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Research Article

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Significance of Magnetic Resonance Imaging for the Diagnosis and Long-Term Follow-Up of Recurrent Aneurysmal Bone Cyst after Definitive Radiation Therapy

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Abstract

Aneurysmal Bone Cyst (ABC) is a rare benign tumor in childhood, requiring definitive radiotherapy in case of inoperable recurrence. Magnetic resonance tomography (MRT) is an imaging study that is used for the diagnosis and differential diagnosis, supporting the pathohistological examination of ABC.

Against the background of a rare clinical case in a 13-year-old boy, we present the significance of MRI in assessing tumor response, namely late sclerosing bone processes in ABC after definitive intensity-modulated radiotherapy (IMRT).

Morphological sclerosing changes in the recurrent aneurysmal bone cyst after definitive radiotherapy are an extremely slow process that gradually leads to stabilization of the bone structure.

Keywords: Recurrent aneurysmal bone cyst, Magnetic resonance tomography, Intensity-modulated radiotherapy, late sclerosing bone proces

1. Introduction

Aneurysmal bone cysts (ABC) are aggressive benign lesions, the name of which was first placed by Dr. Jaffe and Lichenstein in 1942 [1,2]. In the general population, ABC has a predilection for children and young individuals, is diagnosed more commonly in the second decade of life and has a male to female ratio of 1:1.16 [3]. ABC typically affects teenagers up to the age of 20 and is present in the femur, tibia, fibula, humerus, skull and spine [4]. ABC is an expansive, lytic pseudotumorous bone lesion composed of blood-filled spaces separated by connective tissue septa formed by reactive bone tissue, fibroblasts, and osteoclast-type giant cells [5]. Microscopically, these cavernomatous spaces have fibrous walls that lack the normal features of blood vessels, including endothelium, elastic lamina, and muscle [6]. Typical ABCs in MRI can show internal cystic cavities, low signal intervals of varying size and signal inten sity, fluid planes within the lesion, cortical bone fractures and pushing at the border of the soft tissue, with equal or slightly low signal in T1WI sequences and mixed little high or high movement in T2WI [7]. Against the background of a rare clinical case in a 13-year-old boy, we present the significance of MRI in assessing tumor response, namely late sclerosing bone processes in recurrent ABC after definitive intensity-modulated radiotherapy (IMRT).

2. Clinical Case

In September 2022, a 13-year-old boy complained of slowly progressive pain in the lower spine, radiating to the back of his right leg. The pain was difficult to control with painkillers. After consultation with a neurosurgeon and imaging studies (CT and MRI), the child was admitted for surgical treatment. From ***MRI of the spine/November 2022*** - Tumor process on the right, involving the body and right pedicle of the fifth lumbar vertebra (L5), exerting anterolateral compression on the dural sac and nerve root at this level, with MR imaging data for an aneurysmal bone cyst **(Figure 1)**.

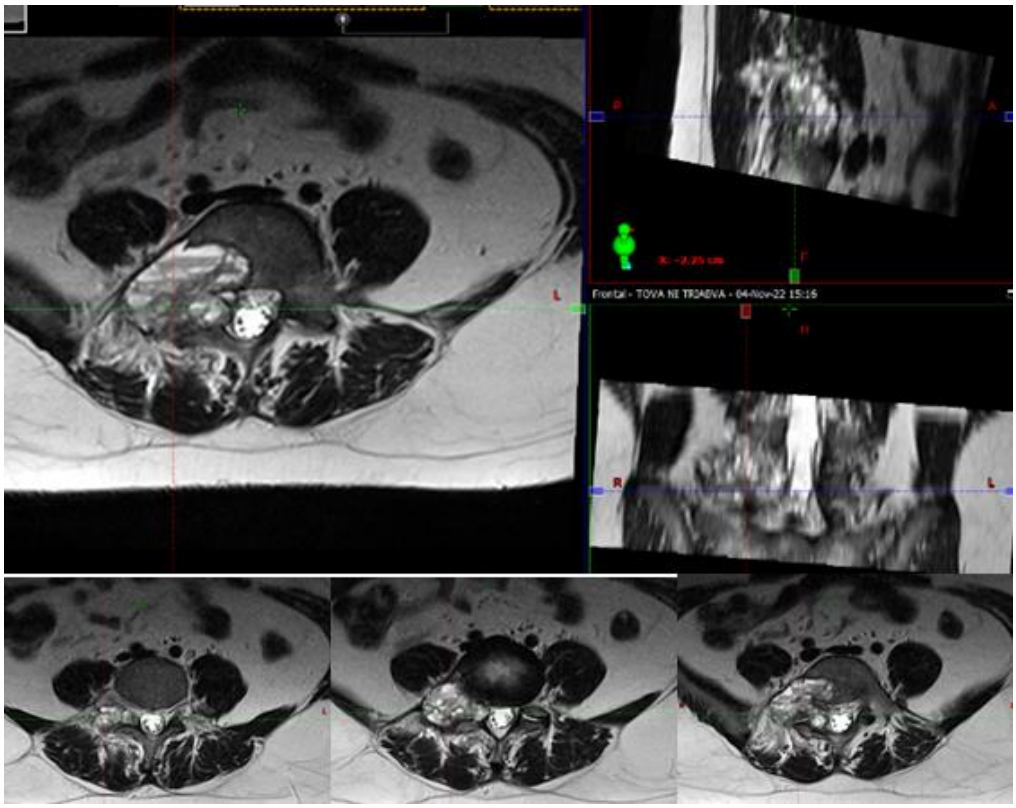

Figure 1: MRI of the spine/November 2022; Tumor process on the right, involving the body and right pedicle of the fifth lumbar vertebra (L5), exerting anterolateral compression on the dural sac and nerve root at this level

Intraoperatively: A hemilaminectomy was performed at L5 and a partial one at L4. An extradural mass was found in the body of L5 with ventral compression on the dural sac, concentrically involving the right spinal nerve root at this level. The lesion was soft, profusely bleeding, with the presence of multiple hemorrhagic cysts and fibrous tissue in the periphery, with macroscopic features of a primary bone tumor of the body of L5. A subtotal tumor resection was performed with wide decompression of the neural structures and vertebroplasty of the body of L5.

Pathohistological result /08.12.2022: Fresh hemorrhages, as well as cystic spaces without clearly formed vascular walls, predominate in the examined material. Peripherally, proliferation of mesenchymal cells and accumulation of osteoclast-type giant cells are visible. Single bone lamellae are found. The lesion is partially encapsulated. Diagnosis - Aneurysmal bone cyst. Due to the resumption of pain syndrome three months after the operation, a control MRI of the spine was performed, which established progressive tumor persistence, compressing the spinal canal.

MRI of the spine / March 2023: The inhomogeneous, mostly high-signal T1 multicystic mass, encompassing the body of L5 on the right with the corresponding peduncle, arch and transverse process, has the following dimensions - anteroposterior about 6.1 cm, transverse 7.8 cm - and craniocaudal 5.1 cm, with the increase being at the expense of the transverse dimension/towards the body medially and to the left. The stenosis of the neuroforamen on the right persists, as the corticalis is not disrupted. Conclusion - MRI data for an advanced aneurysmal bone cyst **(Figure 2)**.

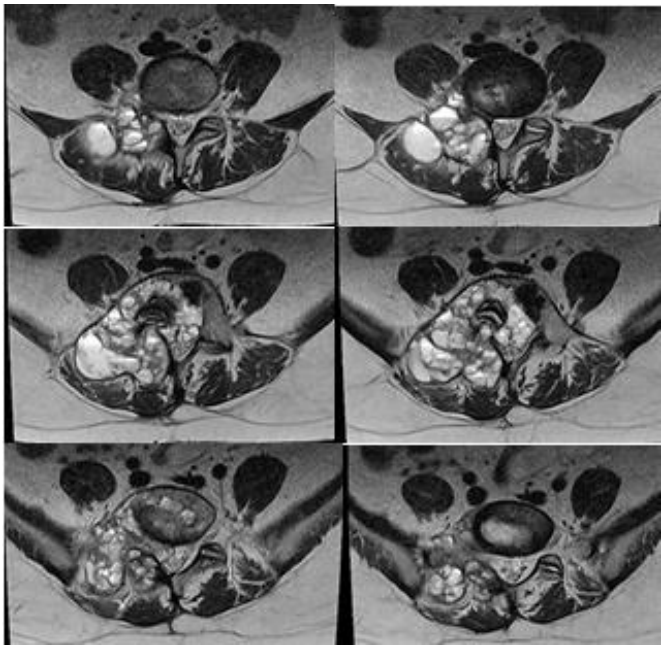

Figure 2: MRI of the spine / March 2023- The inhomogeneous, mostly high-signal T1 multicystic mass, encompassing the body of L5 on the right with the corresponding peduncle, arch and transverse process, has the following dimensions - anteroposterior about 6.1 cm, transverse 7.8 cm - and craniocaudal 5.1 cm, with the increase being at the expense of the transverse dimension/towards the body medially and to the left.

Given the benign nature of the tumor, the child's age and the impossibility of radical tumor extirpation, we decided to perform intensity-modulated radiotherapy (IMRT) using the VMAT method to a total tumor dose of 30 Gy with a daily dose of 2 Gy. Non-steroidal analgesics were required during radiotherapy, as the pain syndrome did not subside until its end. After 3 months from the completion of IMRT, the heterogeneous volume formation presented with significant regression and reduction of a large part of the cystic bone components with the absence of sediments between them in the form of levels **(Figure 3)**.

6 months after completion of IMRT, the control MRI **(Figure 4/A)** did not report a significant reverse reduction of the aneurysmal bone cyst. MRI after 9 months of IMRT - The observed heterogeneous volumetric formation presents without dynamics in its imaging characteristics compared to the transitional MRI examination - affects the body of the fifth lumbar vertebra, swells the right pedicle, lamina and facet joint. Causes stenosis of the corresponding neuroforamen. The structure of the formation is heterogeneous with confluent hyperintense areas in T2 and STIR, hypointense in T1, as well as reticular osteosclerotic areas. There is no noticeable cortical erosion. The body of the fifth lumbar vertebra is wedge-shaped in dorsal aspect with deformation to the right. Intraspongially, osteosclerotic material is observed, presented as low signal in all dimensions **(Figure 4/B)**.

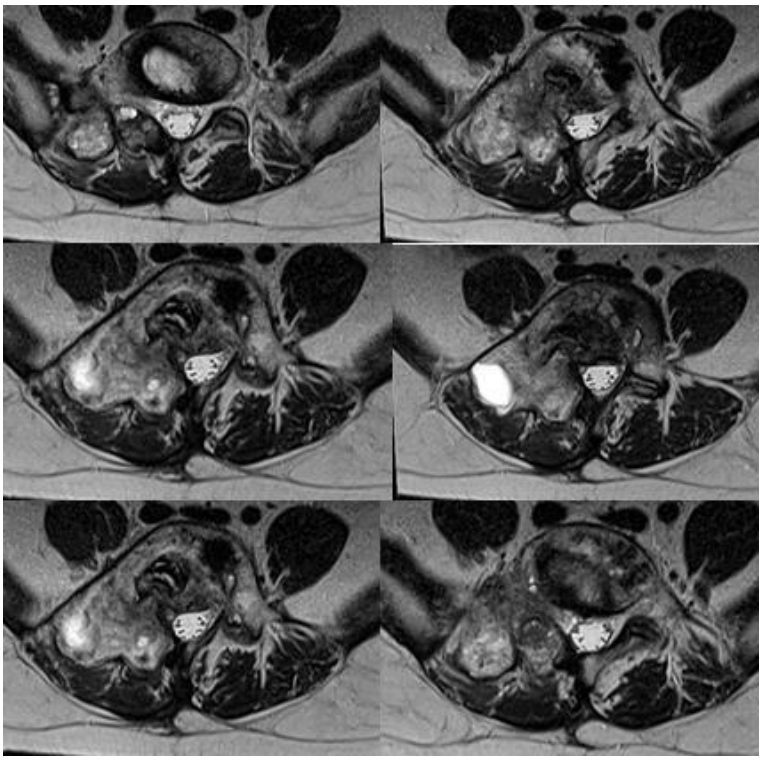

Figure 3: MRI 3 months after completion of IMRT - The tracked heterogeneous mass is presented with significant regression and reduction of most of the cystic bone components with no sediments between them in the form of levels. The bone structure is heterogeneous, with confluent T2 and STIR hyperintense areas, without cortical erosion. Residual cyst lateral to the protruding bone structure of L5 measuring 1.5/1.9 cm.

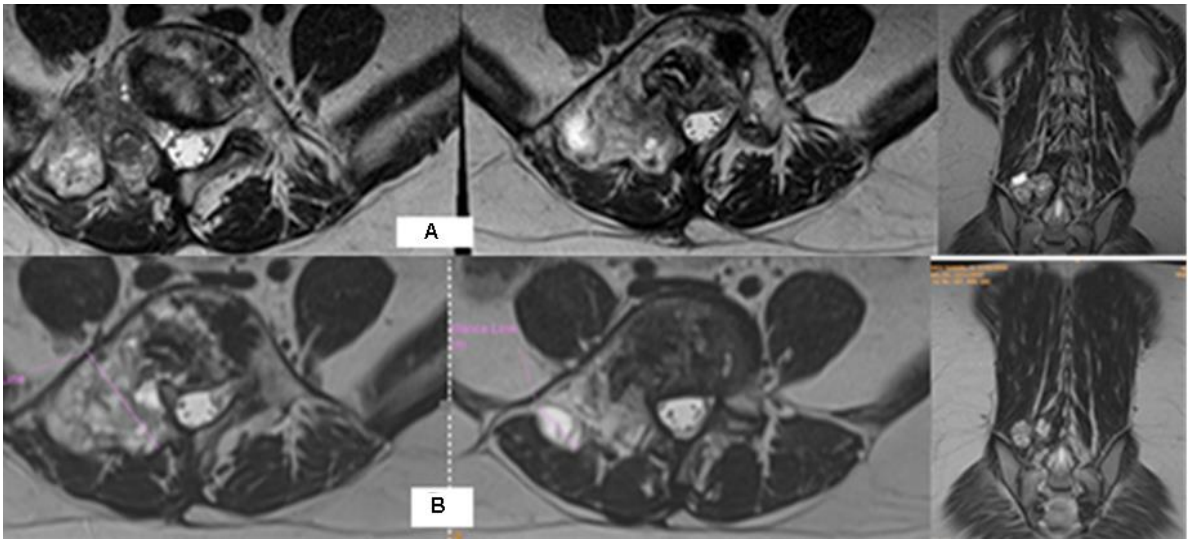

Figure 4: A/ MRI after 6 months of IMRT - without significant reverse dynamics of the aneurysmal bone cyst; **B/ MRI after 9 months of IMRT -** The structure of the formation is heterogeneous with confluent hyperintense areas in T2 and STIR, hypointense in T1, as well as the presence of reticular osteosclerotic areas.

MRI of the spine after 1 year of IMRT/ 22.05 24: The monitored heterogeneous volumetric formation presents without significant dynamics in its axial dimensions of 4.3 / 7.9 cm with transitional 4.3 / 8.4 cm with some advancement of its cystic components. The formation involves the body of the fifth lumbar vertebra, inflates the right pedicle, lamina and facet joint of the fifth lumbar vertebra. It causes stenosis of the corresponding recess and neuroforamen. The structure of the formation is markedly heterogeneous with

cystic areas, as well as with reticular osteosclerotic areas. Without palpable cortical erosion. Without dynamics of the residual cyst with current dimensions of 2.1 cm, prominent towards the erector spinae muscle. The intervertebral disc at the level of L 4/5 is slightly lowered, with a right eccentric disc protrusion foraminally homolaterally - without dynamics. Level L5/S1 - no significant disc pathology, free neuroforaminals. The body of the fifth lumbar vertebra presents as a wedge-shaped depression in dorsal aspect with a deformation to the right **(Figure 5)**.

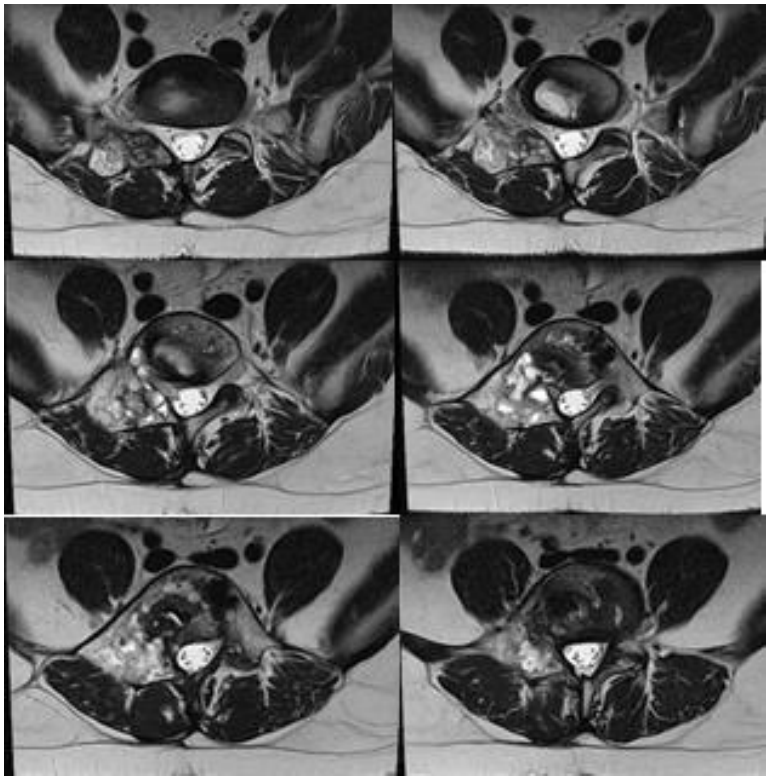

Figure 5: MRI of the spine after 1 year of IMRT/ 22.05 24. The observed heterogeneous volumetric formation presents without dynamics in its axial dimensions of 4.3 / 7.9 cm compared to the transitional axial dimensions of 4.3 / 8.4 cm and with some progression of the cystic components. The structure of the formation is markedly heterogeneous with cystic and reticular osteosclerotic areas.

After 2 years of IMRT/ 26.06 25 - Deformity of L5 on the right with a residual formation with reduced axial dimensions of 4.01cm/7.64cm compared to the examination of 15.11.2024. **(Figure 6)** Currently, the boy feels well, without pain.

3. Discussion

Aneurysmal bone cysts are non-malignant, tumor-like, vascular lesions comprised of blood-filled channels. Although they can occur in any bone, they are most common in the femur, tibia, and vertebrae [8]. In the latest WHO classification (2020) for bone tumors, there is a change in nomenclature, in which the terms "ABC" and "ABC-like changes" are proposed instead of "primary ABC" and "secondary ABC" [9,10]. ABC of the spine accounts for 15% of primary bone tumors [11]. Magnetic resonance imaging (MRI) demonstrates similar findings as CT. T1 contrast-enhanced and T2 weighted images can emphasize the septa within the lesion, revealing rims of low T1 and T2 signal [12].Fluid-fluid levels, in particular, are strongly suggestive of an ABC [13]. MRI plays a uniquely diagnostic important role, as the modality can depict the involvement of adjacent soft tissues, presence of solid components, and extent of surrounding edema, which is frequently underestimated on radiographs and CT scans. The presence of enhancing solid components on MRI raise concern for an associated malignant lesion; however, solid variants of ABC have been described [14]. The term "solid variant" ABC has been used interchangeably with a histologically indistinguishable entity, giant-cell reparative granuloma. This term has commonly been used in the literature to describe lesions that involve gnathic sites, either the mandible or maxilla, or the short tubular bones of the hands and feet [15]. The term "solid variant" ABC is classified as an osteoclastic giant cell rich tumor (WHO 2020) [16]. Radiographically, solid variant ABC is typically lytic and expansile with a variable degree of mineralization; however, it can be non-expansile in approximately one-third of the cases [17].

Combination of both radiographic and MRI features help clinch the diagnosis although histological examination is often warranted to exclude other malignant lesions [18]. The differential diagnosis of primary ABC includes other giant cell-containing tumors of the bone, particularly giant cell tumor (GCT) [19]. A combination of

both radiographs and MRI, alongside histological examination, is required to increase the likelihood of an accurate diagnosis [20].

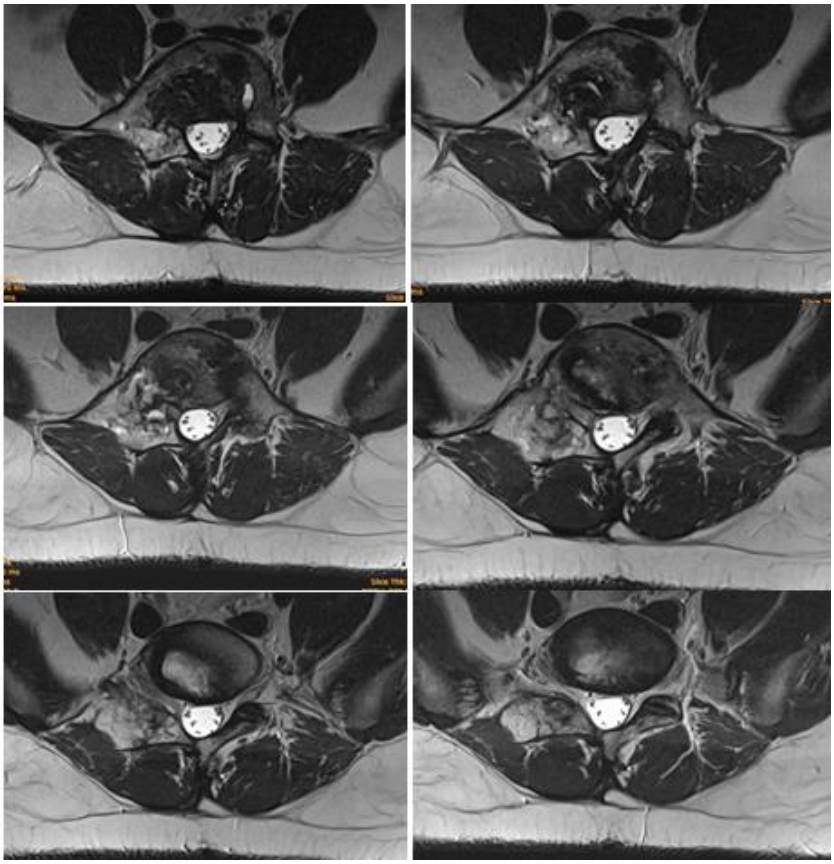

Figure 6: MRI after 2 years of IMRT/ 26.06. 2025 - Deformity of L5 on the right with a residual formation with reduced axial dimensions of 4.01cm/7.64cm compared to the examination of 15.11.2024.

Prognosis and management differ among these lesions, and their imaging features often overlap. Imaging plays an important role in assessing their size, location, aggressiveness, degree of soft-tissue extension, and presence of solid components to help guide optimal management [21]. Aneurysmal bone cysts are expansile lytic lesions consisting of thin-walled cavities containing blood or serosanguineous fluid with a thin shell of overlying bone [22]. The cysts are of a variable signal, with a surrounding rim of low T1 and T2 signals. Focal areas of high T1 and T2 signal [23] are also seen, presumably representing areas of blood of variable **(Figure 1)**.

In a study of 53 patients, Rajeswaran et al. found that fluid-fluid levels distributed throughout a bone lesion was almost always indicative of benign process and 78% of the lesions corresponded to ABCs [24]. On MRI, fluid-fluid levels are the hallmark of the lesion but are not pathognomonic as these are seen in several bone neoplasms. Soft-tissue and bone marrow edema, which are best depicted on MRI due to its high contrast resolution, can be present in ABCs [15].

According to the morphological classification of Capanna et al. [25], which can be applied to the radiographic and macroscopic appearance of the preparation, the presented ABC is type II, which means expansile tumor with cortical thinning involving the entire bone segment **(Figure 2)**.

Multiple treatment options have been described, which can be used alone or in combination, including curettage with or without bone grafting, complete tumor resection, selective preoperative embolization, radiotherapy (RT), chemotherapy, and intralesional injections [26]. Radiotherapy is an option that is currently reserved for patients at high risk for surgery or for those who are refractory to surgical treatment, especially considering the potential risks of post-radiation myelopathy or sarcomatous transformation [27]. Five German institutes have collected data on the clinical characteristics, treatment concepts, and treatment outcomes of patients with ABC after local percutaneous RT over the past 30 years. All seven patients reported tumor control without recurrence or pain during follow-up [28]. RT is effective in recurrent ABC with a total radiation dose of 25–36 Gy, which has satisfactory results [29,30].Although many authors consider RT in ABC to be an aggressive treatment approach similar to amputation, RT is reserved for inoperable recurrences after orthopedic surgery in the vertebral area. To present the effect of treatment expressed

in tumor sclerosis two years after radiotherapy with a relatively low total radiation dose of 30 Gy, we show the dynamics of the morphological characteristics of the tumor by follow-up MRI.

When comparing the MRI images of the tumor, we report a significant reduction in its volume after 3 months of completing RT **(Figure 3)**, but only after 9 months do radiation-induced sclerotic tumor changes appear, expressed through osteosclerotic zones **(Figure 4 B)**. The residual formation after 2 years has reduced axial dimensions compared to the previous year **(Figure 7)**. We see that structural changes in aneurysmal bone cyst after definitive RT are an extremely slow process that gradually leads to stabilization of the bone structure.

4. Conclusion

Aneurysmal bone cyst is a rare benign bone tumor in children and adolescents. Before treatment, a biopsy is required to confirm the diagnosis, accompanied by preoperative MRI. In case of recurrent and inoperable spinal ABC, despite the benign nature of the disease and the adolescent age, percutaneous radiotherapy with a low total dose of 30 Gy-36 Gy is required. MRI is a valuable imaging, necessary for long-term follow-up after RT. When comparing the MRI images of the recurrent tumor, we report a significant reduction in its volume immediately after 3 months of completing RT, but only after 9 months do radiation-induced sclerotic tumor changes, expressed through osteosclerotic zones, appear. We see that structural changes in recurrent ABG after definitive RT are an extremely slow process, which gradually leads to stabilization of the bone structure.

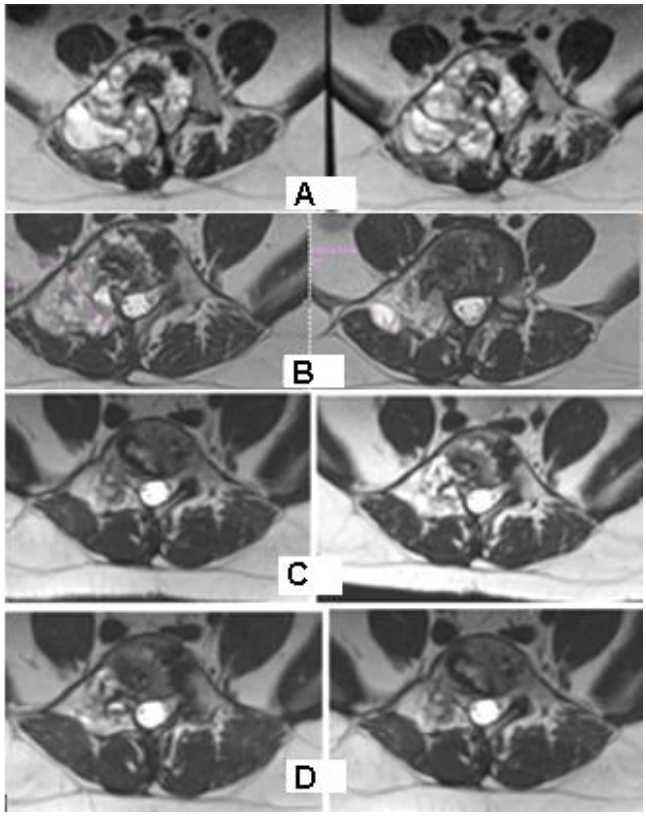

Figure 7: A/ Comparison of MRI before IMRT; B/ MRI after 9 months of IMRT -The structure of the tumor is heterogeneous with confluent hyperintense areas in T2 and STIR, hypointense in T1, as well as the presence of reticular osteosclerotic areas; **C/ MRI after 1 year from IMRT /2024** -Residual tumor with axial dimensions of 4.3 / 7.9 cm with transitional 4.3 / 8.4 cm with some advancement of its cystic components. **D/ MRI after 2 years from IMRT /2025** -Residual tumor with reduced axial dimensions compared to MRI from 15. 11. 2024 / now 4.01 cm/7.64 cm

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