PEDIATRIC OSTEOBLASTOMA OF C3 LATERAL MASS: CASE REPORT AND LITERATURE REVIEW

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The paper presents a clinical case report. Level of evidence — V. Surgical treatment of osteoblastoma of the lateral mass of C3 vertebra was performed in a 6-year-old child. Follow-up period was 9 months after radical operation including tumor en block resection and local screw fixation of C2—C3 vertebrae. The principles of differential diagnosis of small-sized osteoblastoma (1.5 cm) with nonspecific clinical and radiological symptoms are described.
Key Words: benign bone-forming tumors, osteoblastoma, cervical spine tumors, cervical spine screw fixation in children.

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Primary bone tumors of the spine account for 0.4 % of all tumors located at various sites and 4.2 % of all spinal tumors [1]. Certain types of primary spinal tumors are characteristic of each age period (Table 1) [2, 9, 20]. It should be noted that various types of primary bone tumors involve certain vertebral segments and vertebral zones. For example, osteoid osteoma and osteoblastoma usually grow from of the posterior structures of the cervical or lumbar vertebrae and rarely from the thoracic ones [20]. Osteoid osteoma and osteoblastoma are similar in histological structure and consist of osteoblasts, producing osteoid and reticulofibrous membranous bone tissue. In some cases, differential diagnosis of these tumors is a challenging problem.

Osteoblastomas were described in less than 1 % of patients with benign spinal tumors, and osteoid osteomas were described approximately in 9 % of patients [20]. The primary differential diagnosis is based on the tumor size: osteoblastoma reaches a large size (more than 2 cm in diameter), while osteoid osteomas are usually smaller than 1 cm. Tumors sized 1 to 2 cm in diameter cannot always be clearly differentiated based solely on their size [20].

Hereafter we report a clinical case of a 6-year-old patient with a difficult-to-differentiate osteoblastoma of C3 lateral mass sized 1.5 cm.

Clinical presentation. A 6-year-old patient complained of neck pain radiating to the occipital region. The child's parents observed periodic episodes of torticollis, for which the child received an outpatient treatment since the age of 3 years. Neck pain occurred daily and did not depend on the time of day. During pain attacks, neck movements and its palpation are moderately painful. There were no focal neurological symptoms during pain attacks or any other time. Pain was relieved by administration of non-steroidal anti-inflammatory drugs *(NSAIDs)*, and the boy had *NSAID* dependence for the last six months. There were no concomitant diseases, family history of cancer diseases was not burdened.

Thus, neurologically intact patient (grade E on the Frankel scale) preoperatively had Lansky score of 70.

Laboratory tests sowed mild leukocytosis.

X-ray examination. X-ray examination of the cervical spine (Fig. 1) showed slight nodosity at the left intervertebral

Table 1
Primary spinal tumors in children, depending on age

Age, years	Benign tumors	Malignant tumors
<5	Eosinophilic granuloma	—
5 to 10	Eosinophilic granuloma, osteoblastoma, osteoid osteoma, aneurismal bone cyst	Osteosarcoma, Ewing's sarcoma
>10	Osteochondroma, aneurysmal bone cyst, osteoid osteoma	Osteosarcoma, Ewing's sarcoma

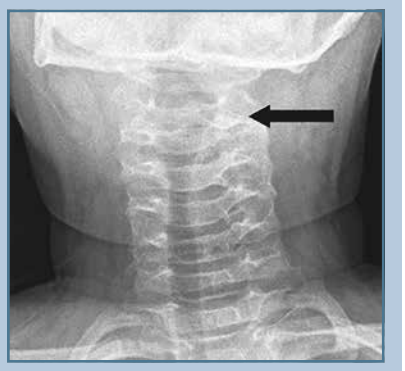

Fig. 1
X-ray image of the cervical spine of the 6-year-old patient (frontal view): the arrow indicates the tumor position

joint C2—C3, and obliterated joint space line.

SCT of the cervical spine (Fig. 2) showed a tumor sized 16 x 15 x 14 mm and located near to the superior left articular process of C3 vertebra (zones 2, 3 and 4 according to Tomita; sectors 1, 2, 3 and zones B, C, D according to Weinstein-Boriani-Biagini classification). According Tomita classification, this tumor belongs to B4 type. Its SINS (spinal instability scale) score is 5 points (the spine is stable). The tumor increased by 20 % compared to CT of the cervical spine 6 months before this examination (13 x 13 x 12 mm). CT showed that the tumor had homogenous structure with peripheral foci of increased signal with hypodense signal from the center of the tumor; there was no pronounced perifocal sclerosis. MRI of the cervical spine showed expansively growing local inhomogeneous neoplasm (isodense and hypodense mass in T1-weighted images, hypodense and hyperdense mass in T2-weighted images). There were no signs of bone marrow edema. The tumor did not accumulate contrast in gadolinium-contrasted studies.

Persistent pain syndrome (for more than three years) with periodic transient episodes of torticollis was the main indication for surgery in this patient. Secondary indications included dependence on analgesics and signs of local aggressive tumor growth accompanied by destruction of the internal endplate of the vertebral canal (Fig. 2) and the walls of the intervertebral foramen C3–C4.

Operation. The patient underwent a one-step operation: posterior approach to the C2–C3 vertebrae, total resection of the tumor followed by treatment of the adjacent articular surface of the inferior articular process of C2 vertebra. Macroscopically, the tumor was friable and moderately bleeding. Polyaxial screws were placed on both sides into the *pars interarticularis* of C2 vertebra and into the lateral masses of C4 vertebra, modeled implant made of treated allobone was laid in place of the resected tumor. Compression maneuver was made on both sides to fix the implant.

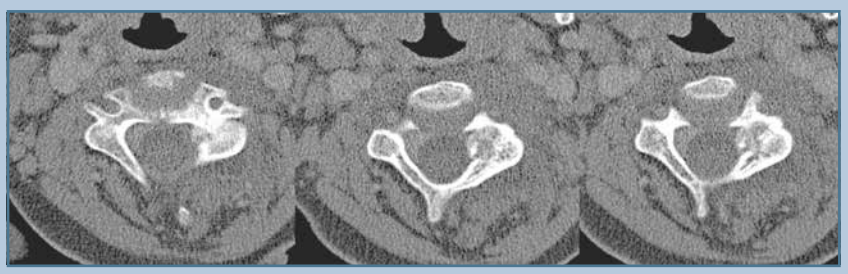

Fig. 2
Preoperative CT of the cervical spine of the 6-year-old patient showing the tumor of C3 lateral mass: homogeneous structure with hypodense center

Histological examination. Along with typical tissue fragments of bone, cartilage, and fibrous structures, microscopic visualization of microslides showed fragments of osteogenic tumor mass represented by a network of intertwining thin, predominantly osteoid, trabeculae characterized by uneven weak mineralization, surrounded by friable fibrous and fibroreticular tissue with high content of cell elements and pronounced vascularization (Fig. 3).

There are transition regions between the tumor mass and surrounding tissues. Fig. 4 shows tumor adjunction to the cartilage (white arrow); there are trabeculae consisting of coarse-fibered bone tissue in the narrow transition zone between the cartilage and tumorous trabecular network (black arrow).

The portions of intact bone adjacent to the tumor have spongy patches, somewhat rarified, with thinned trabeculae and expanded intertrabecular spaces.

Differential diagnosis between osteoblastoma and osteoid osteoma was based on the fact that tumor foci were larger than 1 cm and there was no area of extensive perifocal sclerosis characteristic of osteoid osteoma.

There was no evidence of malignant growth in the form of cell polymorphism and atypia and increased mitotic representation, including pathological one. Based on these findings, we excluded malignant variants of osteogenic tumors during the differential diagnosis. There were also no symptoms of inflammatory lesions.

Treatment outcome. Postoperative radiographs and CT: the tumor was totally removed, position of metal structure is satisfactory (Fig 5, 6). Immediate and late postoperative period was uneventful. Neurological status remained at the preoperative level (level E on the Frankel scale), Lansky score increased to 90 points. Pain was completely eliminated, there were no torticollis episodes, control CT scan 6 months after surgery showed satisfactory spondylosynthesis; there were no evidence of tumor recurrence.

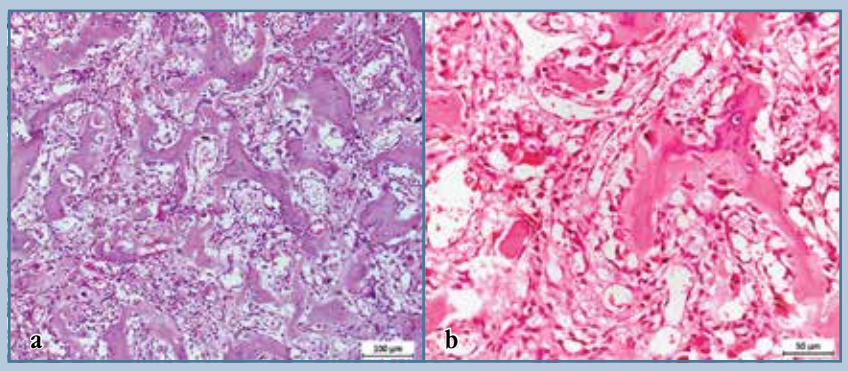

Fig. 3
Trabecular network at high magnification: **a** — magnification x250; **b** — x500; H & E stain

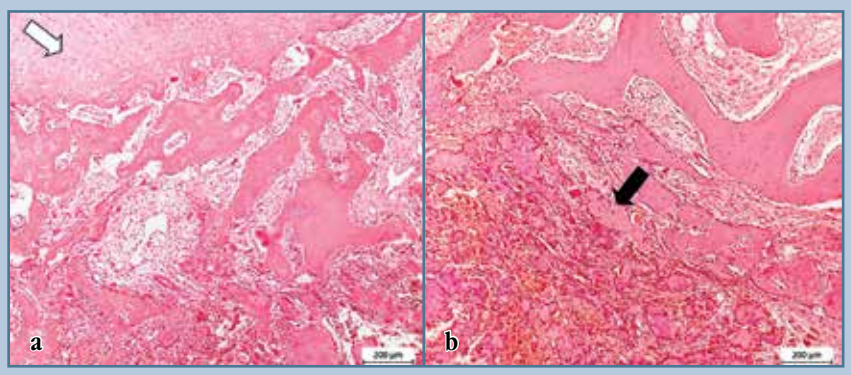

Fig. 4
Transition areas between the tumor and cartilage **(a)** between the tumor trabecular network and mature bone trabeculae **(b)**; tumor trabecular network is located in the lower portions of the pictures; magnification x250; H & E stain

Discussion

In some cases, the differential diagnosis of osteoid osteoma and osteoblastoma is quite challenging and is based on the analysis of clinical and radiological presentation and morphological characteristics (Table 2).

The main clinical symptoms of cervical spine osteoblastoma include neck pain, movement restrictions, torticollis, scoliosis, and, more rarely, signs of myelopathy [5, 13, 18, 25]. Macroscopically, osteoblastoma is represented by friable bleeding tissue with hemorrhage sites, well circumscribed from the surrounding bone tissue. It grows from the cancellous tissue of the posterior vertebral elements, the vertebral body is expansively involved from the pedicle of the vertebral arch [15, 16, 20, 28]. The cases of osteoblastoma malignization were reported in 12–25 % of patients [16, 19, 20,27]. CT image of osteoblastoma looks like frosted glass with hypodense signal in the center [20].

Osteoid osteoma often presents with local pain at the tumor site and sometimes radicular pain. Typically, the pain is intense, intermittent, increases during activation, and is associated with movements. Nocturnal pain, decreasing after intake of aspirin, is one of its typical signs. Macroscopically, osteoid osteoma is a dense, sclerosed tumor, which may have a granulomatous component in its center [20]. CT image of osteoid osteoma shows a uniform hyperdense signal at the periphery due sclerotic area around it and hyperdense heterogeneous signal inside due to sclerotic foci; there are no signs of bone destruction. T2-weighted MRI of osteoid osteoma showed hyperintensive signal and bone marrow edema at the tumor site.

The final diagnosis in this patient was based on the following signs: tumor larger than 1 cm, frosted-glass-like appearance on CT with hypodense center, no signs of bone marrow edema, growth from the cancellous tissue of C3 lateral mass, locally invasive growth towards the intervertebral foramina, spinal canal with destruction of the vertebral cortical bone, characteristic structure after resection (friable, bleeding), no pronounced sclerotic area around the tumor as shown by microscopic examination.

Selection of the type of surgery for spinal tumors depends on the following factors: the type and size of the tumor, location of the tumor with respect to the spinal cord, neural roots, and vertebral arteries [4], individual characteristics of patient, including age, spinal balance [29], and aggressive growth signs [21]. The following options of surgical treatment for cervical osteoblastomas in children have been described: resection of the vertebral body tumor from the anterior approach without fusion [7, 28], resection of the vertebral tumor from the combined (anterior and posterior) approach with fusion using autobone and wire [26], resection through the single-stage or two-stage combined approach (anterior and posterior) accompanied by instrumented fixation [6, 10, 13, 23, 29], tumor resection from the posterior approach with non-instrumented fusion [14, 17], tumor resection from the posterior approach with instrumented fixation [3, 6, 8, 22].

In some cases, tumor relapses necessitating reoperations have been reported [6, 7, 26]. En bloc resection of osteoblastomas is preferred over excochleation followed by irradiation, since this operation is associated with lower number of

Table 2
Differential diagnosis of osteoblastoma and osteoid osteoma

Tumor	Symptoms	Imaging data	Growth characteristics	Histologic characteristics	Treatment
Osteoid osteoma	Nocturnal pain, good effect of NSAIDs	CT: sclerosis, hyperdensity; T2-weighted MRI: hyperintensity, smaller than 1 cm	Benign	Dense, sclerosed, granulomatosis site at the center	Operation, follow-up, radiofrequency ablation
Osteoblastoma	Pain, weak effect of NSAIDs	CT: frosted-glass-like appearance, hypodense center, larger than 2 cm	Benign, locally invasive	Friable, bleeding	Operation

relapses [6]. Non-instrumented fusion and fusion with additional sublaminar fixation with wire does not provide reliable fixation and results in higher incidence of pseudoarthrosis and complications [8, 11, 24]. Safety and reliability of the instrumented screw fixation of the cervical spine in children was proved by morphological and experimental studies and for this reason, it has been widely used in practice during the last 5–7 years [11, 12].

In the reported case, the patient underwent en bloc resection of the osteoblastoma within the healthy bone. Since the lateral mass of C3 vertebrae was completely resected, the stabilizing step was carried out, i.e. local posterior instrumented fixation of C2–C3 vertebrae with a screw system. We preferred the posterior instrumented spinal fusion through a single approach. Furthermore, the tumor did not spread into the body of C3 vertebra and therefore no additional anterior fixation was required.

Conclusion

1. In some cases, differential diagnosis of osteoblastomas and osteoid osteomas is a challenging problem. It is based on the analysis of clinical and radiological patterns and morphological characteristics.

2. Surgical treatment for cervical spine osteoblastoma is indicated in the case of symptomatic course. Additional indications for surgery include tumor growth and local aggressive impact of the tumors on adjacent tissues.

3. En bloc resection of osteoblastoma within the healthy bone is preferred over excochleation.

4. Local posterior instrumented fixation using a screw system is a method of choice to stabilize the spine in the case of the tumor of the posterolateral structures of cervical vertebrae.

The study was not sponsored. The authors declare no conflict of interests.

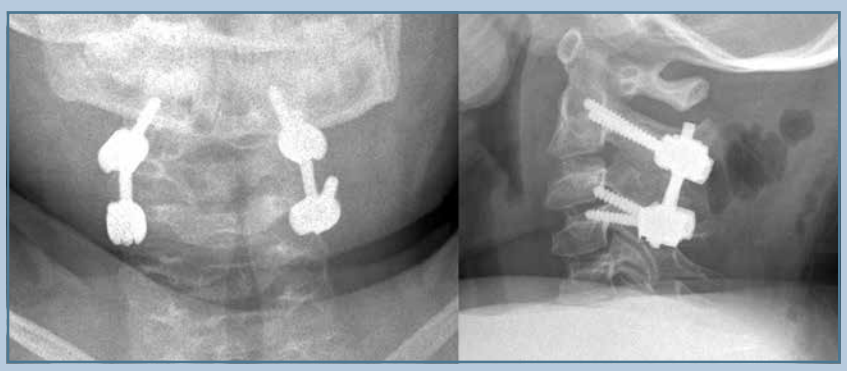

Fig. 5
Postoperative X-ray images of the 6-year-old patient (frontal and lateral projections)

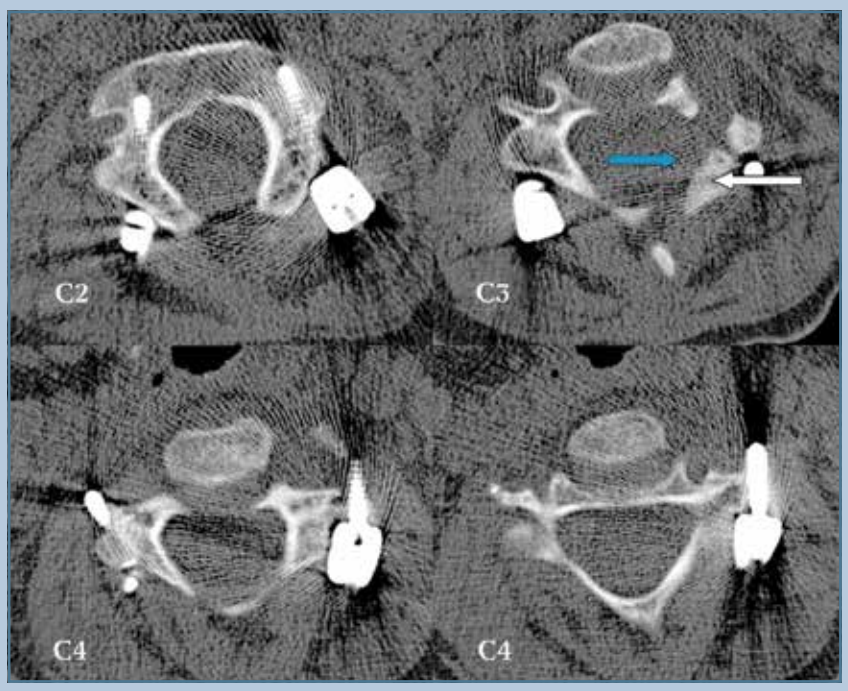

Fig. 6
CT of C2–C4 vertebrae: screws are inserted into the *pars interarticularis* of C2 on both sides and into C4 lateral mass from both sides; white arrow shows allograft, blue arrow shows lateral mass defect after tumor resection

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