

Intensity Modulated Radiotherapy For Recurrent Spine Aneurysmal Bone Cyst In Adolescence-Case Report And Review of Literature

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Submitted: 05 Oct 2023; Accepted: 11 Oct 2023; Published: 25 Oct 2023

Citation: Marinova L, Vasileva V, Gabrovski I, Kahchiev N, Radev R (2023). Intensity Modulated Radiotherapy For Recurrent Spine Aneurysmal Bone Cyst In Adolescence-Case Report And Review of Literature. *Medical & Clinical Research*, 8(10), 01-06.

Abstract

Aneurysmal bone cysts are rarely diagnosed aggressive benign bone tumors that require CT and MRI imaging and strict histopathological analysis. This clinically and histologically benign tumor is mainly diagnosed in children and adolescents in the long bones of the limbs, but in 12%-30% it is localized in the spine.

Since the value of external beam radiotherapy (EBRT) for aneurysmal bone cyst (ABC) has not been well defined, we present our observations and treatment outcome after the realization of intensity modulated radiotherapy (IMRT) up to total dose 30 Gy for recurrent spine aneurysmal bone cyst in adolescent age. In this article, for the first time in the world medical literature, an MRI imaging finding after performed IMRT in recurrent ABC is presented.

Our observations lead to the conclusion, that radiation therapy is an effective treatment method for relapsed or inoperable disease, achieving not only a symptomatic pain-relieving effect, but also a significant tumor reduction and restoration of the bone structure.

Keywords: Aneurysmal Bone Cyst, Benign Bone Tumor, Magnetic Resonance Imaging, Histopathological Analysis, Intensity Modulated Radiotherapy.

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]. It is an expansive, lytic pseudotumoral bone lesion composed of blood-filled spaces separated by connective tissue septa formed by reactive bone tissue, fibroblasts, and osteoclast-type giant cells [3]. Most cases have been identified in the first two decades of life; it is rare to find them after the age of 30 [4]. Treatments have ranged from simple curettage, with or without bone graft, injection of a fibrosing agent, complete resection surgery, radiotherapy (RT) and selective arterial embolization to a combination of these methods [5]. Since the value of external beam radiotherapy (EBRT) for ABC has not been well defined, we present our observations and treatment outcome after the implementation of 30 Gy RT by the VMAT method for lumbosacral ABC in a 14-year-old boy.

Clinical Case

Since September 2022, a 13-year-old boy presented with slowly progressive low back pain that spread to the posterior surface of the right leg. The pain was hardly affected by conservative

treatment. After CT and MRI imaging studies and consultation with a neurosurgeon, the child was admitted for surgical treatment. From Spine MRI/ November 2022-Evidence of a tumor process on the right involving the body and right pedicle of the fifth lumbar vertebra (L5), exerting antero-lateral compression on the dural sac and on the nerve root at that level, with imaging features of aneurysmal bone cyst (Figure 1).

Intraoperative: Percutaneous vertebroplasty was performed through the left pedicle at L5. A hemilaminectomy was performed at L5 and partial at L4. An extradural tumor formation was encountered from the body of L5 with ventral compression on the dural sac, concentrically involving the right root. The mass was soft, profusely bleeding, with the presence of multiple intradural hemorrhagic cysts and fibrous stroma at the periphery, with macroscopic features of a primary bone tumor of the body of L5. The tumor was subtotally resected, achieving wide decompression of the neural structures. Vertebroplasty of the bony tumor bed was performed.

Pathohistological result: mFragments of cystic spaces with areas of proliferated spindle cells, among which giant cells of osteoclast

type are present. Adjacent, extensive hemorrhages with ectased vascular spaces are reported. Bone beds are also found in places.

Conclusion: The histological picture corresponds to an aneurysmal bone cyst.

Revision of the histological result/08.12.2022: Fresh hemorrhages predominate in the examined material, followed by cystic spaces without clearly formed vascular walls. In their periphery, proliferation of mesenchymal cells and clustering of osteoclast-type giant cells can be seen. Single bone lamellae are found. The lesion is partially encapsulated. The diagnosis of aneurysmal bone cyst is confirmed. Due to the resumption of the pain syndrome, in January 2023, a follow-up MRI of the spine was performed, which established a recurrence of the disease with tumor size advancing -7.5/6/5.1 cm with compression on the spinal canal.

Spine MRI/January 2023

Dorsal body of L5 presents wedge-shaped depression with deformation and erosion on the right. Osteosclerotic material intraspongious. The tracked heterogeneous volume formation has a complex lobulated shape with current maximum dimensions transversely 7.5 cm, ventro-dorsally 6 cm and cranio-caudally up to 5.1 cm. Considerable advancement of the tumor lesion is reported. The tumor has sharp contours and a characteristic multicystic septate structure with formed liquid-liquid levels. It causes ventral and lateral stenosis of the spinal canal with dural compression and obliterates the corresponding root canal. It latero-dorsally compresses the paravertebral muscles and in particular the erectus vertebrae muscle.

Conclusion: Advanced aneurysmal bone cyst at L5. Postoperative changes. Condition after L5 vertebroplasty.

Spine MRI /March 2023

The inhomogeneous mostly high-signal T1 multicystic mass involving the body of L5 on the right with the corresponding peduncle, arch and transverse process persisted. The dimensions in anterior-posterior dimension are about 6.1 cm, transverse 7.8 cm and caudal-cranial 5.1, the advancement 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 disturbed.

Conclusion: MRI data for an advancing aneurysmal bone cyst (Figure 2). After consultation with a pediatric hematologist and a pediatric radiation therapist, a decision was made for radiation therapy given the available literature data for a beneficial effect in recurrent or inoperable aneurysmal bone cysts. Considering the benign nature of the lesion and its volume, we performed intensity modulated radiotherapy (IMRT) using the VMAT method up to a total dose of 30 Gy with a daily dose of 2 Gy (Figure 3 and 4). During RT, non-steroidal analgesic drugs were required, as the pain syndrome did not resolve until the RT end. 6 months after the completion of IMRT, we performed a control MRI (Figure. 5), which showed a significant reverse reduction of the aneurysmal bone cyst against the background of complete anesthesia without the use of pain medication (Figure. 6).

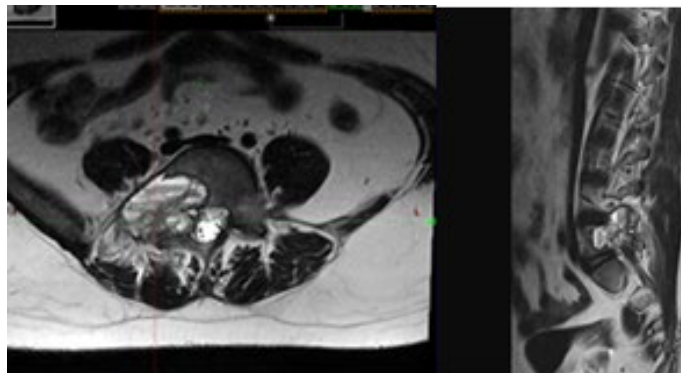


Figure 1: Spine MRI/ November 2022-Evidence of a tumor process on the right involving the body and right pedicle of the fifth lumbar vertebra (L5), exerting antero-lateral compression on the dural sac and on the nerve root at that level, with imaging features of aneurysmal bone cyst.

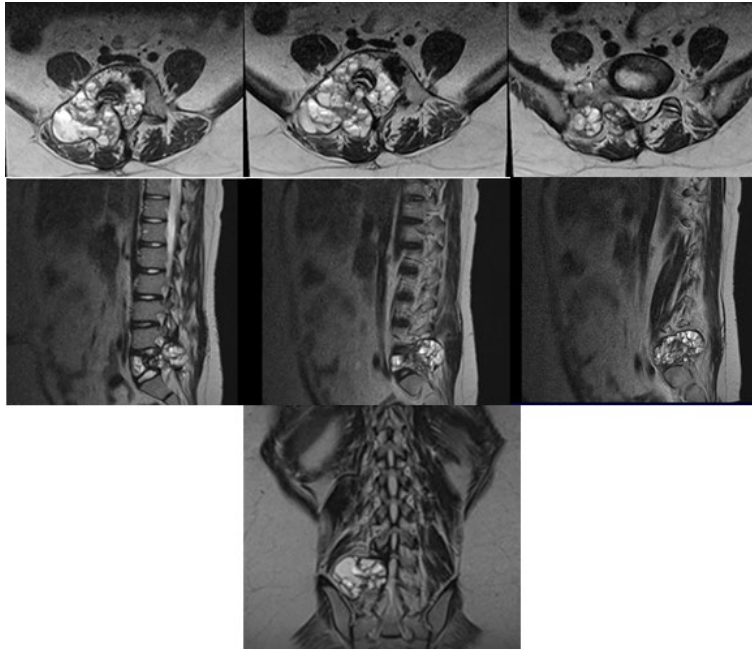


Figure 2: Spine MRI /March 2023-The inhomogeneous mostly high-signal T1 multicystic mass involving the body of L5 on the right with the corresponding peduncle, arch and transverse process persisted. The dimensions in anterior-posterior dimension are about 6.1 cm, transverse 7.8 cm and caudal-cranial 5.1, the advancement 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 disturbed. Conclusion-MRI data for an advancing aneurysmal bone cyst.

Discussion

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 [6]. In the last WHO Classification of Tumors of Bone (2020), there has been a shift in nomenclature in which the terms “ABC” and “ABC-like changes” that are found within certain preexisting primary bone neoplasms are suggested instead of “primary ABC” and “secondary ABC,” respectively [7]. The spine ABC represent 15% of primary bone tumors [8]. Because there are few reported aggressive case reports of spinal ABC, diagnostic and treatment algorithms remain controversial and variable [4]. They are usually expansive blood-filled cavities within the bone lined by

proliferative fibroblasts, giant cells, and trabecular bone [9]. The majority of ABCs are primary, and some are secondary, but both primary and secondary ABCs can develop into osteosarcoma, and certain benign or malignant tumors may be combined with ABCs [10]. The pathohistological analysis of the presented clinical case is a typical characteristic of ABC, including fragments of cystic spaces with areas of proliferated spindle cells, among which giant cells of osteoclast type and extensive hemorrhages with ectased vascular spaces. Strict pathohistological analysis requires a differential diagnosis with a number of benign and malignant bone tumors such as unicameral bone cyst (UBC) and telangiectatic osteosarcoma, therefore a biopsy is recommended before treatment [11].

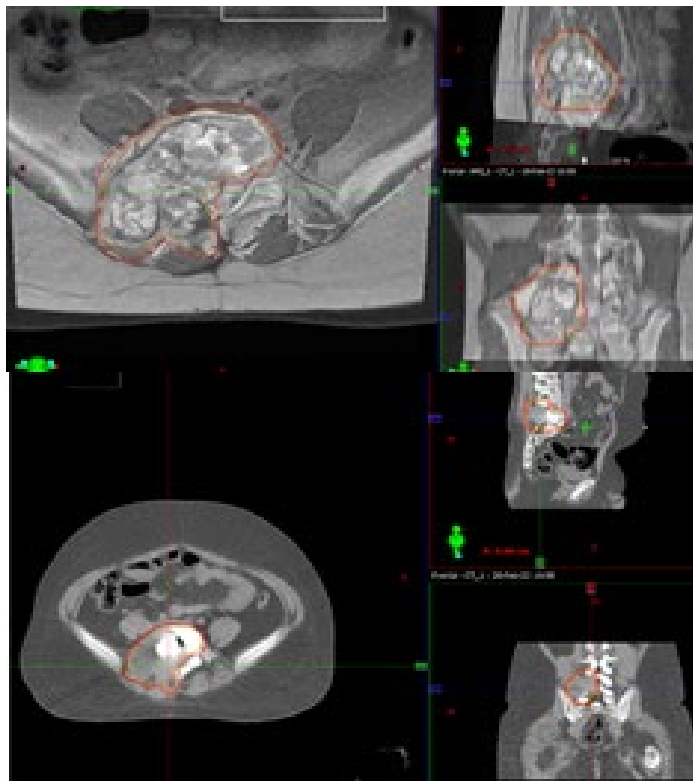


Figure 3: Contouring of the CTV after overlaying the MRI with the planning CT in IMRT planning.

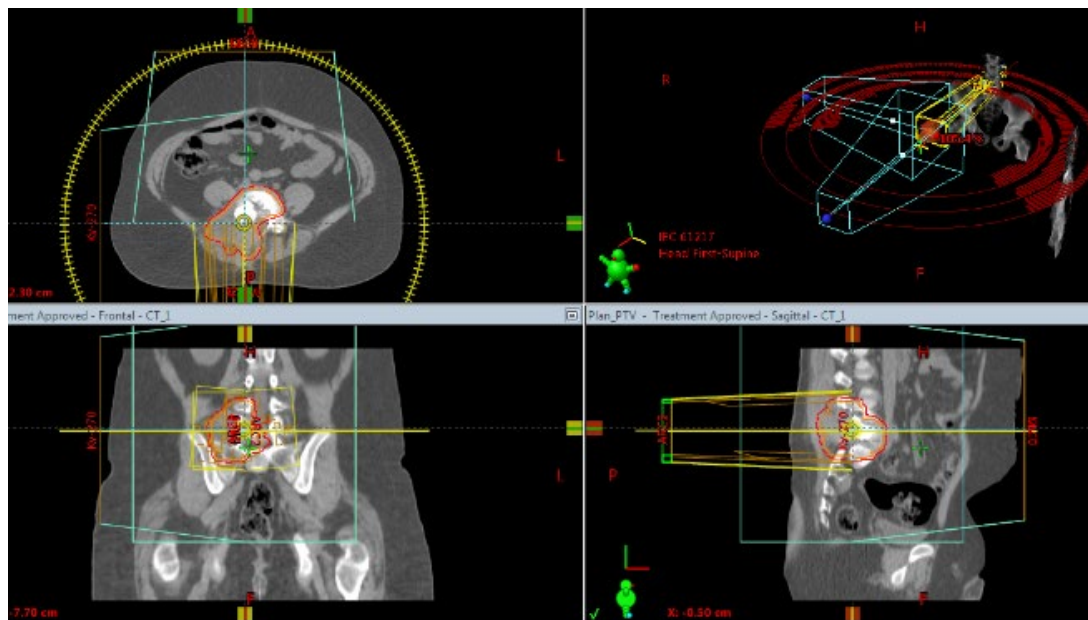


Figure 4: Intensity modulated radiotherapy (IMRT) using the VMAT method up to a total dose of 30 Gy with a daily dose of 2 Gy.

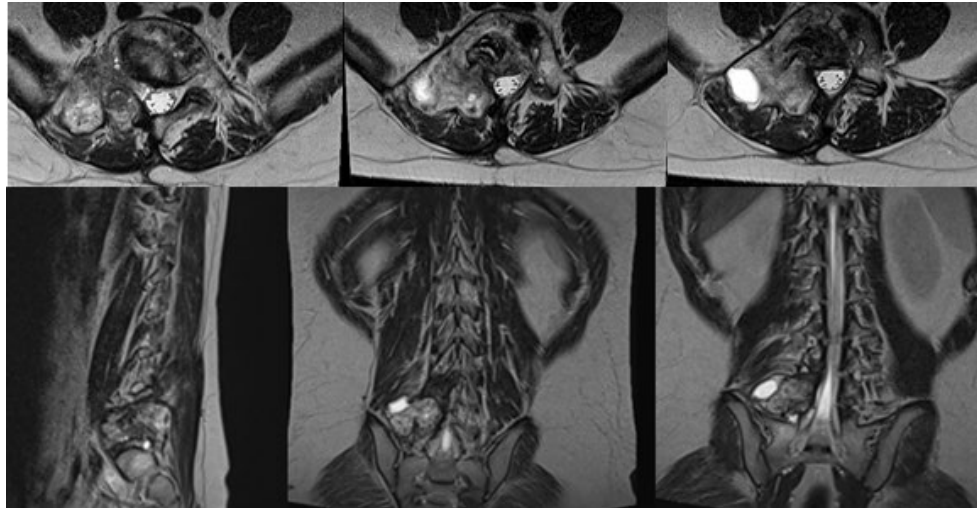


Figure 5: The follow-up MRI 6 months after the completion of IMRT/November 2023-The tracked heterogeneous volume formation is presented with a significant reverse development and reduction of a large part of the cystic bone components with a lack of sediment among them in the form of levels. The bone structure is heterogeneous, with confluent T2 and STIR hyperintense areas, without cortical erosion. Residual cyst lateral to the bulging bony structure of L5 measuring 1.5/1.9 cm.

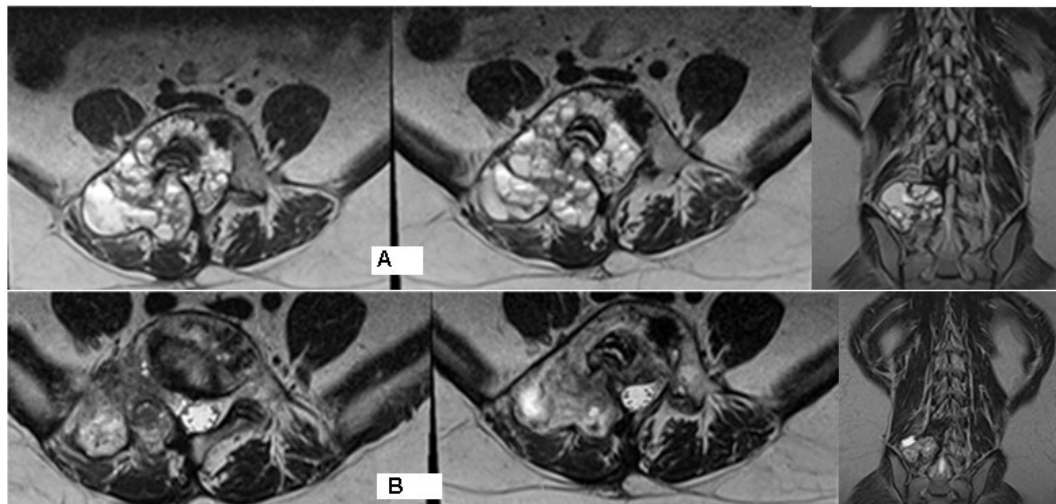


Figure 6: Comparison of MRI images before and after IMRT.

On MRI presence of multiloculated fluid-fluid-filled cysts is characteristic of ABC [1]. Because it can be difficult to differentiate an ABC from a malignancy based on imaging alone, confirming the diagnosis by means of a biopsy before treatment is imperative [12]. In the presented patient, the aneurysmal bone cyst was diagnosed in September 2022, and after subtotal surgery, progression was reported with a typical MRI image including the tumor with sharp contours and a characteristic multicystic septate structure with formed liquid-liquid levels. ABC causes ventral and lateral stenosis of the spinal canal with dural compression and obliterates the corresponding root canal (Figure 2). Multiple treatment options have been described, to be used alone or in combination, including curettage with or without bone grafting, complete tumor resection, selective preoperative embolization, radiotherapy, chemotherapy, and intralesional injections [13]. Radiotherapy is an option currently reserved for patients at high

risk of not withstanding surgery or for those who are resistant to surgical treatment, even more so considering the potential risks of post-radiation myelopathy or sarcomatous transformation [14]. Five German institutions collected data regarding clinical features, treatment concepts, and outcome for patients with ABC who had been referred for local EBRT over the past 30 years. All 7 patients exhibited a radiological response and experienced no recurrent disease activity or pain during follow-up [15]. Radiotherapy seems to be effective for recurrent cases of ABC and a dose of around 25 to 36 Gy could be effectively delivered with satisfactory results [16,17]. In the presented patient, low back pain radiating to the right leg was unresponsive to pain medication. Since the value of external beam radiotherapy (EBRT) for ABC has not been well defined, we assessed the need for radiotherapy and conducted IMRT. Considering the benign nature of the lesion and its volume, we performed intensity modulated radiotherapy (IMRT) using the

VMAT method up to a total dose of 30 Gy (Figure 3 and 4). The follow-up MRI 6 months after the completion of IMRT/ November 2023-The tracked heterogeneous volume formation is presented with a significant reverse development and reduction of a large part of the cystic bone components with a lack of sediment among 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 bulging bony structure of L5 measuring 1.5/1.9 cm. The body of L5 appears slightly wedge-shaped in dorsal aspect with deformity and erosion on the right (Figure 5). This imaging finding showed a significant tumor reduction (Figure 6), and the boy was completely anesthetized and without neurological symptoms. In this article, for the first time in the world medical literature, an MRI imaging finding after performed IMRT in recurrent ABC is presented.

Conclusion

Aneurysmal bone cyst is a rare benign bone tumor in children and adolescents. A strict histopathological analysis accompanied by preoperative and postoperative MRT imaging is necessary. The main treatment is radical surgery. In case of recurrent and inoperable spinal ABC, despite the benign nature of the disease and the child's and adolescent age, external beam radiotherapy is required. IMRT up to a total dose of 30 Gy achieves significant tumor reduction with complete analgesia and absence of neurological symptoms.

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