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Giant cell tumor of the maxilla - case report and long-term follow-up results

Guz olbrzymiokomórkowy szczęki – opis przypadku i wyniki długoterminowej obserwacji

INTRODUCTION

Giant cell tumors (GCT) constitute approximately 3-7% of bone tumors. In about 70-90% of the cases they involve the end of long bones, however, they occur in the area of the head and neck in 2% of the cases. They are mainly located in the mandible, more rarely in the maxilla, sphenoid bone, ethmoid bone, temporal bone, zygoma and frontal bone. In the years 1991-1996, 107 cases of giant cell tumor occurring in the area of the skull were described [5]. GCT occurs more frequently in children and adolescents. The mean age of GCT patients is 8.2 years. GCT is more frequent in girls than in boys and 2/3 of the cases occur in the mandible. The tumor is connective tissue neoplasm; it develops from nonosteogenic stromal tissue of the bone marrow [2,5].

The clinical examination reveals asymptomatic oedema, tooth migration, root resorption, perforation of cortical plate. The patients may complain about pain, paresthesia, tooth loss [2].

In the histological imaging mononuclear fusiform or oval cells of the stroma and giant cells are visible. Giant cells arise as a result of mononuclear cell fusion and division of cell nuclei without simultaneous division of cytoplasm. Such cells reach considerable size, up to 100 nuclei, and are located quite evenly over the whole tumor. The nuclei shape is round or oval, and they are hyperchromatous. The tumor stroma consists of fibroblasts and rounded cells [5].

Tumor tissue is soft, reddish or red-brown, fragile, easily-bleeding. The cross-section reveals yellowish stains connected with lipid accumulation in histiocytes. Giant cell tumor malignancy grade is difficult to establish in histopathological examination [5].

GCT must be differentiated from giant cell granuloma, brown tumor occurring in hyperparathyroidism, histiocytosis chordoma, aneurysmal bone cysts [2,4,5].

In the diagnosis of GCT computer tomography as well as magnetic resonance imaging are used [5].

C a s e R e p o r t. 17-year-old patient presented at the Outpatient Department of the Oral Surgery Clinic of Medical University in Lublin with slowly growing, painless lesion on the maxilla alveolar process, which she noticed a year before, as reported in the patient's history.

Clinical extraoral examination revealed a tumor with a diameter of 3 cm which involved teeth 13-16 in the oral vestibule, on the maxilla alveolar process, on the right side, it was covered with mucosa with intensive vascularity, slightly cyanosed. On the day of the examination panoramic tomography was performed and showed clearly separated osteolytic loss with a diameter of about 8mm in the region of teeth 13-14. The patient was sent for oncological consultation, where samples were taken for histopathological examination. Due to the character of the tumor, which suggested the possibility of brown tumor, the patient was examined for hyperparathyroidism. However, in the Endocrynology Clinic of Children's University Hospital in Lublin the diagnosis of hyperparathyroidism was excluded. After conducting blood cell count, coagulation tests (the results were within normal range) operation was performed with local anesthesia. The mucoperiosteal flap was incised, detached and then the tumor was uncovered. The tumor was clearly separated from the surrounding tissues; it lay superficially on the bone of the maxilla alveolar process. It was brown in color, very fragile, however, easily separable from its base. The whole tumor was removed within the borders of macroscopically healthy tissues. The bone edges were milled. The tumor caused partial destruction of the maxilla bundle bone in the section 13-16. Interdental spaces and the area of denuded roots of teeth 13-16 were thoroughly cleaned. The wound was sutured. The sample material was sent for histopathological examination. The result: tumor gigantocellularis.

The patient still remains under clinical monitoring and 2 years after the surgery there are no clinical or radiological symptoms of the disease recurrence.

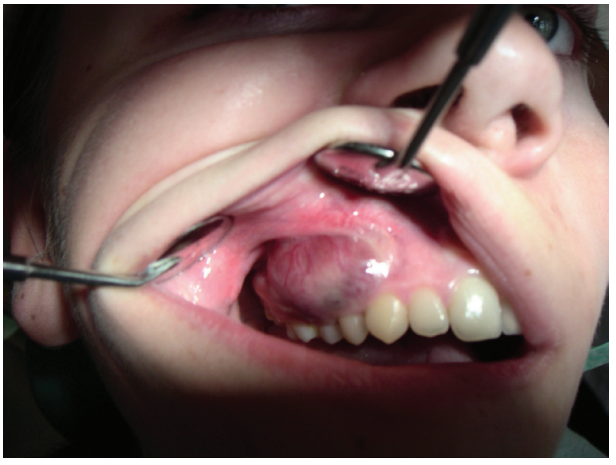
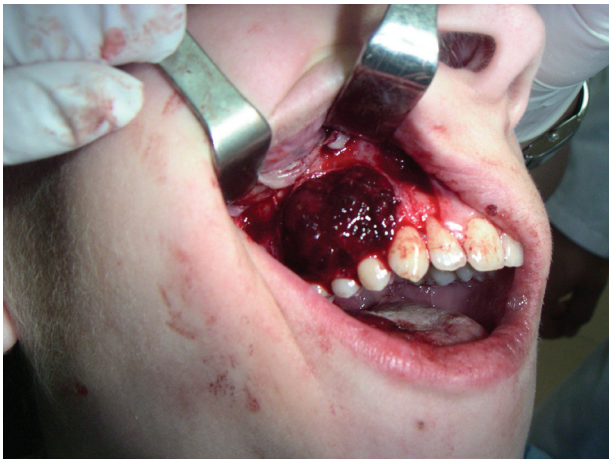
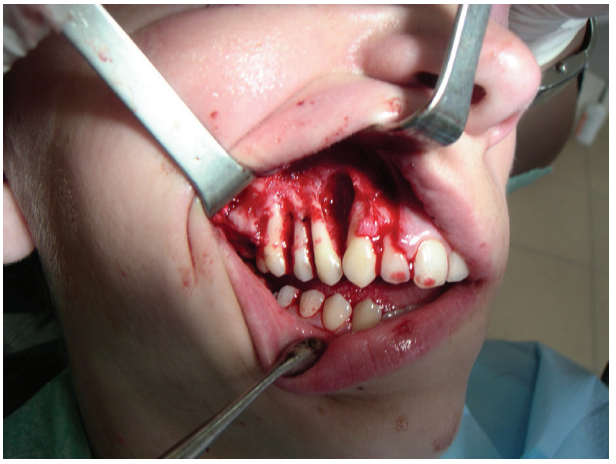


Figure 1. Intraoral picture demonstrating tumor located on maxilla alveolar process





Figures 2, 3. Picture taken during surgery. Tumor destroyed bundle bone and denuded tooth roots

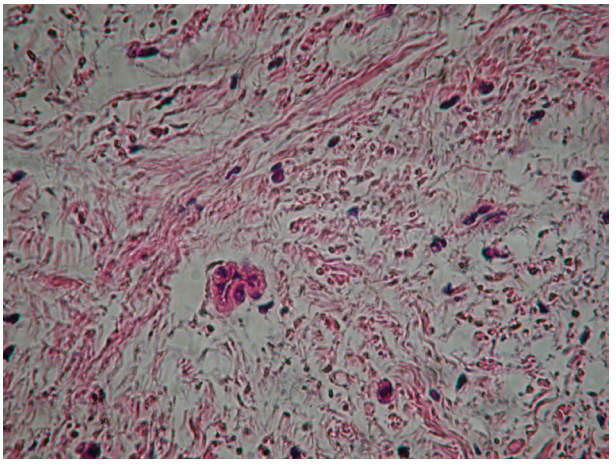


Figure 4. Giant cells with large number of nuclei H plus E staining, magnification of 200

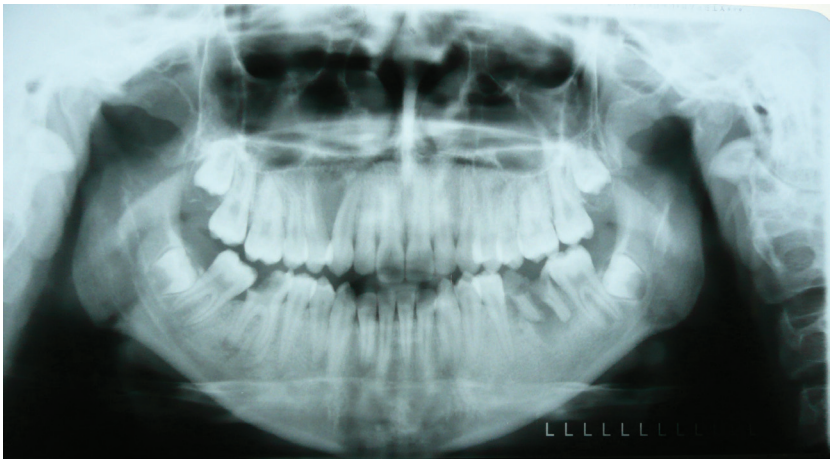


Figure 5. Panoramic radiograph demonstrated clearly separated osteolytic cavity with a diameter of about 8 mm in the area of teeth 13-14

DISCUSSION

Giant cell tumor is defined both as malignant and benign lesion. Malignant type of tumor is large, grows rapidly; patients complain of pain, tooth loss or displacement, bleeding. The radiological examination demonstrates root resorption of teeth located in the lesion and perforation of the bundle bone. The recurrences occur in 70%. Benign type of giant cell tumor does not have any of the features described above. It is relevant that both malignant and benign tumors often have the same histological structure which is characterized by great vascularity [2].

There were attempts of pharmacological treatment of GCT. Assuming inflammatory etiology of the tumor corticotherapy was applied. There were conducted studies on the use of calcitonin which inhibits bone resorption. Leonard Kaban et al[1] described the case of giant cell tumor of the mandible in 5-year-old child where interferon alfa-2a was used as an adjuvant therapy after surgical treatment. Interferon alfa-2a is angiogenesis inhibitor. It inhibits the production of mRNA and proteins of the two well-known angiogenetic agents, bFGF and interleukin-8. The urine analysis showed largely elevated concentration of bFGF. A year after interferon administration the post-surgery recurring tumor disappeared, the mandibular bone regenerated and the level of bFGF in urine returned to the norm. No complications were observed during the therapy and it was recommended to continue it for the following years [2].

The methods of giant cell tumor treatment in the area of the head and neck bones are controversial. Standard treatment ranges from surgical curettage to wide resection [1-3,5]. Instead of wide bone resection in GCT patients, tumor extirpation followed by interferon therapy is conducted more and more frequently. Angiogenetic therapy combined with curettage of the lesion is a promising treatment method for aggressive giant cell tumors [1,2]. Considering tumor location and the consequences of radical surgical treatment of seventeen-year-old patient the surgery was restricted to tumor excision within the borders of macroscopically healthy tissues. The patient is being

monitored by the local clinic (check-up every 6 months). In the case of recurrent disease implementing of angiogenetic therapy with interferon alfa-2a should be considered.

CONCLUSIONS

A patient with giant cell tumor requires detailed clinical, radiological and differential diagnosis in order to choose treatment method that will be appropriate for a particular case.

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SUMMARY

Giant cell tumor is a rare disease of the head and neck region. Predominantly giant cell tumor involves the end of long bones. Sometimes it occurs in the head and neck areas, predominantly in mandible and maxilla. The tumor arises from the nonosteogenic stromal cells of the bone marrow. Giant cell tumors are defined as either aggressive or nonaggressive lesions. An aggressive type tumor is large, rapidly growing, loose and displaced teeth. Nonaggressive type giant cell tumors do not exhibit these signs. Surgery is the most accepted treatment of giant cell tumor. Sometimes surgery can be performed in association with new approaches, in an attempt to avoid recurrence. Surgical treatment can be followed by local injection of steroids or antiangiogenic therapy using interferon alfa-2a.

We report a 17-year-old girl with a large, slowly growing giant cell tumor of the maxilla. The patient was treated with surgical excision.

Keyword: giant cell tumor, tumor gigantocellularis, surgical treatment

STRESZCZENIE

Guz olbrzymiokomórkowy jest rzadkim schorzeniem w obszarze głowy i szyi. Najczęściej guz olbrzymiokomórkowy spotykany jest w nasadach kości długich. Rzadko pojawia się w obszarze głowy i szyi, głównie okolicy żuchwy i szczęki. Nowotwór powstaje z komórek podścieliska w szpiku kostnym.

Guzy olbrzymiokomórkowe są definiowane jako zmiany agresywne lub nieagresywne. Agresywny typ nowotworu jest najczęściej dużą, dynamicznie rozwijającą się zmianą, powodującą rozchwianie i przemieszczenia zębów. Nieagresywne guzy olbrzymiokomórkowe nie wykazują tych cech. Leczenie chirurgiczne jest najbardziej akceptowaną metodą guzów olbrzymiokomórkowych. Leczenie chirurgiczne może być stosowane przy użyciu miejscowych iniekcji sterydów lub terapii antyangiogennej za pomocą interferonu alfa-2a.

Przedstawiamy przypadek 17-letniej dziewczynki z guzem olbrzymiokomórkowym składającym się z wolno rosnących komórek. Pacjent był leczony poprzez chirurgiczne usunięcie zmiany.

Słowa kluczowe: guz olbrzymiokomórkowy, tumor gigantocellularis, leczenie chirurgiczne