



OSTEOID OSTEOMA AND OSTEOBLASTOMA OF THE SPINE IN CHILDREN: Features of intravertebral zoning and results of surgical treatment With a follow-up of at least one year

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Objective. To analyze the features of zonal localization of osteoid osteoma and osteoblastoma and the results of their surgical treatment in pediatric patients.

Material and Methods. The data of 41 children aged 4 to 17 years who underwent surgery for osteoid osteoma (29) and osteoblastoma (12) of the vertebrae were compared retrospectively within a monocentric cohort. The features of the tumor zonal location and the results of surgical treatment were assessed taking into account the risk of relapse and the need for instrumental stabilization.

Results. Osteoid osteoma and osteoblastoma are localized in the posterior structures of the vertebrae (sectors 2-4 and 9-11 according to the Weinstein – Boriani – Biagini classification) in 93 and 75 %, respectively, with a predominance of right-sided localization of osteoid osteoma (sector 9-11). Osteoid osteomas are predominantly located in zones B and C, while the spread of osteoblastomas to zone D indicates a more aggressive course with the possibility of developing neurological symptoms in 50 % of cases. Complete removal of tumors in the volume of marginal resection due to unilateral localization in the posterior elements of the vertebra is safe and allows, in the absence of intraoperative destabilization, to complete the operation without additional instrumental fixation; local bone fusion is sufficient to prevent local instability.

Conclusion. Osteoid osteoma and osteoblastoma of the vertebrae in children differ in localization, zoning according to the Weinstein – Boriani – Biagini classification, and clinical aggressiveness of the course. Features of intraorgan zoning of tumors with proximity to nerve structures limit the use of ablation technologies, while wide marginal resection provides a full-fledged cure. The need for instrumental stabilization of the spine depends on the volume of resection.

Key Words: spine; tumor; osteoid osteoma of the vertebra; osteoblastoma of the vertebra; children.

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Primary spinal tumors in children represent 10 % of primary neoplasms of the musculoskeletal system and less than 5 % of all spinal tumors [1-3], and approximately 70 % of these tumors are benign [4]. Osteoid osteoma (ICD-O classification, 9191/0) and osteoblastoma (ICD-O classification, 9200/1) hold a special place among them: the incidence of spinal lesions is rated in the range of 3-25 % for osteoid osteoma and 30-46 % for osteoblastoma [4–10].

Both tumors are osteogenic, characterized by a similar morphological pattern and twice as common in male patients, with a peak incidence in the second or third decade of life [6–9]. Some authors [10, 11] believe that both tumors are rarely present with neurological disorders. The differentiating feature is the size of tumors: the diameter of osteoid osteoma foci does not exceed 2.0 cm, while space-occupying lesions are considered to be osteoblastomas [12]. In addition to differences in size, however, osteoblastoma is a locally aggressive tumor with replicative potential, the possibility of postoperative recurrence, and malignant transformation. The main complaint of all the patients is pain syndrome. At the same time, pain syndrome in osteoid osteoma is usually transient, increases at night, and is relieved by taking salicylates or other non-steroidal anti-inflammatory drugs (NSAIDs) [6]. Unlike osteoid osteoma, the pain syndrome in osteoblastoma is less pronounced; nocturnal pain is less severe, but NSAIDs are effective in only 7 % of cases [6, 13–16]. The histological similarity of osteoid osteoma and osteoblastoma, on the one hand, and the different clinical potential, on the other hand, motivated us to analyze the peculiarities of these tumors.

The objective is to analyze the features of zonal localization of osteoid osteoma and osteoblastoma based on the Weinstein, Boriani, Biagini (WBB) classification [17] and the results of their surgical treatment in pediatric patients.

Design: a monocentric retrospective cohort clinical study.

Material and Methods

In 1994–2023, 29 patients were operated on for histologically distinctive osteoid osteoma and 12 patients for vertebral osteoblastoma at the Clinic of Pediatric Surgery and Orthopedics, St. Petersburg Research Institute of Phthisiopulmonology. The subject matter of the analysis was general data (age, gender, tumor localization level), as well as the main clinical and imaging manifestations of tumors, evaluated according to the following criteria:

• neurological impairment according to the Frankel scale of the ASIA standard;

• pain syndrome according to 10-point pain rating scale (VAS);

• extent of the pathological process at the level of the affected vertebra according to CT and MRI data in accordance with the WBB zonal-sectoral classification (Fig. 1) with an addition for tumors of the cervical spine [18].

According to the WBB classification, on the axial section of the vertebra, sectors 2-4 and 9-11 correspond to the pedicle, arch, articular and transverse processes; sectors 12-1 correspond to the spinous process; and sectors 5-8 correspond to the vertebral body. There are also 5 concentric zones (A to E) from periphery to center, with zone A corresponding to perivertebral spread, zones B and C to peripheral and central intraorganic (intraosseous) sections, zones D and E to epidural and intradural position, respectively, and zone F, evaluated only for the cervical spine, to involvement of the spinal artery canal.

All patients underwent preoperative CT, MRI, and radioisotope scanning of the spine. The type of surgery, complications, and recurrences were evaluated when analyzing the outcomes. The clinical features of the disease and its imaging manifestations were the grounds to determine the variant of surgical strategy. Transcutaneous trephine biopsy under C-arm control was performed to verify the process in patients with atypical imaging findings (presence of soft tissue component, dural sac compression, pronounced osteosclerosis). Postoperative follow-up was performed 3, 6, and 12 months after surgery using a distant survey and CT control and then once a year for the first three years. In case of recurrence of complaints, an intravenous contrast CT and MRI were performed in the event of neurological disorders.

Statistical analysis. Statistical analysis was performed using SPSS Statistics software, version 26. Each study sample was assessed for conformity to a normal distribution using the Shapiro-Wilk test. The age distribution of patients was described using median (Me) and interquartile range [IQR] because of non-normal distribution. The one-sample chi-square test was used to evaluate the difference between the distribution of patients in the studied samples and the standard distribution. Differences in distributions were accepted as statistically significant at p < 0.05.

Results

Data from 29 patients with osteoid osteoma (19 boys and 10 girls; 66 % and 34 %, respectively) and 12 patients with osteoblastoma (7 boys and 5 girls; 58 % and 42%, respectively) who underwent surgery at the hospital in 1994–2023 were included in the study. The median age of patients in the total sample was 14.0 [11.0-16.0] years. The minimum age at the time of surgery was 4 years, and the maximum was 17 years, including 3 children under 7 years old, 12 children of 8 to 12 years old with a median of 11.0 [10.0-12.0] years, and 26 children over 12 years old with a median of 15.0 [14.0-17.0] years. The postoperative follow-up period was on mean more than 8 years.

The age structure of the sample proves that the occurrence of both tumors in children is typical for children, mainly in the second decade of life, with only osteoid osteomas being diagnosed in preschool patients. The majority (69 %) of osteoid osteoma cases were recorded in the lumbar and sacral spine, while cervical and thoracic vertebrae predominate in the structure of the level distribution of osteoblastoma (75 %; Table 1).

At the time of admission, the main symptom in all patients was vertebrogenic pain, which corresponded to 3 points on the VAS in 38 % of cases. In patients with osteoid osteoma, pain was completely resolved by taking NSAIDs, while narcotic analgesics were additionally administered at the outpatient stage in two cases of osteoblastoma because of the ineffectiveness of NSAIDs. Secondary spinal deformity in the form of antalgic scoliosis (5 patients; 17 %) and torticollis (3; 10 %) was found in 8 (28 %) cases of osteoid osteoma in children. In osteoblastoma, secondary spinal deformity was observed in two (17%) cases in the form of antalgic scoliosis and localized angular kyphosis and torticollis.

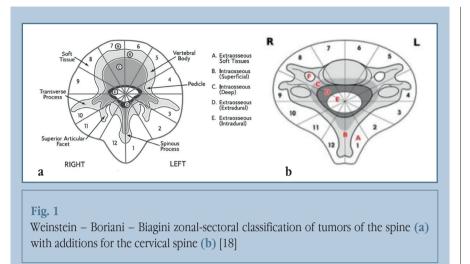
Four (14 %) patients with osteoid osteoma experienced radicular pain syndrome, in one case followed by monoparesis (Frankel type D with S1 lesion level). In osteoblastoma, neurological disorders were found in 4 (33 %) patients, including three cases of type D and one of type C.

According to the WBB classification, osteoid osteoma was localized in sectors 2–4 and 9–11 in 27 (93 %) of 29 cases, with only in 2 (6.9 %) cases in the vertebral body (sectors 5–8). Considering lesion side, the distribution of patients with osteoid osteoma was found to be statistically significantly different from the uniform distribution (chi-square test; p < 0.01): according to the WBB classification, right-sided tumor location in sector 9–11 (20; 69 %) and zone C (55 %) was significantly predominant; chi-square test; p < 0.01 (Table 2).

Osteoblastomas were also localized in sectors 2–4 and 9–11 in 9 (75 %) of 12 cases, and in 3 (25 %) cases – in sectors 5–8 (vertebral body) with spread to zone D in half of the cases (Table 2).

No lesions of the spinous process (sector 12–1) were identified in any observation.

The indication for transcutaneous trephine biopsy to exclude a malig-



nant process was an unusual imaging pattern in two patients (one case of osteoid osteoma and one case of osteoblastoma). In the first case, there was pronounced osteosclerosis spreading from the arch to the vertebral body; in the second case, there was a rapidly progressive soft tissue component with dural sac compression. The pathological exclusion was verified by preoperative biopsy in a patient with osteoblastoma.

Considering the location of the tumors, the vast majority of patients (97 % with osteoid osteoma; 67 % with osteoblastoma) underwent tumors removal from the posterior approach. In one case, a C2 body tumor, initially treated as an osteoblastoma but morphologically diagnosed as an osteoid

osteoma, was removed via transoral approach (Fig. 2).

According to current guidelines, a benign spinal tumor must be resected completely during surgical treatment; its intralesional resection is considered sufficient [19, 20]. This is absolutely justified, as the extension of the resection, which is excessive for total, including en bloc resections of malignant tumors, seems unnecessary for benign tumors in most cases. The zones of reactive sclerosis and microcirculatory edema in the vertebrae, which do not require resection, may spread much wider than the original tumor nidus.

According to postoperative CT evaluation of the volume of procedures, surgeries were divided into 3 types: wide marginal resection, when the plane of section runs along the periphery of the reactive (sclerotic) zone through normal tissue;

 marginal resection, where the plane of dissection runs along the reactive zone of sclerosis;

- intralesional resection, when morcellation with preservation of the margins of the lesion is performed.

The treatment strategy is given in Table 3. The vast majority of patients, 15 (52%) with osteoid osteoma and 10 (83%) with osteoblastoma, underwent wide marginal resection. Accordingly, marginal resection was performed in 10 (35%) patients with osteoid osteoma; this type of surgery was not performed in patients with osteoblastoma; intralesional resection of osteoid osteoma accounted for 4 (14%) surgeries, and that of osteoblastoma – 2 (17%).

Tumor resection with preservation of intervertebral joints was performed in 17 (41%) patients, unilateral surgery with posterior local fusion – in 16 (39%), and additional instrumentation – in 8 (20%) patients.

Osteoid osteoma was resected without additional stabilization of the spine in 14 (48 %) cases; it was followed by posterior local osteoplastic autofusion in 13 (45 %) cases; posterior instrumentation was required only in 2 (7 %) cases of wide hemilaminectomy to stabilize the spine.

In patients with osteoblastoma, 3 (25 %) procedures were limited to tumor

Table 1

Distribution of patients with vertebral osteoid osteoma and osteoblastoma by age, gender and tumor location, n (%)

Indicator	Osteoid osteoma ($n = 29$)		Osteoblastoma ($n = 12$)	
				,
Age, years	Me [IQR]	min-max	Me [IQR]	min-max
	13.0 [11.0-16.0]	4-17	14.0 [13.0-15.0]	10-17
Gender				
Female	10 (34.5)	p = 0.09	5 (41.7)	p = 0.564
Male	19 (65.5)		7 (58.3)	
Tumor location				
Cervical spine	6 (20.7)	p = 0.188	5 (41.7)	p = 0.343
Thoracic spine	3 (10.3)		4 (33.3)	
Lumbar spine	10 (34.5)		2 (16.7)	
Sacrum	10 (34.5)		1 (8.3)	

Table 2

Distribution of patients with vertebral osteoid osteoma and osteoblastoma according to the WBB classification, n (%)

Indicator	Osteoid osteoma ($n = 29$)		Osteoblastoma ($n = 12$)	
Tumor location by sect	ors			
2-4	8 (27.6)	p < 0.01*	2 (16.7)	p = 0.174
9-11	19 (65.5)		7 (58.3)	
12-1	-		-	
5-8	2 (6.9)		3 (25.0)	
Tumor location by zone	es			
А	-	p < 0.01*	-	p = 0.127
В	5 (17.2)		2 (16.7)	
С	16 (55.2)		2 (16.7)	
D	6 (20.7)		6 (50.0)	
Е	-		1 (8.3)	
F	2 (6.9)		1 (8.3)	
' Differences in distribu	tions are statistically significant ($p < 0.05$).			

resection only, while a variety of spinal stabilization options were performed in 9 (75 %) cases: posterior local osteoplastic fusion in 3 (25 %) cases, posterior instrumentation in 2 (17 %) cases, the surgery was added with anterior stabilization in 1 (8 %) case, anterior and posterior instrumentation after 360-degree reconstruction and decompression surgery in 3 (25 %) cases.

In three cases of spinal artery canal involvement, there was 1 case of planned (suspected) bleeding in a patient with osteoblastoma of the C3 vertebral arch that was treated with local hemostasis.

Analyzing the long-term outcomes followed for a mean of 8 years (min -2, max -12 years), no clinical or radiological signs of spinal instability were observed in any case. Therefore, unilateral surgery in the absence of obvious local intraoperative destabilization can be completed without instrumentation, and local osseous fusion in such cases is sufficient to prevent instability.

The greatest attention was given to evaluating the causes for local tumor recurrences noted in three patients who underwent a total of 5 repeated surgeries. Intralesional resections were initially performed in all cases. One of these observation is of particular interest for several reasons. Postoperative recurrence was noted twice within 5 months. After the first and second surgery, the postoperative diagnosis was twice reported by pathologists as osteoid osteoma, which was also consistent with the clinical and radiological pattern of the disease. After the second recurrence, the tumor was treated as a possible osteoblastoma despite the previous findings, and a wide marginal resection with monolateral monosegmental posterior instrumentation of the spine was performed in revision surgery. The histological report also confirms osteoblastoma (Fig. 3).

Discussion

In 1932, Jaffe and Mayer mentioned a formation of the metacarpal bone that they called "osteoblastic osteoid tissueforming tumor," and 3 years later, Jaffe first introduced the term "osteoid osteoma" to describe an osteoblastic process originally seen as inflammatory. It is typical that Virchow reported a similar process in 1863 and Bergstrand in 1930. It was only in 1954 that Dahlin and Johnson mentioned osteoma, which they described as "giant osteoid osteoma" [7], and in 1956 Jaffe proposed the term "benign osteoblastoma" for this tumor [20], which was entered into the classification of bone tumors published by him and Lichtenstein in the same year [7].

Up to 25 % of osteoid osteomas and 46 % of osteoblastomas are located in the

spine (including 70 to 100% of tumors in the posterior vertebral bodies), accounting for up to 10 % and 1 % of all benign spinal tumors, respectively [13, 21].

While osteoid osteoma is considered to be a single tumor, there are reports of multilevel lesions [22]. The main clinical manifestation of osteoid osteomas is pain, usually aggravated at night; in 80 % of patients, it is effectively but temporarily controlled by taking NSAIDs (the socalled aspirin test). Very often (63–70 % of observations), non-structural (antalgic) scoliosis develops in adolescents because of pain, and the tumor is usually located at the apex of the concave side of the deformity. As the duration of the therapeutic pause increases, scoliosis may become structural as a result of asymmetric inhibition of vertebral apophysis growth [23]. Although the tumor is most often located in the posterior regions of vertebral bodies, it also may spread to the vertebral body, and very rarely - isolated lesion of the vertebral body [14, 21].

A typical feature of pain in osteoblastoma is the lack of relief from NSAIDs. In rare cases, systemic symptoms such as fever and weight loss may be observed, which is explained by an increased immune response to the tumor [24]. While scoliosis is less common in osteoblastoma than in osteoid osteoma, neurological disorders are observed in more than half of patients [25, 26].

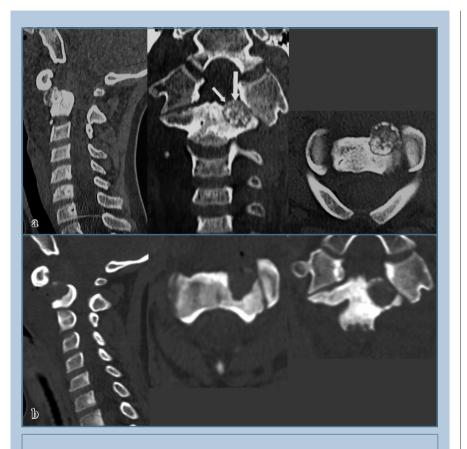


Fig. 2

CT scan of the cervical spine of an 8-year-old child with a tumor of the left half of the C2 vertebral body (sagittal, frontal and axial planes): \mathbf{a} – before surgery, tumor in sectors 5–8, zone A–B according to the WBB classification; \mathbf{b} – after marginal resection of the C2 body through a transoral approach; histological report – osteoid osteoma

Table 3

Distribution of patients with vertebral osteoid osteoma and osteoblastoma by surgical treatment technique, n (%)

Surgical treatment	Osteo	Osteoid osteoma		Osteoblastoma			
	(n	(n = 29)		(n = 12)			
Marginal resection	10 (34.5)	p = 0.043*	—	p = 0.021*			
Intralesional resection	4 (13.8)		2 (16.7)				
Wide en block resection	15 (51.7)		10 (83.3)				
* Differences in distributions are statistically significant ($p < 0.05$).							

CT is the primary imaging technique used to clarify the location and extent of both tumors. The use of MRI as an option of primary imaging of osteoid osteoma is less frequent since it can be misleading because of the spread of reactive edema to the bone and soft tissues, which is misdiagnosed as infection or an aggressive tumor process [6]. Alternately, osteoblastomas on MRI may demonstrate low signal intensity on T1- and T2-weighted images and are consistent with highly mineralized lesions with variable adjacent signal, similar to edema, and signal amplification in the bone marrow and surrounding soft tissues. In some cases, a fluid level can be visualized, making the image similar to aneurysmal bone cysts [15].

Osteoid osteomas are characterized macroscopically by the presence of dense excess bone (hyperostosis), and their bed is sclerotic bone surrounding a foci of osteoid tissue with high vascularization. Its central part microscopically contains differentiated osteoblasts visible as a single laver around trabeculae of unmineralized or mineralized bone tissue. Conversely, macroscopically, osteoblastoma is usually manifested by a red-browncolored mass reflecting its intense vascularization. Microscopically, the tumor is similar to an osteoid osteoma [6, 14]; its margins are usually well-defined, and there are no signs of destructive invasion outside the bone, which, as well as the absence of atypical mitotic figures, is the most reliable histological sign for differentiating osteoblastoma from osteosarcoma-like osteoblastoma [27]. The main sign of differentiation between osteoid osteoma and osteoblastoma is considered to be the frontier size of the tumor. estimated at 20 mm. However, in such an anatomical formation as the spine with the spinal canal and radicular foramen, this sign, in our opinion, should be considered controversial.

The analysis of the two groups of children reported by us, while matching with the literature data on the frequency distribution of tumors among themselves and the predominant lesion of posterior vertebral structures (in our cohort, sections 2–4 and 9–11 according to the WBB classification were affected in 93 % and 75 % of cases of osteoid osteomas and osteoblastomas, respectively), suggests some peculiarities.

1. Predominantly right-sided location of osteoid osteomas (sector 9-11) has no explanation but is consistent with the data of some foreign researchers [28].

2. According to the WBB classification, the location of osteoid osteomas in zone C in more than half of cases (55 %) and the spread of 50 % of osteoblastomas into the canal to zone D confirm the more aggressive course of the latter and explain the possible onset of neurological symptoms.

3. The inconsistency of clinical and radiological interpretation and similarity of morphological pattern may result in errors in differential diagnosis and treatment strategy of osteoid osteoma and osteoblastoma, which is shown in our observation. The formal determination in this case of a tumor size of 16 mm provided the reason for the diagnosis of osteoid osteoma, which possibly could have induced pathologists to make the same diagnosis.

4. The administration of NSAIDs for osteoid osteoma should be considered only as symptomatic treatment. A surgical treatment is strictly indicated when there is no effect of NSAIDs, in cases when side effects of their use develop, and, obviously, when neurological disorders develop [5, 11].

5. The technique of percutaneous radiofrequency ablation has been developed for patients with osteoid osteoma with pain only, who respond well to NSAIDs [5]. However, its use on the spine is limited due to the potential risk of injury to neural structures. Rosenthal et al. [28, 29], the first developers of the technique, suggested that thermal necrosis of neural structures during this procedure is likely to occur within a radius of up to 13 mm from the area of exposure. Therefore, the technique cannot be used in children with the predominant location of tumors in the zone C of sectors 2-4and 9-11, which are usually close to the nervous structures (especially in osteoblastomas involving the zone D according to the WBB classification). The potential for widespread use of this technique in pediatric patients is also reduced, on the one hand, by controversial results of small series of minimally invasive techniques for the treatment of osteoid osteoma of the spine [30-32] and, on the other hand, by the lack of histological verification of the tumor during radiofrequency ablation.

6. Selective embolization of nutrient arteries has been described in some arti-

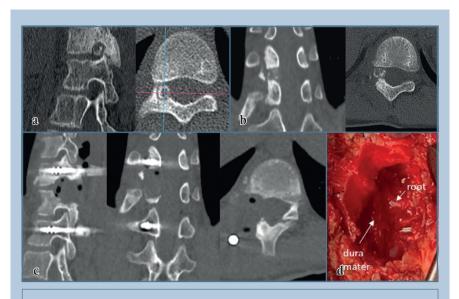


Fig. 3

CT scan of the thoracic spine (a) in the sagittal and axial planes of a 13-year-old child with a space-occupying lesion of 16 mm in diameter in the area of the pedicle of the T11 arch (sector 9–10; zone D according to the WBB classification), extending into the radicular foramen, with moderate perifocal sclerosis without hyperostosis, persistent pain syndrome up to 5/6 points according to VAS. Based on clinical and radiological data, the tumor was assessed as osteoid osteoma, which was confirmed histologically based on the removed material. On the coronal and axial CT sections (b) 5 months after the first surgery with significant clinical improvement, but persistent pain syndrome of 1-2 points according to VAS, a recurrence of the tumor (sector 9-11; zone D according to the WBB classification) with the onset of hyperostosis was noted. Histological report after marginal resection: osteoid osteoma. Another 6 months later, also with clinical improvement (rare, once a week, disturbing local sensations not exceeding 1 point on VAS), almost identical findings were revealed by CT. The sagittal, coronal and axial CT sections (c) and intraoperative images (d) show the outcomes of wide resection with decompression of the right regions of the dural sac and spinal root, unilateral instrumented stabilization. Histological report - osteoblastoma. Follow-up for 3 months after the surgery - no subjective complaints

cles as a step in the surgical treatment of osteoid osteoma and osteoblastoma of the vertebrae, which was used when the lesions seemed particularly aggressive or the diagnosis was initially uncertain [33]. This procedure was unnecessary in our cohort.

Antalgic scoliosis, which usually regresses on its own within a few months after tumor resection, has been excluded from the discussion.

Our results confirm that complete resection of the neoplasm in most cases of unilateral monosegmental intralesional resection due to location in the posterior elements of the vertebra is still safe and does not result in delayed spinal instability. It rather suggested to the extremely limited indications for performing this stage of treatment. There is no clear answer in the literature to the question about the advisability of the removal of reactive hyperostosis.

Limitations of reliability: retrospective collection of material; quite a long period of material collection (1994 to 2023), during which the information about these tumors and the possibilities of their treatment, including minimally invasive treatment, has been changed.

Conclusion

Despite the similarity of morphological pattern, osteoid osteoma and vertebral osteoblastoma have certain differences in their location, zoning according to the WBB classification, aggressive course, and presence of neurological complications.

Wide marginal resection of both tumors provides complete recovery of patients. The peculiarities of intraorganic zonation of both tumors prompt a rather reserved attitude to such minimally invasive procedures as percutaneous radiofrequency ablation. Nevertheless, the indications for instrumented stabilization of the spine, depending on the volume of resection, in the considered tumor variants seem to us to be very limited; therefore, the surgeon has the right to justifiably refuse them.

It is controversial to differentiate tumors based on such a characteristic as size. We believe that a more significant feature is their zonal location: osteoid osteomas are mostly characterized by intraosseous location (zone C according to the WBB classification), while osteoblastoma is characterized by expansion into the soft tissues of the spinal canal (zone D).

The study had no sponsors. The authors declare that they have no conflict of interest.

The study was approved by the local ethics committees of the institutions. All authors contributed significantly to the research and preparation of the article, read and approved the final version before publication.

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