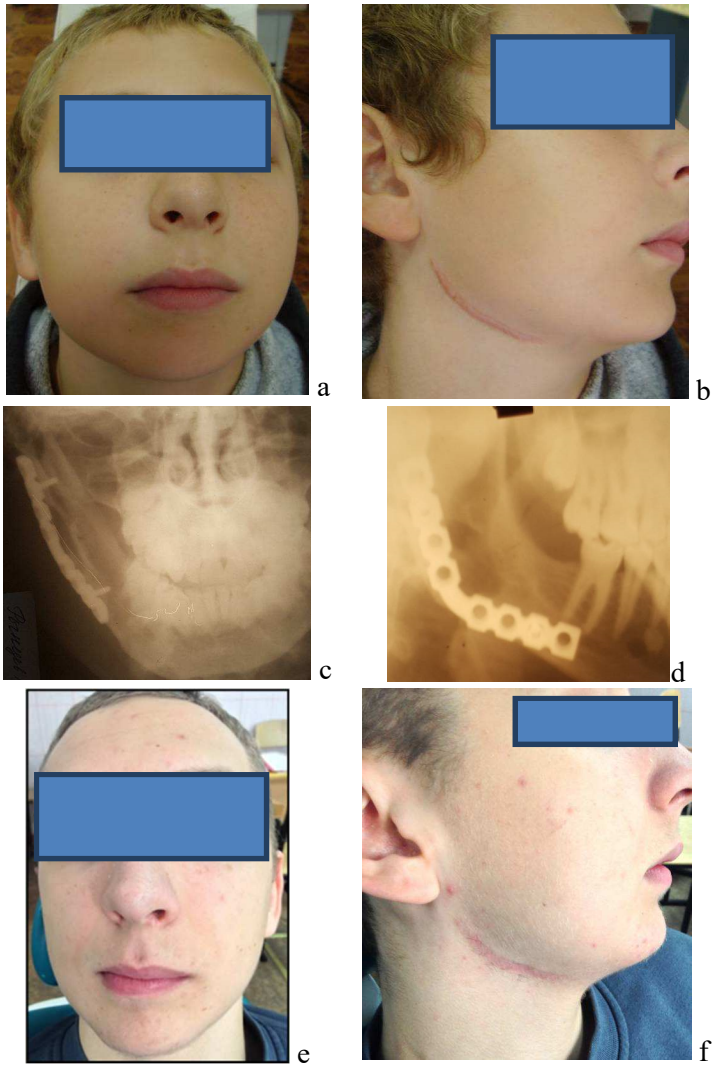

Fig. 3. General view of the child's face (a, b) and radiographs (c, d) 1 year after the surgery of AB on the right mandible (c, d – reconstructive titanium plate is observed) and general appearance of the face of the same child 7 years after AB resection (facial asymmetry and postoperative scar in the submandibular area are determined)

During surgery for FD, it was removed by partial resection of the jaw involved in the pathological process, leaving, if possible, a bony edge.

Basically (9 cases – 81.8 %), preference was given to curettage of the affected areas with leveling the edges of the bone tissue. Usually, during surgery, bone grafting was performed simultaneously according to generally accepted methods.

We did not observe any complications during surgery or in the postoperative period (fig. 3). However, we should not forget about the likelihood of mandibular pathological fracture.

In all cases, AB microscopically consisted of round or irregular shaped islets, bounded by odontogenic epithelium and with process polygonal stellate-shaped cells in the center, resembling the structure of an enamel organ. Dystrophic processes in these structures led to the cystic formation (Fig. 4 a). In general, this morphological picture corresponded to the structure of follicular AB.

In turn, the microscopic picture of FD was classic: the foci are represented by fibrous tissue, among which low-calcified bone beams of primitive structure and osteoid beams were determined. The fibrous tissue in some areas consisted of randomly arranged bundles of mature collagen fibers and spindle–shaped cells, while in others it consisted of newly formed (thin) collagen fibers and stellate-shaped cells. Sometimes there were myxomatous foci, cysts, clusters of osteoclast cells or xanthoma cells, islets of cartilage (fig. 4 b).

By comparing tomograms and the results of pathomorphological studies, we identified AB spurs, which penetrated into healthy bone and in most cases were not determined by radiography, which should be taken into account when radically removing the formation.

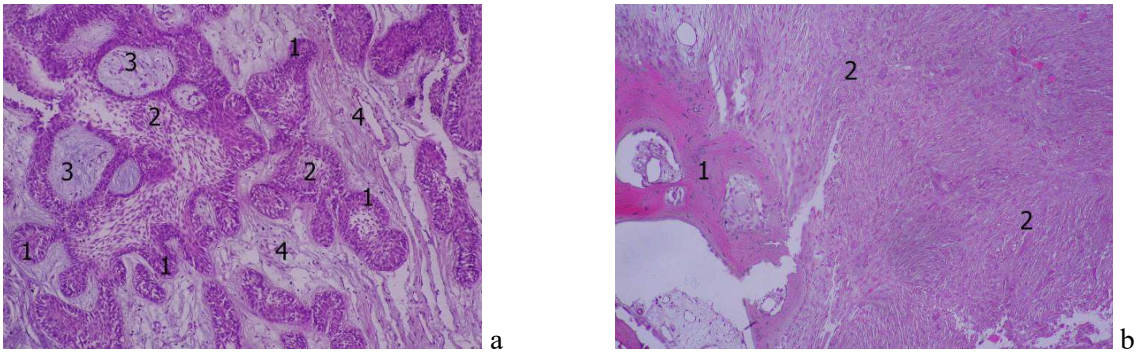

Fig. 4. Microscopic structure of ameloblastoma (A) and fibrous dysplasia (B). Hematoxylin-eosin staining. Objective lens: 10^x, Ocular lens: 7^x. a: 1 – odontogenic epithelium; 2 – polygonal cells resembling the pulp of the enamel organ; 3 – cysts; 4 – connective

tissue stroma. b: 1 – unchanged bone tissue; 2 – fibrous connective tissue with randomly arranged bundles of mature collagen fibers and spindle-shaped cells.

Therefore, ameloblastoma and fibrous dysplasia are quite similar diseases, although they have many differences. Thus, ameloblastomas are more common in 7–16-year-old patients, although they can be found at other ages in both women and men, accounting for up to 7–18 % of all BT and tumor-like formations of the jaws. Being the only tumor that originates directly from the cellular elements of the enamel organs, in almost 94 % of cases it occurs on the lower jaw with a "favorite" localization in the area of its angle and ramus [1, 2 5].

In contrast, fibrous dysplasia in most cases is a defect in bone development in both embryonic and postnatal periods of child development and manifests itself mainly in early childhood (which coincides with our data) with slow progression until puberty. Interestingly, FD occurs mainly in females in all continents, but for some reason the maximum frequency of facial skull lesions is observed in West Africa [3, 8, 9].

Although the true cause of FD is uncertain, most researchers believe that the basis of the disease is a tumor-like process associated with impaired mesenchymal stem cell osteogenic differentiation [5], and depending on the spread of the process, monostotic and polyostotic forms of FD are distinguished [8].

In all our cases, the follicular form of AB was determined, but scientists describe its other varieties: plexiform, acanthomatous, basal cell, granular cell [7].

There are some features of the histological picture and FD of facial bones: the dense component in the foci of dysplasia may be represented by cement-type tissue (cementicle-like structures) [7, 8], but we have not observed such cases.

There are isolated cases of malignancy of AB and FD [5, 8], so we should not forget about cancer vigilance in children [1-3, 9–10].

Conclusions

Ameloblastomas are more common in the lower jaw in 7-16-year-old patients. In contrast, fibrous dysplasia, areas of which in MFA are also localized mostly on the lower jaw, is manifested mainly in early childhood.

Both tumors have a unique clinical and morphological picture, so the final diagnosis should be determined by clinical objective examination, analysis of X-ray and CT or MRI, puncture and incisional biopsy and histological examination of postoperative material. Treatment of patients should be fairly balanced and comprehensive with the involvement of doctors of related specialties in the postoperative period and during rehabilitation.

Prospects for further research lie in the fact that the given material can become a basis for further in-depth scientific and practical researches on comparison of clinical manifestations and immunohistochemical features of ameloblastoma and fibrous dysplasia of different anatomical localization depending on age of patients.

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