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ANEURYSMAL BONE CYST WITH LUMBAR SPINE INVOLVEMENT: A CASE OF ATYPICAL GROIN PAIN MISDIAGNOSED AS AN INGUINAL HERNIA

LOMBER VERTEBRA TUTULUMU GÖSTEREN ANEVRİZMAL KEMİK KİSTİ - İNGUİNAL HERNİ İLE KARIŞAN ATİPİK KASIK AĞRILI BİR OLGU SUNUMU

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SUMMARY:

Aneurismal bone cysts (ABCs) make up 1.4% of all bone tumors and 15% of all primary spinal tumors. Some benign primary bone lesions, such as ABCs, are aggressive (such as osteoblastoma and giant cell tumor). The clinical symptoms can be confused with other pathologies if attention is not paid. In a 23-year-old male patient who received surgery due to suspicions of hernia as a result of pain in the inguinal region, a rare aggressive ABC with involvement of the lumbar 1 vertebral posterior neural arch was detected, with neurological symptoms.

The mass, which was causing compression of the posterior neural arch and medulla, was totally removed, and pedicular screw fixation was applied to one upper and one lower level. Postoperatively, an early full recovery was obtained, and no recurrence occurred at the end of a five-year follow-up. The results were clinically and radiologically improved.

Key words: Aneurysmal bone cyst, benign primary bone tumor of spine, aggressive bone tumors of the lumbar spine, differential diagnosis, surgical treatment.

Level of evidence: Case report, Level IV.

ÖZET:

Anevrizma kemik kisti (AKK) yaklaşık olarak tüm kemik tümörlerinin % 1.4'nü ve tüm primer omurga tümörlerinin % 15'ni kapsar. AKK gibi omurganın bazı benign primer kemik lezyonları agresif seyirlidir (Osteoblastoma ve dev hücreli tümör gibi). Klinik semptomları dikkat edilmezse başka patolojiler ile karıştırılabilir. İnguinal bölgedeki ağrı şikayeti için herni şüphesi ile daha önce opere edilen 23 yaşındaki erkek hastada, nörolojik semptomlarla birlikte Lomber 1 vertebra posterior nöral ark tutulumlu nadir rastlanan agresif AKK tespit edildi.

Posterior nöral ark ve medulla üzerinde bası oluşturan kitle total olarak çıkarılıp bir üst ve alt seviyeye pediküler vida fiksasyonu uygulandı. Postoperatif erken dönem tam iyileşme sağlandı, 5 yıllık takip sonucu rekürrensle karşılaşılmadı, klinik ve radyolojik olarak sonuçların çok iyi olduğu belirlendi.

Anahtar Kelimeler: Anevrizmal kemik kisti, omurganın primer kemik tümörü, lomber omurga benign agresif kemik tümörü, ayırıcı tanı, cerrahi tedavi.

Kanıt Düzeyi: Olgu sunumu, Düzey IV

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INTRODUCTION:

Primary bone tumors of the spine are rarely observed[14]. 90% of tumors involving the spinal column have a metastatic origin[30]. Primary ABCs are observed at a low rate of 0.14–0.32 in every 100,000 people[18, 24, 34]. ABCs make up 1.4% of all primary bone tumors[8]. They are mostly located in the femur, tibia, humerus and fibula regions, and have a limited effect in the metaphysis region of the bone. These locations represent about 52% of all cases[18]. Others are commonly located in the cranium and posterior elements of the spine, and are frequently seen in the first two decades of life. ABCs are the most common malignancies affecting the posterior elements of the spine (with osteoid osteoma and osteoblastoma)[26]. Involvement of the vertebral column is observed in 3–20% of cases[1,33]. It has been reported that this is caused by mobile vertebrae, often in pediatric cases and with a ratio of 10–30%[2]. Lesions are commonly located in the thoracic and cervical vertebrae, and most patients are male[25]. There are many theories about ABC pathophysiology, and vascular, traumatic and genetic etiologies have been suggested[5,19,24,27].

The treatment algorithm for spinal primary tumors should contain the complex relationship of neural elements and the recurrent tendency of these lesions. Some benign primary bone lesions of the spine, such as ABCs (osteoblastoma, giant cell bone tumor), are considered aggressive. ABCs are rarely observed in the spine, although despite the small number of patients there are case reports in the literature. A variable histology is present in aggressive benign bone tumors of the spine. Despite these differences, the anatomical surgical treatment should include the complete resection of the tumor[14].

Diagnosis of primary benign bone tumors of the spine cannot be always performed accurately. Due their rarity, diagnosis is difficult. An early and accurate diagnosis positively affects the success rate, especially for patients with aggressive ABCs. In our study, symptoms that developed due to a posterior neural arch-located aggressive ABC, and the clinical and radiological results of the treatment, were evaluated in a young male patient who was misdiagnosed due to confusion of the clinical symptoms with an inguinal hernia, and who received surgery for hernia repair. The differential ABC diagnosis and treatment options are discussed.

CASE PRESENTATION:

Lower back pain in a 23-year-old male patient was present for one year. He received surgery based on a diagnosis of an inguinal hernia due to worsening pain in the right inguinal region and spreading to the anterior of the femur. The patient then applied to our clinic after the second week postoperatively, with a continuing increase in complaints. Hypoesthesia in the region corresponding to the L1 dermatome, lower back pain when walking, neurogenic claudication in the right lower extremity, 4/5 reduction in muscle strength and palpation and sensitivity in the L1 vertebral spinous projection were observed. AP-LAT lumbar X-rays, computerized tomography (CT), magnetic resonance imaging (MRI), scintigraphic examinations and biochemical tests were performed. Biopsy, pathological examination and surgery were planned in the same session.

For this patient, with clinical signs similar to a lumbar disc hernia, a detailed radiological examination was performed due to the clinical localization of the pain. A posterior laminar

osseous pathology narrowing the foraminal exit was detected in a lateral direct X-ray (Figure-1). By CT and MRI, in addition to radiological signs, a lobule-contoured, 3.5 × 2.5 × 2.2 cm osseous lesion was detected, which showed levelling at the L1 vertebra right pedicle, transverse projection, facet joint, and lamina, and had bone cortex destruction, expansion and spreading to the inside of the spinal canal, and a compressed dural sac and right L1 and L2 nerve roots (Figure-2.a-c).

Figure-1. Lateral X-ray showing foraminal stenosis caused by tumoral tissue on the lamina.

After preoperative planning, pathological examination of the material taken from the affected posterior lamina region in the same session was performed with direct microscopy, and a benign primary bone tumor was detected (Figure-3). A posterior approach was applied between T12 and L2. A cortical defect was detected on the right lamina of the L1 vertebra. As planned preoperatively, the tumoral tissue expanding to the right lamina of the L1 vertebra, the spinous projection, a part of the right transverse projection and the medullar canal involving the right pedicle were carefully removed, without leaving any pieces in adjacent tissues. All posterior elements of the L1 vertebra, except the left superior facet joint and transverse projection, were carefully excised. It was observed that the tumor tissue aggressively spread to the right L1 root, like a thin sheath over the medulla spinalis. In addition to the pathological tissues covered by the sheath, all tissues of the L1 root were removed without damaging the neural tissues (Figure-4).

45 mm polyaxial pedicular screws with a 5.5 mm diameter were used in the T12 vertebra, and 50 mm polyaxial pedicular screws with a 5.5 mm diameter were used in the L2 vertebra (Figure-5.a-b). Autogenous grafts taken from the patient were placed on the left transverse projection to support fusion.

As mentioned in other studies in the literature, the pain in the right lower extremity and the neurological complaints of the patient disappeared after surgery[5]. He was mobilized on the second day postoperatively and discharged on day five, with a thoracolumbar corset to be used temporarily, for five months.

The pathology results were evaluated and an ABC was diagnosed. No recurrence, complaint about implant relaxation or complications were encountered in the patient after the fifth year postoperatively (Figure-6). The patient returned to his daily active life (Figure-7.a-c).

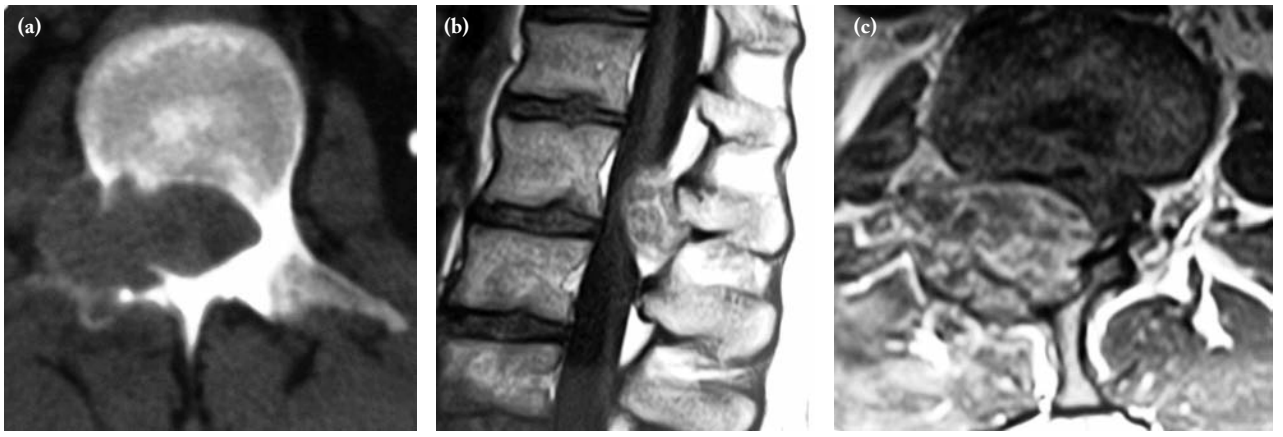

Figure-2. a. Damage of cortex inside canal by tumor tissue invading lamina and pedicle in CT, **b.** Compression of spinal cord by tumoral tissue in sagittal MRI. **c.** Involvement of L1 vertebra right pedicle, transverse projection, facet joint and lamina in axial MRI section.

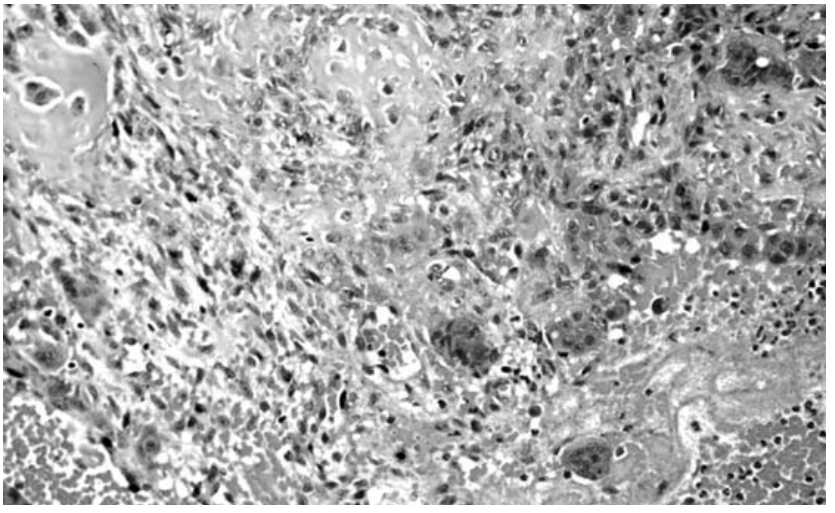

Figure-3. Pathology of ABC sample taken from lesion region (H.E.)

Figure-4. Spinal cord decompression during surgery and pathological tumor tissue on spinal cord

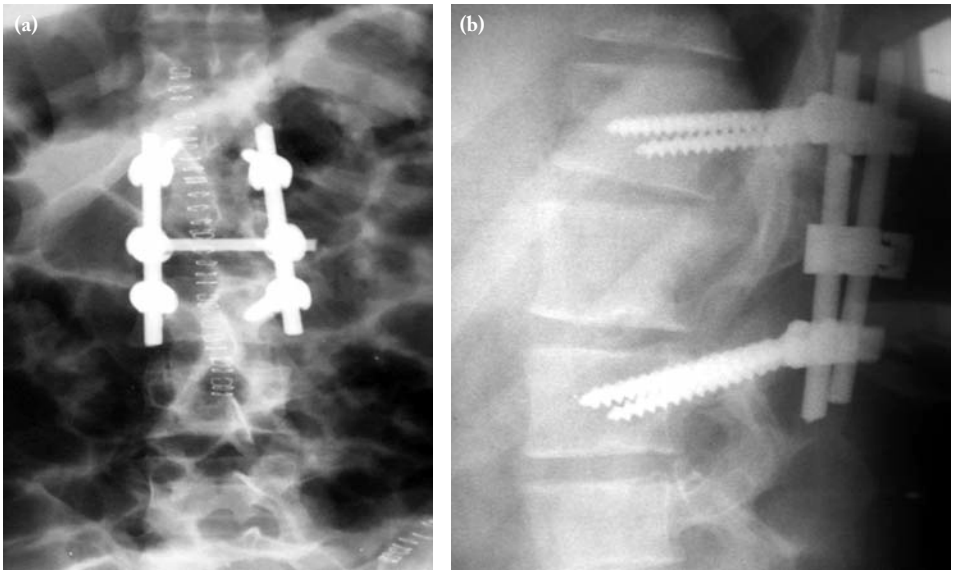

Figure-5.a. Postoperative AP X-ray, **b**. postoperative lateral X-ray.

DISCUSSION:

ABCs were first defined as a separate clinical and pathological form by Jaffe and Lichtenstein in 1942[16,21]. ABCs are accepted as non-neoplastic. They are a type of tumor that generally affects a pediatric population, which spread to their thin-walled environment and include cystic cavities filled with blood[14]. These bone lesions are benign and cause problems related to pain, swelling or spreading of the mass to itsenvironment[26].

They are generally observed in individuals between 1 and 59 years of age, with a median age of 12–13 years[18,34]. It is thought that about 70% of cases are primary lesions and 30% have originated secondarily from different primary tumors[7]. ABCs represent about 1.4% of all bone tumors and 15% of all primary spinal tumors[1,8]. Additionally, ABCs can involve any bone of the skeletal system, and often affect the flat bones of the pelvis and the metaphysis of long bones. About 10–30% of cases involve the spine, and thoracic and cervical vertebral involvements are mostly observed[25]. Generally, they emerge in the posterior elements of the spine. They result in pathological fracture and neurological complications by spreading to the pedicle, vertebral body, and the inside of the spinal canal. Rarely, some of these cysts become silent or spontaneous regression occurs. Progressive growth is the most common clinical scenario[14]. Before treatment, an accurate diagnosis of an ABC should be performed.

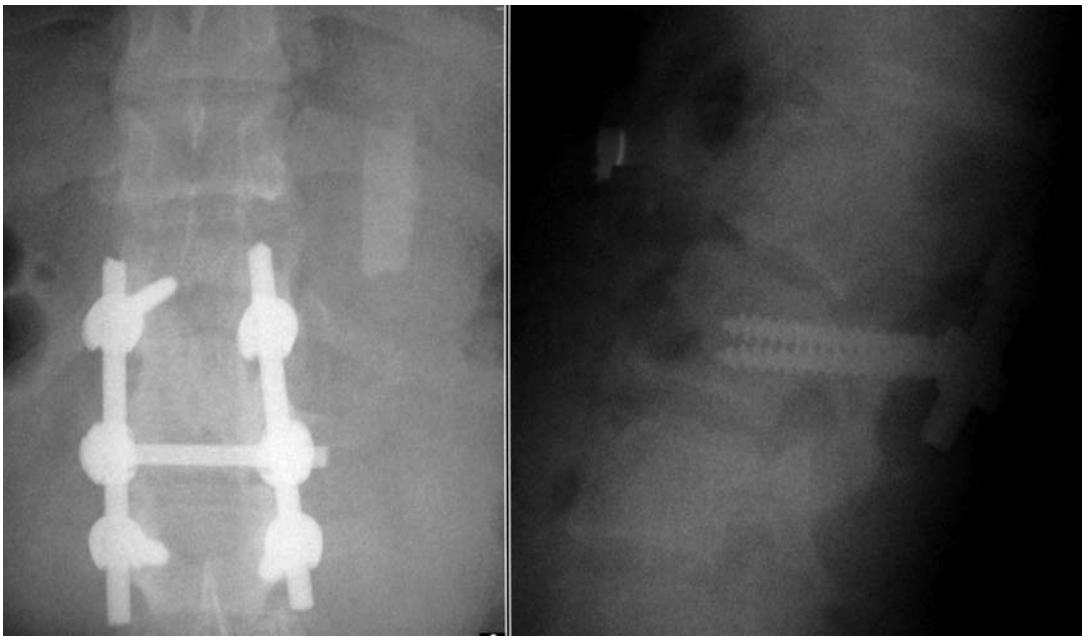

Figure-6. Postoperative last follow-up X-ray.

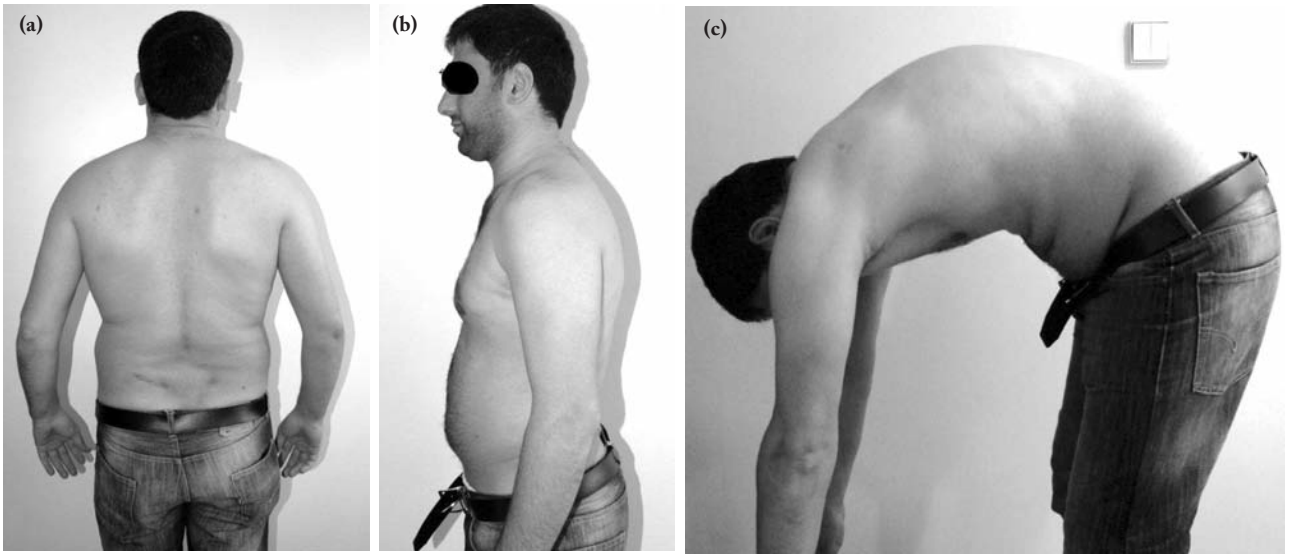

Figure-7.a. Posterior image of the patient in postoperative last follow-up, **b.** lateral image of the patient in postoperative last follow-up, **c.** bending forward in postoperative last follow-up.

After diagnosis, treatment generally includes intralesional curettage and bone grafting (with or without adjuvant treatment), large en bloc resection, preoperative arterial embolization, cryotherapy, sclerotherapy, radionuclide ablation, radiation therapy, or a combination of all these methods[1-2,4,20,26,28,32-33].

To categorize ABCs, various classification systems have been developed. Enneking divided lesions into three groups: inactive, active, and aggressive. An inactive tumor is the most benign tumor, due to its limited lesion.

It rarely spreads to the environment and causes less inflammation or periosteal reaction. Active

lesions typically cause mild symptoms such as pain, and are differentiated from a normal bone image in radiographies by cortical thinning and spreading, like a reactive bone layer. Aggressive tumors rapidly spread to their environment and damage adjacent tissues[12]. As in our case, the aggressive type is generally the most symptomatic kind.

A suspected ABC can be initially examined with a conventional radiograph. The classic radiological image of an ABC is a radiolucent cystic lesion in the bone metaphysis.

The effects of the lesion can be destructive and can spread into the adjacent bone tissue. The lesion can increase bone by a periosteal reaction, and the surrounding of the cortex can be limited with a thin shell. CT can be used preoperatively to better define the bone borders. Characteristic fluid-fluid levels of solid blood layers found inside the cystic areas of the lesion can be observed in contrast-enhanced MRI. This sign is quite valuable for diagnosis of ABCs, but is not pathognomonic, as it is also present in telangiectatic osteosarcoma, giant cell tumors, secondary ABCs and simple bone cyst fractures. Mahnken et al. found that MRI is superior to conventional radiography in terms of specificity. However, sensitivity, specificity and positive predictive value become higher when both are used together[22,26].

In young adult patients, the main reason for commonly-observed lower back and leg pain can be hard to determine. The problem needs to be detected by evaluating clinical and basic radiological examinations together, and then adding specific examinations when necessary[2].

In AP radiography, evaluation of the pedicles is routinely performed for every patient. Insufficient physical examination and missing radiological and clinical examinations resulted in surgery of our patient for another reason, and the diagnosis and treatment of the main disease were delayed. CT is the best imaging method for diagnosis, as the fluid level can be observed. Epidural spreading is seen well by MRI. Malign degeneration is not observed and the recurrence rate is about 20–30%. If there is incomplete excision, this ratio increases[25]. Embolization, surgical excision and instrumentation for spine stabilization can be required in treatment. Radiation therapy is not recommended because it can cause sarcoma.

The best treatment for ABCs is still controversial. The clinical progression of ABCs cannot always be predicted, and it has been shown that local recurrence can occur with various treatment types[2]. It seems that the recovery ratio is high with complete excision or en bloc resection. A spinal diagnosis provides correction of deformity and instability in addition to a high ratio of local control and protection[2,10,14,25]. Complete resection of an ABC should include all abnormal tissues, all cyst walls considered to be spongious, and all bone surfaces covered with fragile and hypervascular membranes[2-3,10,20,25]. It is recommended to reach healthy bone tissue by performing aggressive curettage with high-speed drills[14]. For large and common lesions, fusion with instrumentation is required because complete resection may result in iatrogenic instability[14,20]. In our case, complete excision and posterior instrumentation were applied to avoid iatrogenic instability, due to the presence of the ABC in the thoracolumbar region and the need for wide resection.

In the last radiological imaging, MRI and scintigraphic examinations of our patient, no

recurrence or relaxation of the instrumentation were observed.

In a large study with total resection by an intralesional technique and without radiotherapy, no local recurrence was observed in any patients[1-2,10,15,25]. The latest study by Boriani et al. reported no recurrence in 13 patients treated with complete resection intralesional curettage without radiation[2]. In a study in which incomplete excision was applied with curettage, about 25% local recurrence was reported two years after treatment[14-15,33]. Boriani and Hay reported local progression due to incomplete curettage in one out of five patients and seven out of 28 patients, respectively[2,15]. It has been reported that 90% recurrence due to incomplete excision was observed 6–12 months after surgery[2,25]. Recurrence of ABCs is unusual after two years and rare after four years[31]. If complete excision is performed, the possibility of recurrence is quite low[6,33]. In a study by Garg et al. that evaluated spinal ABCs retrospectively, while eight patients had no recurrence after surgical techniques including intralesional curettage, electrocautery, high-speed burr and bone graft, four recurrences were reported in four patients that were treated with only curettage and bone graft [(13)]. In our case, total resection surgery was a good option due to the involvement of the lesion in the posterior vertebral elements.

Local progression has been reported in ABC patients treated with radiation therapy after surgery and biopsy without significant surgical excision, and in ABC patients treated with only radiation therapy [(2,15)]. If the ABC is associated with pathological fracture, spinal deformity, instability, or neurological disorder, radiation therapy should be considered[14].

Complications of radiation therapy are possible growth disorder and post-radiation myelopathy[2,19,29,31,33]. Papagelopoulos et al. reported a post-radiation osteosarcoma in one case, seven years after surgery[25]. Despite this, radiation therapy is commonly used as an extra treatment after incomplete resections. No recurrence was reported after treatment[1,2]. As a result, radiation therapy provides supportive treatment for patients with inoperable lesions, high surgical risks due to aggressive recurrence and medical conditions, or incomplete excision, and it has limited primary indications. However, therapeutic embolization treatment should be considered before radiation therapy, due to the risks of radiation[9,11]. Therefore, it was not considered to be a treatment option in our case.

To reduce intraoperative bleeding, embolization can be primarily applied as selective arterial embolization (SAE) and/or direct lesion injection before surgery[9,11]. It has been reported that preoperative embolization reduces intraoperative blood loss[11]. Although successful treatments with embolization have been reported for ABCs of the pelvis and long bones, its role in treatment in the spine is quite limited. Recurrence has been reported in patients with SAE[2]. Embolization cannot be applied to small and peripherally located tumors. For lesions located in the thoracic or upper lumbar vertebrae, a careful approach is necessary, because accidental embolization of the Adamkiewicz artery can cause cord ischemia and anterior cord syndrome. Lesions that include the spine require discussion with an experienced spinal surgeon[26].

After a five-year follow-up, no instability, tumor recurrence or pain were observed in this young adult patient treated with posterior stabilization

after total excision of the tumor in order to avoid the development of instability in thoracolumbar junction region. Surgical total resection gave a sufficient result and there was no need for extra treatment. The patient satisfaction was quite high.

In the literature, it has been reported that ABCs can be observed with groin and lower back pain clinically, and cases have shown the involvement of the pubis and proximal femur[17,23]. As far as we know, no other case in which confusion resulted in surgery for an inguinal hernia instead of an ABC in the lumbar region has been reported previously.

As a result of treatment, the patient returned to his job and was satisfied with limited surgical excision and stabilization. No recurrence, implant failure, sagittal instability in the thoracolumbar region or pain were observed in the early-term follow-up. This case study shows that lumbar region involvements of aggressive ABCs can be confused with disc hernia and inguinal hernia, and so caution is required.

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