OMURGADA ANEVRİZMAL KEMİK KİSTİ: BİR ÜÇÜNCÜ BASAMAK HASTANE DENEYİMİ

Aneurysmal Bone Cyst of the Spine Experience at a Single Tertiary Hospital

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ÖZET

Amaç: Anevrizmal kemik kisti (AKK), lokal agresif yapıda iyi huylu bir kemik tümörüdür. Bu çalışmanın amacı, omurgada seyrek görülen ve geç tanı konulan AKK'nde uygulanan tedavilerin ve sonuçlarının değerlendirilmesi amaçlanmıştır.

Gereç ve Yöntem: 2005 ve 2019 yılları arasında hastanemizin online veritabanında omurgadaki AKK araştırması yapılmıştır. Toplamda altı hasta (4 erkek, 2 kadın) tespit edilmiş olup, hastaların ağrı başlangıcından tedavi edilinceye kadar geçen süre ortalama 2,7 yıldır.

Bulgular: Hastalarımızdan ikisinde küretaj uygulanmıştır. Üç hastamızda ise tümörün çıkarılması sonrasında iatrojenik instabilite gelişmiş, bu nedenle kısa segment posterior enstrumentasyon uygulanmıştır. Bir hastada ise eşlik eden komorbiditeler nedeniyle selektif arteriyel embolizasyon ile tümörün büyümesi kontrol altına alınmış olup komorbiditenin tedavi edilmesi sonrasında cerrahi planlanmaktadır.

Sonuç: Nörolojik bulgunun olmadığı ve deformite gelişmemiş hastalarda embolizasyon kullanılabilir. Enneking evre 2 vakalarda küretaj (ve greftleme) uygun cerrahi seçenektir. Ancak tümörün yeri ve büyüklüğüne göre, cerrahi sonrasında instabilite gelişebilir, bu nedenle kısa segment posterior enstrumentasyon uygulanabilir.

Anahtar Kelimeler: *Anevrizmal kemik kisti; Omurga; Sakrum; Selektif arteriyel embolizasyon; Cerrahi*

ABSTRACT

Aim: Aneurysmal bone cyst (ABC) is a benign osseous tumor, with a local aggressive nature. It is rarely seen in the spine which causes a delayed diagnosis and treatment. The aim of this study is to present results of ABC of the spine in our clinic and investigate the current treatment options.

Material and Method: A search of the ABCs of the spine between 2005 and 2019 years was conducted on online hospital database system. There were 6 patients (4 male, 2 female), with a duration of symptoms until treatment 2.7 years.

Results: Two patients were treated with curettage, posterior instrumentation was added due to iatrogenic instability after tumor removal in three patients, and selective arterial embolization was used to control the tumor growth in a patient with comorbidities, and surgery was planned for the patient after treatment of comorbidities.

Conclusion: Embolization can be used in patients without neurological symptoms and in the absence of deformity. Curettage and bone grafting is the viable surgical treatment method in stage 2 lesions. However more extensive surgery may be necessary in patients with spinal instability, such as posterior instrumentation and fusion, and en bloc resection with neural structure sacrifice in recurrent or primary stage 3 lesions.

Keywords: Aneurysmal Bone Cyst; Spine; Sacrum; Selective arterial embolization; Surgery

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INTRODUCTION

Aneurysmal bone cyst (ABC) is defined as aggressive, expansile and osteolytic lesions, accounting for about 2.5% of all primary bone tumors and roughly 15% of all primary spine tumors (1–3). Approximately 6% to 22% of ABCs develop in the mobile spine; 13% to 21% occur in sacrum (4–8). It is seen mostly in the second decade of life with a predilection for the posterior elements, yet the lesion may extend into the anterior column if of sufficient size.

ABC can be classified as primary or secondary, associated with bone tumors such as giant cell tumors, telangiectatic osteosarcoma, osteoblastoma or chondroblastoma (9). ABC of the spine typically present with pain, deformity, or neurological dysfunction (4,10–15). The pain may originate as localized pain from the lesion itself or due to pathologic fracture, and radicular pain from neurologic compromise.

Definitive diagnosis is obtained from radiological and histological findings (11–13,16). The characteristic radiologic features on X-ray and computerized tomography (CT) are expansile osteolytic lesions, with defined sclerotic margins and fluid-fluid levels. On magnetic resonance imaging (MRI) series, the cysts mostly demonstrate marked septations and fluid-fluid levels which often accompanied by peripheral enhancement (11,17–19). Macroscopically, these lesions appear as spongy hemorrhagic masses covered by a thin shell of reactive bone. Histologically, they appear as multiple large blood-filled spaces separated by fibrous or osseous septae and lined by fibroblasts and histiocytes. Osteoclastic giant cells and mitotic figures are typically present.

The treatment of spinal ABCs includes surgery, radiotherapy, selective arterial embolization (SAE), and a combination of these modalities. The treatment requires special consideration on tumoral, mechanical and neurological perspectives. From the tumoral point of view, excessive intraoperative bleeding particularly may be an adversity in approaching these lesions. Also, the inaccessibility of the tumor due to complex structure of the spine may inevitably result in inadequate resection and recurrence in intralesional curettage treatment, or increased morbidity is the result when en bloc resection is performed as treatment. Pre- and postoperative spinal instability in ABC of the spine have paramount importance. The lesion's proximity to the major neural structures may limit the available treatment options. A wide range of treatment options are defined as watchful waiting, SAE, cryoablation, cementation, intralesional injection (concentrated autologous bone marrow, denosumab or doxycycline), radiotherapy, curettage, en bloc resection, curettage or en-bloc resection after SAE (9,20,21).

The aim of this study is to evaluate the results of surgical treatment in ABC of the spine and investigate the treatment strategies in this rarely-seen anatomic location.

MATERIALS AND METHOD

After obtaining local ethics committee approval for the study, a retrospective review of the patients who underwent surgery with the diagnosis of spinal ABC, between 2005 and 2019 was performed.

X-ray, MRI and CT were used for radiological evaluation. Physical examination findings, duration of symptoms from the start of complaints till the surgery date, and neurologic findings were recorded.

All patients were consulted by the multidisciplinary orthopedic-oncology council prior to treatment procedure. The patients were staged according to Enneking system: latent, active and aggressive, stage 1 to 3, respectively (22) and Weinstein-Boriani-Biagini classification for tumor localization was used for surgical planning (23); described as twelve sections of axial spine image, beginning from left side of spinous process, turning clockwise, and A – F from prevertebral to the dural involvement.

The follow-up was carried out as 2nd, 6th, 12th weeks, then every three months during the first two years, then annually. Radiological evaluation of the spine and pelvis were conducted to assess recurrence and vertebral imbalance (spinal deformity), neurological examination was performed at each follow-up was noted.

Surgical Procedure

Biopsy was performed prior to treatment to confirm the diagnosis of ABC, except in patient 5. All patients, except patient 4 were operated with a posterior approach only. Patient 4 was treated with SAE. Curettage, cementation, posterior instrumentation (arthrodesis), selective arterial embolization (SAE) and their combinations were the applied treatment methods in our series (Table 1).

In stage 2 patients, intralesional curettage was the surgical procedure performed. Excision was performed in stage 3 patients. The lesion localization was determined with fluoroscopy. High speed burr was used to remove the cortex to approach the tumor. After identification of the tumor, aggressive curettage was performed. Tumor removal was finished with high speed burr till reaching the cortex in all available/safe areas. In patients with stage 3 lesions, posterior pedicle screw instrumentation one level above and below the pathology was performed due to iatrogenic instability secondary to tumor removal.

RESULTS

Six patients (4 male, 2 female) with a mean age of 25 (16 – 49) years were included in the study. All patients had complete clinical and radiological follow-up data. Written informed consent was obtained from all participants before accessing patient data.

The mean follow-up was 77.5 (13.8 – 161.5) months. Details of surgical procedures performed were given in Table 1.

There was hypoesthesia and muscle weakness on right lower extremity in Patient 1, preoperatively. Muscle strength was 5/5 and there was no hypoesthesia during the last follow-up.

Patient 2 had localized pain for 5 years until diagnosis of ABC, she was treated successfully with simple curettage and bone grafting. There was no recurrence in the last follow-up.

Table 1. Patient demographics are given. Enneking staging for tumor and WBB classification were used for surgical planning. Physical examination findings and surgical procedures performed are given.

No	Age	Sex	Location	Enneking	WBB	Duration of Symptoms (year)	Physical Examination Findings	Surgical Procedure
1	22	M	L1 right pedicle & lamina	Stage 3	10 to 1, D-A	1	Hypoesthesia at L1 dermatome, right lower extremity global muscle power 4/5	1. Right pedicle excision 2. 2-level instrumentation
2	27	F	L3 left pedicle and corpus left side	Stage 2	2 to 4, C	5	No neurologic symptoms	1. Curettage 2. Bone grafting
3	17	F	L4 spinous process	Stage 2	12 to 1, B-C	4	No neurologic symptoms	Intralesional resection
4	16	M	S1-S4 left side	Stage 3	7 to 12, E-B	2	Left L5 & S1 radiculopathy, S2-3-4 anesthesia, left sciatica	Selective arterial embolization
5	49	M	T12 left pedicle & corpus left side	Stage 3	1 to 6, A-D	1	Left T11 radiculopathy	1. Left pedicle excision 2. Cementing 3. 2-level instrumentation
6	16	M	L1	Stage 3		3	No neurologic symptoms	1. Curettage 2. Bone grafting 2. Posterior instrumentation for scoliosis

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Patient 3 had back pain for a couple of years, and had been operated at another center for kyphosis, however her pain continued after posterior instrumentation surgery, and applied to our clinic. She was diagnosed with AKK at the spinous process of L4, simple en bloc resection was performed (Figure 1).

Clinical and radiologic evaluation of Patient 4 was a possible AKK of the sacrum. During preoperative anesthesia examination, hypercalcemia and hyperphosphatemia were detected, so he was referred to another center for the etiology of increased levels of calcium and phosphate. Therefore, SAE was used to control the tumor growth instead of surgical excision During the follow up, the patient was using two crutches for mobilization and was not able to bear weight on left lower extremity. Orthopedic oncology team examined and evaluated the patient: even though radiologic examination showed that ABC was not progressed, yet clinical examination indicated that S1 root compression and L5 stretching due to expansion of the mass anteriorly led to sciatica which resulted as knee contracture and patient could not bear weight on his left leg in last follow-up (Figure 2).

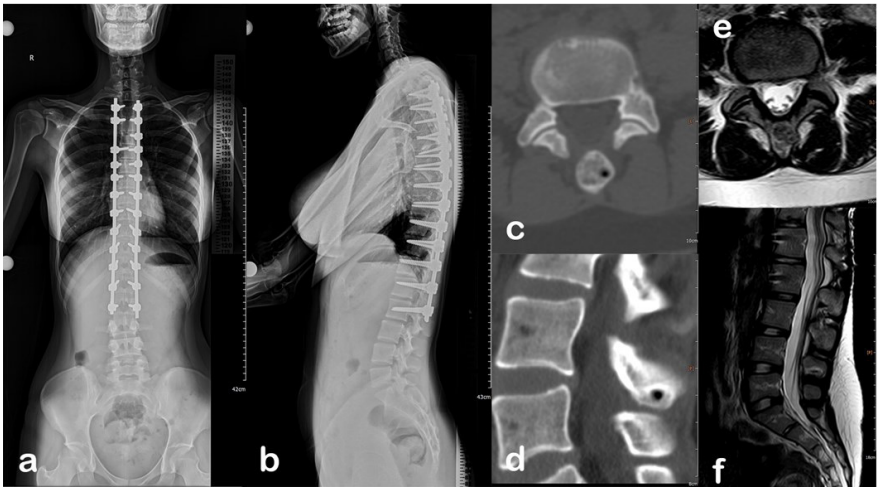


Figure 1. Female, 17 years old. She had complaint of back pain and operated with a diagnosis of (possibly postural) kyphosis at another center (a, b). Primary complaint of her back pain did not resolve after surgery and applied to our clinic. CT and MRI evaluation showed a benign lesion at the spinous process of L4 (c to f).

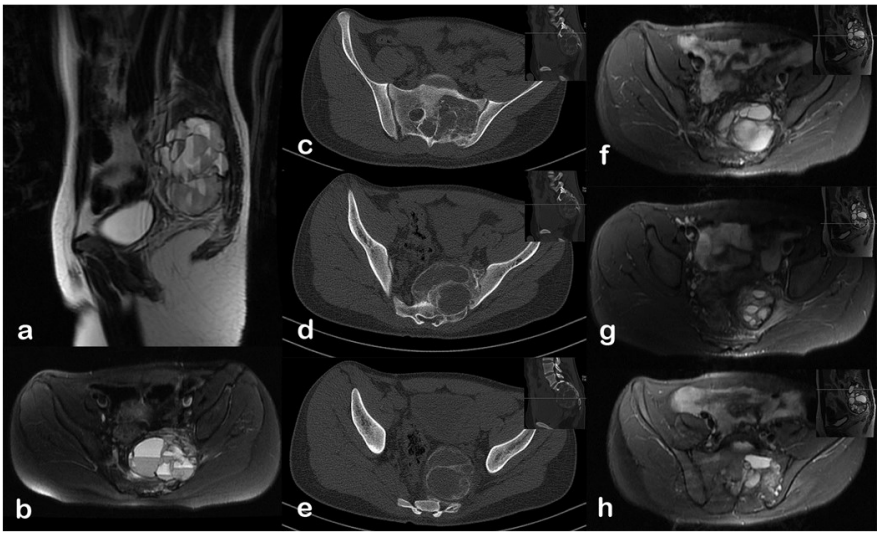


Figure 2. Male, 16 years old. MRI showed fluid-fluid levels on left side of his sacrum, suggesting ABC (a, b). After SAE, tumor growth was controlled, as seen on last follow up CT and MRI (c to h).

Resection was planned in this patient. Left T11 radiculopathy was present in Patient 5 due to foraminal canal occlusion (Figure 3). T11 radiculopathy was resolved in the 3rd postoperative month after surgical excision of the tumor.

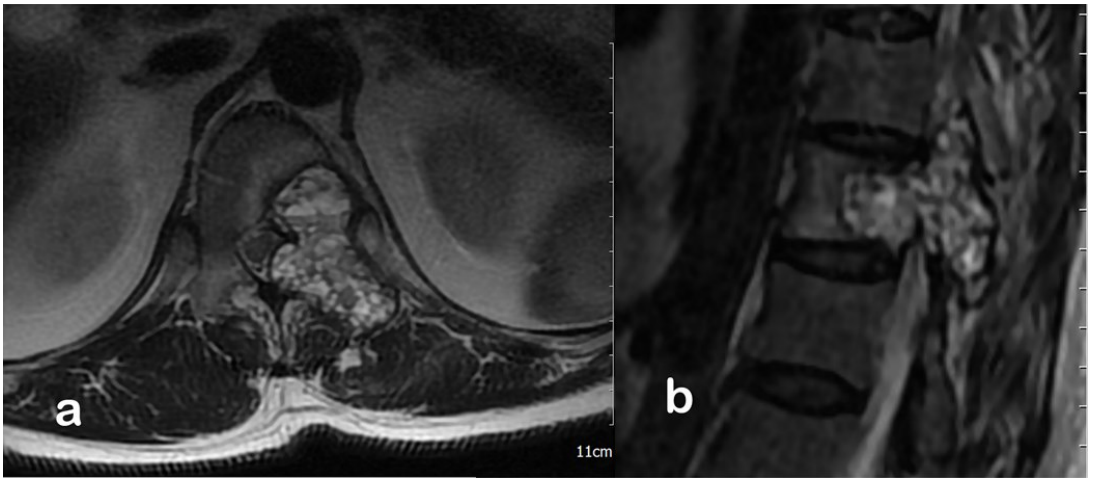
Scoliosis was diagnosed in Patient 6 at another hospital. He applied to our clinic for scoliosis treatment. Initial evaluation revealed that he had a localized pain at L1, and radiological studies showed the mass in the lamina. Simultaneous scoliosis surgery and ABC excision were performed. Even though there were failed screw and pseudarthrosis findings at the lower level of posterior instrumentation, he had no complaint of any pain, and MRI study showed no recurrence in the last follow-up. There was only one superficial infection, Enterobacter spp., treated with oral antibiotics. There were no other early or late complications in patients.

DISCUSSION

ABC is often a primary lesion but may develop secondary to other tumors such as Langerhans cell histiocytosis, chondroblastoma, giant cell tumors, or telangiectatic osteosarcoma. Our series include primary ABC cases, confirmed by histopathological evaluation. There are various treatment options for ABC of the spine. Spontaneous resolution of ABC can be seen on long bones, promoted or not by surgical biopsy (21). However, spontaneous resolution is rare in spine probably because most diagnosed ABCs in the spine are stage 2 or 3 lesions. There is no information about stage 1 lesions of the spine. Since stage 1 lesions may be asymptomatic, therefore patients do not apply to clinics, and are not diagnosed on X-ray incidentally, especially due to the complex anatomy of the spine.

Radiological evaluation includes X-ray, MRI, and CT. MRI should be considered as the gold-standard technique for radiologic evaluation, due to its excellent sensitivity on frequent aggressive presentation, with ABC presenting as large masses. Aggressive spinal tumors may show a T1 hypo intensity and a T2 hyperintensity with a low signal margin with enhancement. Fluid levels are present in only one out of three cases and are a non-pathognomonic sign but very suggestive of ABC. This finding is also a radiographic characteristic of telangiectatic osteosarcoma, giant cell tumor, secondary ABC, and fracture through a simple cyst. Since fluid levels may represent a metastatic malignancy (24), diagnosis of ABC must be confirmed prior to initiating definitive treatment (25). Biopsy was taken for histological confirmation of ABC in all patients, except Patient 5, who asked for single stage surgery. In this patient, the orthopedic-oncology council's decision about diagnosis was ABC or giant cell tumor, and frozen section was not available. Therefore, cement augmentation as adjuvant therapy was used instead of grafting to prevent recurrence in case of histopathologic study revealed giant cell tumor.

Figure 3. Male, 49 years old. Left pedicle and left side of corpus, with mild cord compression is seen on axial image (a). Tumor extention through left T11 foramen is seen on sagittal image (b).



In terms of the proposed protocol for spine ABC treatment, intralesional excision, "en bloc" resection, radiation therapy, SAE, and intralesional drug injection (steroid, calcitonin, denosumab or doxycycline) are all good choices (20). The aim of the surgical treatment of ABCs is to perform a complete resection of the cyst, intralesional or extralesional 'en bloc', to avoid recurrence. Since majority of the cases involve the posterior elements of the spine, posterior approach is the preferred method. If ABC has an extension to corpus, the vertebral body lesion is curetted after resection of the posterior elements. The choice of the surgical approach is challenging for extensive ABCs. The use of combined approaches for vertebral body lesions may be recommended in one or two distinct surgical procedures. Due to the vascularity of the tumor, SAE is helpful in reducing intraoperative blood loss and improving the visualization during surgery (4,9,10,12,26). However, surgical resection should be performed shortly following SAE to prevent revascularization of the lesion. SAE respects surrounding bone tissue and complications are rare, but it is essential to check that the Adamkiewicz artery is not in the embolized territory to prevent paraplegia secondary to medulla spinalis ischemia (10,20). Since embolization failure in terms of tumor growth control does not preclude surgical treatment, SAE was preferred over surgery in the patient with sacral ABC due to the comorbidities in our series even though patient had neurologic symptoms. SAE currently can be used as a standalone medical method to treat spinal ABCs in the absence of neurological symptoms or bone destruction responsible for spinal deformity which requires a surgical treatment (20,26).

En bloc resection in healthy tissue theoretically carries a lower risk of recurrence when compared with intralesional excision, it is also less hemorrhagic. However, en bloc resection is not always possible in spinal tumors without neurologic compromise or without increased morbidity due to proximity of major vascular structures, yet en bloc resection involving neurologic structure excision may be discussed with the patient and performed in select patients with local aggressive tumors of the spine (27).

Radiotherapy has been used and still used as a standalone treatment or in combination with surgery (2,5,9) but it is regarded as a major risk factor for radiation-induced sarcomas (1). Therefore, radiotherapy has not been used in ABCs of the spine in our clinical practice. Another treatment modality, intralesional injection of various drugs, such as steroid, calcitonin, denosumab or doxycycline have been proposed for control of tumor growth (7,10,17,18,20,28,29). We have been using denosumab and steroid injections in the ABCs of the extremities, yet not used in spinal cases. In patients without neurologic symptoms, and in the absence of deformity secondary to osseous destruction of the spine, injection treatment should be considered as a treatment option.

The occurrence of neurological symptoms with benign spine tumors is either because of vertebral canal or intervertebral foramen invasion or by external pachymeningitis. Spinal deformity, especially kyphosis with pathological fracture, may cause neurological complications. However, neurological complaints could be the first symptoms in ABC of the spine before pathological fractures, especially in Enneking stage 3 lesions, where tumor may invade the spinal canal in a short time interval. During initial diagnosis of patient 4, operation was planned, increased calcium and phosphor levels were detected during preoperative evaluation period, he was consulted to internal medicine at another tertiary center. He was diagnosed with a parathyroid adenoma in mediastinum, and thoracic surgeons excised the mass by video assisted thoracic surgery in that tertiary center. Arterial embolization was also done for controlling the ABC of the sacrum in the same hospital, and tumor was stabilized, and the decision to follow up was taken for him by the orthopedic department. However, he developed a sciatica and knee flexion contracture, where contracture release surgery for his knee was performed twice by using external fixator. When the patient re-applied to our clinic, patient was using two crutches, not able to bear weight on his left leg. Even though the tumor size was not expanded after embolization, clinical and radiological examination revealed that left L5 nerve stretching due to anteriorly expanded sacrum, and S1 compression in the sacral

foramen were the cause of sciatica. Therefore, patient and his family were offered the surgery for excision of the tumor with neural structure release to relieve sciatica pain to treat the recurrent knee contracture and to be able to use his left leg for mobilization.

In the study of Mankin et al, local recurrence rate was 20% (6). The lowest recurrence rate is seen with en bloc resection and risk factors for recurrence include young age (<12 years) (30–32), histologically predominance of cellular component over osteoid (33), and a mitotic index of ≥ 7 per 50 fields(34). There was no recurrence in our series. Development of sarcoma following radiotherapy is a well-known entity and transformation of ABCs to sarcoma secondary to radiotherapy has also been reported (1). Yet, there were case reports of ABC transformation to telangiectatic osteosarcoma and fibroblastic osteosarcoma without radiotherapy (35–40). Thus, all patients should be followed regularly for recurrent symptoms or a change of symptoms.

In conclusion, ABC of the spine is a benign, active lesion that may cause significant morbidity in adolescents and adults. The aim of the treatment is prevention or resolution of neurologic compromise, prevention of pathologic fracture or deformity development. Even though malignant transformation is exceedingly rare, patients should be followed regularly for recurrence of symptoms or a change of the symptoms. Excellent success rates have been reported with intralesional drug injection or SAE in patients with ABC, yet surgery should be reserved in cases with spinal deformity, or in the presence of neurologic symptoms.

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