# Combined Percutaneous and Endovascular Treatment of Symptomatic Aneurysmal Bone Cyst of the Spine: Clinical Six Months Follow-up of Six Cases

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**Key words:** aneurysmal bone cyst, percutaneous treatment, sclerotherapy

**SUMMARY** – *We describe the usefulness of endovascular and direct percutaneous treatment as a therapy option for aneurysmal bone cysts (*ABCs*) of the spine. From January 2007 to December 2008, we treated six consecutive patients with symptomatic ABCs resistant to continuous medical management or with acute clinical onset of paraparesis at cervical, thoracic and lumbar spine level. Two patients were treated after emergency laminectomy. All patients were studied with an MRI protocol and multidetector CT with MPR reconstructions followed by angiographic control before treatment. The procedure was performed under general anaesthesia for all patients. Under CT or fluoroscopy guidance, percutaneous treatment was performed either by direct injection of Glubran® diluted at 30% with Lipiodol® only, or combined with endovascular treatment by Onyx® injection. Clinical and X-ray follow-up was performed at three and six months. Combined endovascular and percutaneous treatment for ABCs was successful and led to an excellent outcome in five out of six patients with clinical improvement. There were no periprocedural or subsequent clinical complications and the glue resulted in successful selective permanent occlusion with intralesional penetration. Direct sclerotherapy resulted in immediate thrombosis of the malformation with no progression of symptoms. Complete healing was observed in five out of six aggressive lesions. No major complications were noted. At six month follow-up the symptoms had completely resolved and X-ray control showed a partial or total sclerotic reaction of the lesion with stable clinical results (no partial or clinical abnormalities). One patient had a recurrence of the ABC with spinal cord cervical clinical symptomatology. Combined endovascular and percutaneous treatment or direct percutaneous sclerotherapy with glue alone are important, safe, effective therapy options for symptomatic aneurysmal bone cyst. Results are stable and confirmed by clinical and X-ray follow-up six months after treatment.*

## Introduction

Aneurysmal bone cyst (*ABC*) is a pseudotumoral hyperaemic-haemorrhagic, expansive osteolytic lesion with a thin wall, containing blood-filled cystic cavities. Although benign, ABC can be locally aggressive and can cause extensive weakening of the bony structure and impinge on the surrounding tissues. Malignant transformation is extremely rare [1].

The lesions predominantly afflict children, with 60% of patients younger than 20 years, with a female prevalence [2-6]. The true aetiology remains unknown and ABCs can emerge in virtually any bone in the arms, legs, trunk or skull [7]. The long bones of the lower extremities are most often afflicted with the tibia and femur being affected in 24.7% and 17.3% of cases respectively, followed by the upper extremities (10%) and the pelvis (9%). About 14%

of all ABCs are encountered in the spine, with those in the cervical spine making up only 2% [8]. The expansive nature of the lesions can cause pain, swelling, deformity, disruption of growth plates, neurologic symptoms (depending on its location), and pathologic fracture [8-9].

Sometimes ABCs are asymptomatic, but generally give rise to pain resistant to continuous medical management. Occasionally ABCs can be found during examination performed for other reasons or cause acute onset with spinal cord syndrome.

Treatment of spinal aneurysmal bone cysts is controversial. The traditional surgical procedures are curettage and wide resection but they are frequently complicated by kyphoscoliosis and compression of the spinal cord and nerve roots. Deformity is frequently worsened by the surgical procedures often needed to control these lesions, which can involve more than one level [10]. Haemorrhage or pulmonary embolism may riase as complications during or after surgery [9,11-14]. Other treatment strategies have included radiation therapy, sclerotherapy and endovascular treatment [15-16]. Percutaneous intralesional injections represent a relatively new mini-invasive therapeutic option for the treatment of ABCs, sometimes combined with surgical or endovascular treatment especially for large and resistant lesions, or in combination with surgical therapy [17-19].

The aim of our study is to describe the usefulness of the combined endovascular and direct percutaneous treatment as a therapy option for ABCs of the spine, without any major or minor complications.

## Material and Methods

Approval was given by the hospital medical ethical committee for this therapeutic procedure and retrospective collection of data and analysis at a later date. Informed consent was obtained from all patients. From January 2007 to December 2008, six consecutive patients (*three males and three females, mean age 15 years*) were treated for ABCs of the spine by means of a combined endovascular and percutaneous treatment or direct percutaneous sclerotherapy alone. Four out of six patients with ABCs were symptomatic and resistant to conservative medical management, while two out of six had acute onset with acute paraparesis and needed emergency surgery.

The lesions involved the cervical, thoracic and lumbar levels of the spine with this distribution: the sacrum in one patient, in the arch and body of L4 in two patients, at T9 in one, at L3 in one and in the arch and body of C7 in one patient. Two patients were treated after emergency laminectomy (EL). Three patients were treated by combined endovascular and percutaneous treatment, whereas the other three had percutaneous sclerotherapy alone (figure 1-3). Choice of treatment was based on morphology, vascular anatomy and distribution, vertebral level and size of the lesion.

*Patient selection-exclusion criteria*

Patients were excluded from treatment if they had:

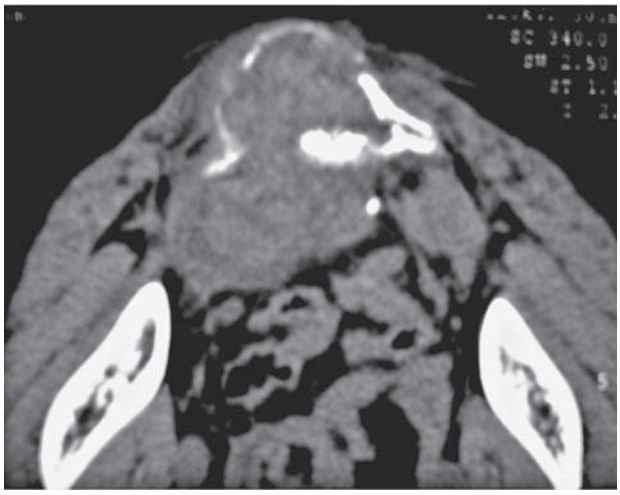
- symptomatic ABC with neurological impairment, spine instability, pathological fracture or spinal deformity
- venous drainage after contrast injection on CTA
- non correctable coagulation disorders
- greater bony destruction
- skin infection.

The six patients were divided into two groups (table 1, patient's list). In group A the procedure was carried out by percutaneous sclerotherapy alone, whereas in group B we performed both percutaneous and endovascular combined treatment. All the procedures were performed under general anaesthesia. Percutaneous sclerotherapy was performed by direct injection of Glubran® diluted at 30% with Lipiodol® generally under fluoroscopy or CT guidance. Endovascular embolization was performed by Onyx® (Onyx Liquid Embolic System) injection under fluoroscopic control also when it was combined with direct percutaneous treatment

*Imaging*

All patients were studied with both MRI and multidetector CT (MDCT), also with contrast injection to provide useful information in examining a possible ABC. Preoperative MDCT examination with MPR was helpful for defining the location and extent of bone destruction and the integrity of the cortical bone, the presence of pathologic fracture, partial or complete vertebral body collapse. CT angiography was performed to define the vascular structure. We performed an MR examination for a proper evaluation and delineation of the contents and the full extent of bone, soft-tissue involvement, including assessment of potential compromise of neural elements.

No bone biopsy under CT or fluoroscopic guid-



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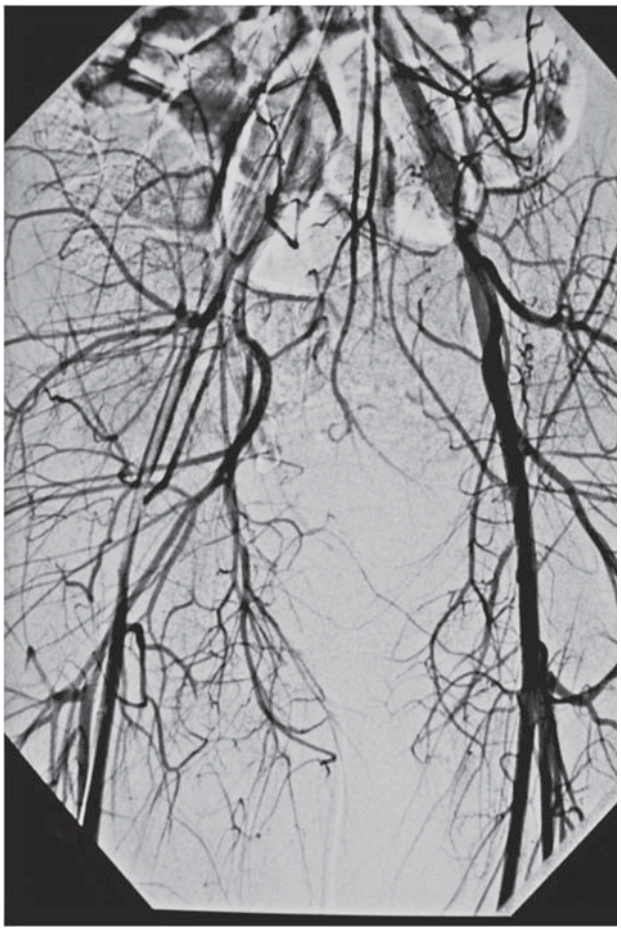

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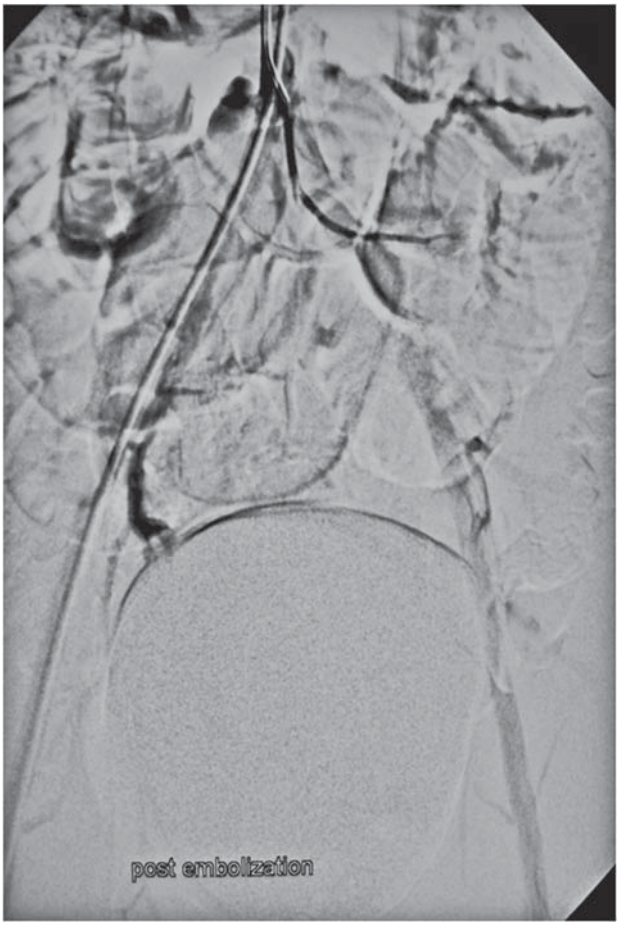


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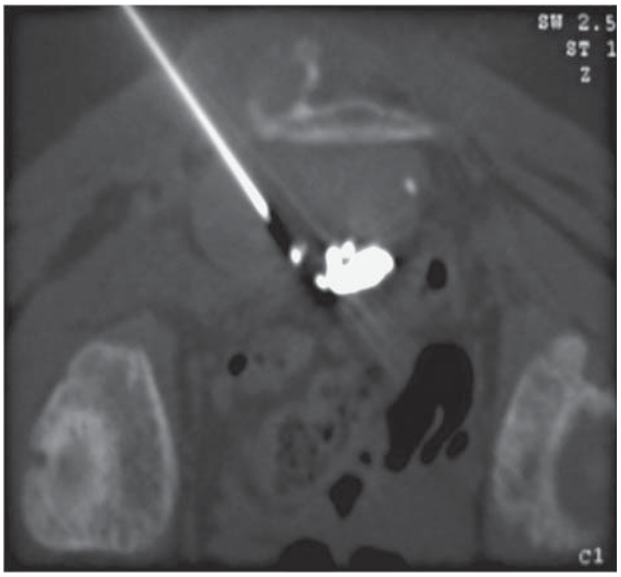


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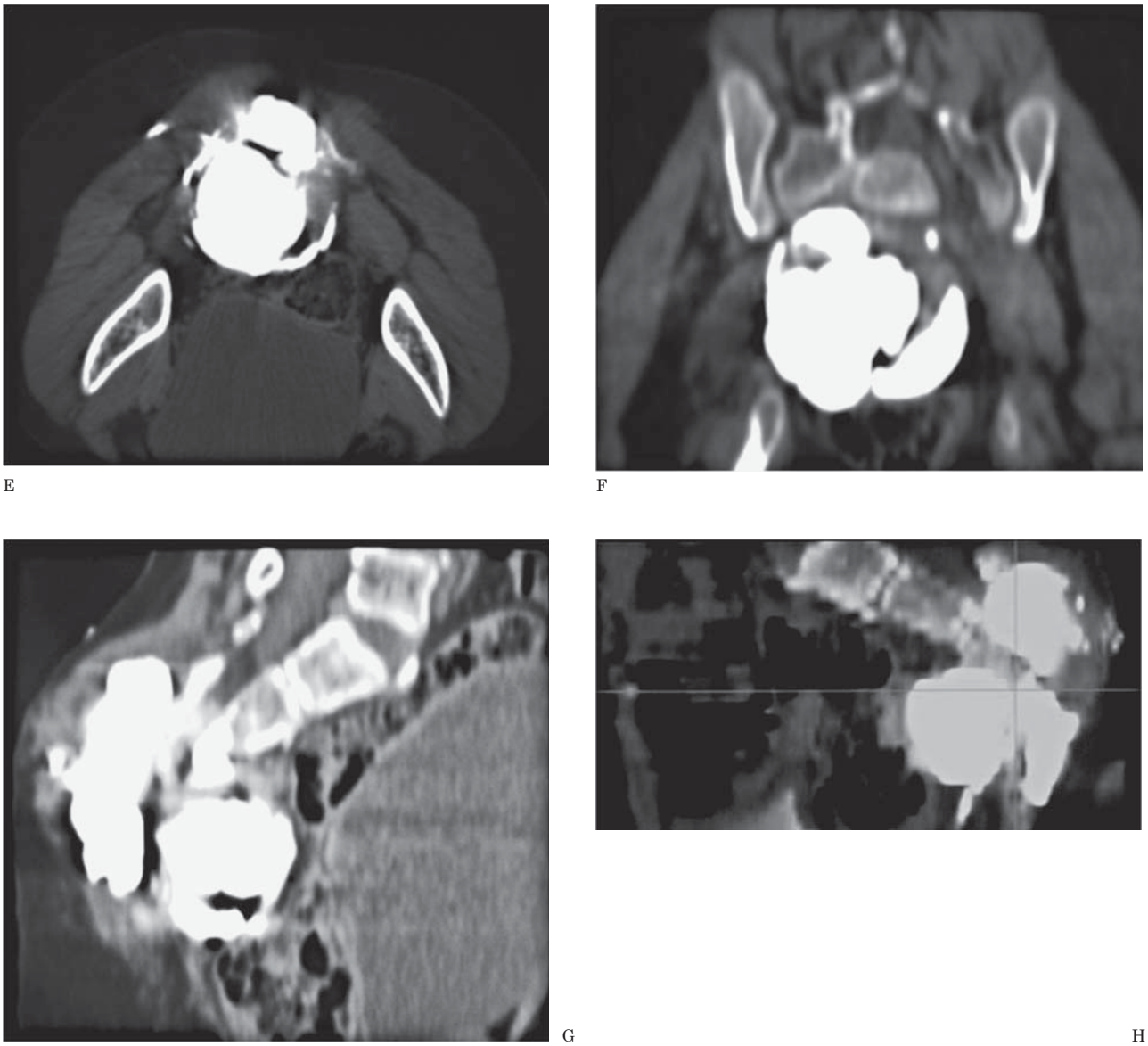

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Figure 1 11-year-old female. Pain and mass in lumbar and presacral region. A) Axial CT shows a large lytic lesion of the sacrum bone. B) Biplane angiography shows a vascular blush of the ABC. C) Angiographic control post embolization. D) Percutaneous direct sclerotherapy under CT guidance. E) Final CT control after percutaneous sclerotherapy. F) Coronal MPR-MDCT reconstruction after direct sclerotherapy. G) Sagittal MPR-MDCT reconstruction. H) Sagittal reconstruction at 1 year follow-up shows a stable result of sclerotherapy-endovascular combined treatment.

ance was performed to determine the nature of the lesion because this was already ensured by morphologic studies and MRI features.

An angiographic control was performed before treatment to evaluate feeding arteries, aneurysmal bone cyst vascularity, occasional arteriovenous shunt and venous drainage. No radioisotope bone scans were performed.

## Technique

All the procedures were performed under general anaesthesia by interventional neuroradiologists. The amount of diluted Glubran 30% varied from 2ml up to 27ml in one case, at sacrum level. The needle used for percutaneous injection was always a 16G. The amount

**Table 1** Patient characteristics

| *Case* | *Group* | *Sex* | *Age (years)* | *Symptoms* | *Localization* | *Treatment* |
|---|---|---|---|---|---|---|
| 1 | B | F | 10 | Pain and mass in lumbar region | Sacrum | Endovascular + percutaneous treatment + EL |
| 2 | B | F | 22 | Pain and mass in lumbar region | Posterior arch of L3 | Percutaneous + Endovascular treatment |
| 3 | A | F | 9 | Acute paraparesis | T9 | Percutaneus sclerotherapy + EL |
| 4 | A | M | 17 | Cervical pain and paresthaesias | Arch and body of C7 | Percutaneous sclerotherapy |
| 5 | B | M | 13 | Pain and mass in lumbar region | Arch and body of L4 | Endovascular + percutaneous sclerotherapy |
| 6 | A | M | 19 | Swelling | Arch and body of L4 | Percutaneous treatment |

of Glubran or Onyx® injected varied depending on the extension of the lesion and vascular structures.

In group A the procedure was carried out by percutaneous sclerotherapy only injecting Glubran® diluted at 30% with Lipiodol®, generally under fluoroscopic or CT guidance due to small arterial feeders. For better flow control injection was performed directly and slowly into the vertebral body malformation by multiple microinjections. In group B, we performed both percutaneous and endovascular combined treatment based on vascular intralesional structures with many arterial feeders. Patients were evaluated after treatment at three and six months by X-ray control and clinical assessment using the VAS scale.

## Results

All the results were evaluated before and after treatment by two other general radiologists up to six months follow-up, comparing clinical data, imaging and the VAS scale before and after treatment. Patients were evaluated after treatment at three and six months by conventional radiography and clinical assessment.

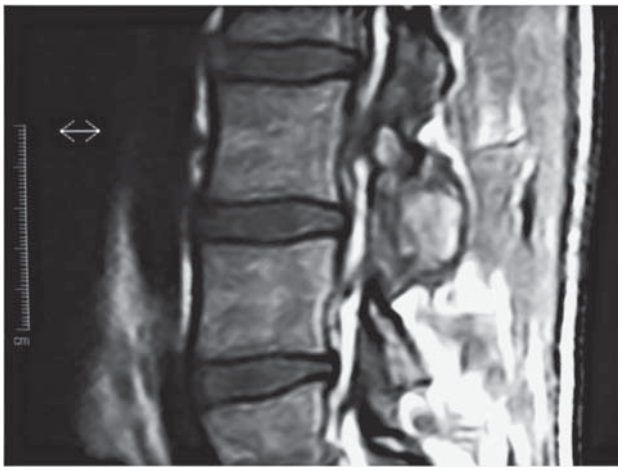
A successful outcome was observed in five out of six patients 24-72 hours after treatment, with a partial or complete resolution of pain caused by a thrombosis of 80% of the volume of the malformation, recording a four point reduction in the VAS evaluation. At six month follow-up, results remained stable with no complications or pathological fracture in five out of six patients. The patient with ABC at C7 level needed surgical spine stabilization after four months follow-up.

## Discussion

Aneurysmal bone cyst (ABC) is a pseudotumoral hyperaemic-haemorrhagic, expansive osteolytic bone lesion that most often affects individuals during their second decade of life and may occur in any bone in the body. ABCs consist of blood-filled spaces separated by fibrous tissue containing multinucleated giant cells. This finding is demonstrated by CT scans and MR imaging, which show separation of the blood content forming levels of different fluid densities. However, the fluid-fluid signal is not exclusive to ABCs, and can be seen in other bone lesions such as chondroblastoma, giant cell tumour and osteoblastoma [20].

Histologically, the cyst appears like a "blood-soaked sponge" containing blood with walls consisting of histiofibroblasts, giant multinucleated cells, thin capillary vessels, granules of hemosiderin and immature bone trabeculae. The septations of the lesion are hypervascularized and the venous injection of contrast media on CT or MRI can help to differentiate the thin and smooth septa of ABC from other tumours, which would have nodularities [21].

Radioisotope bone scans can occasionally



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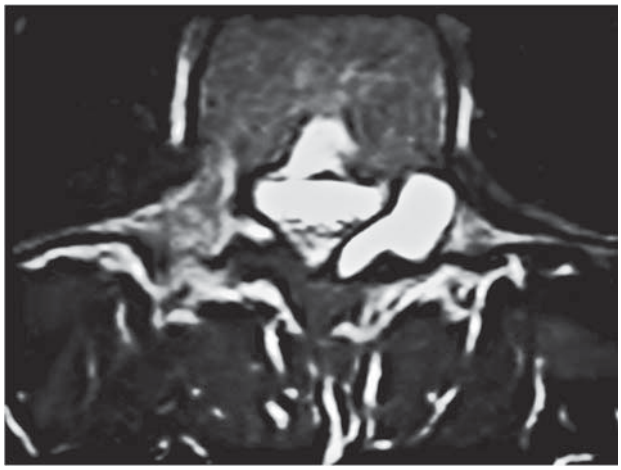

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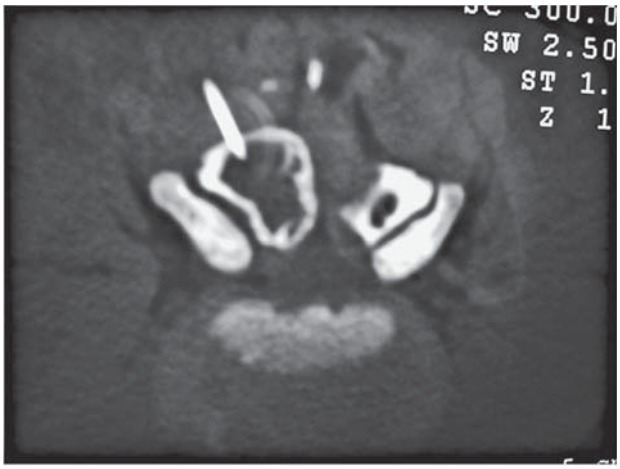


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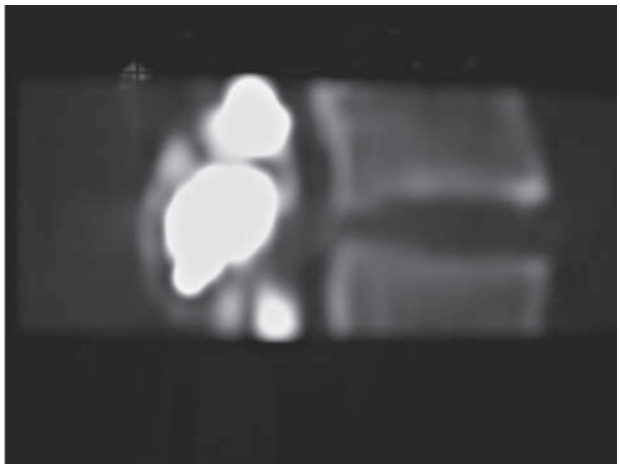

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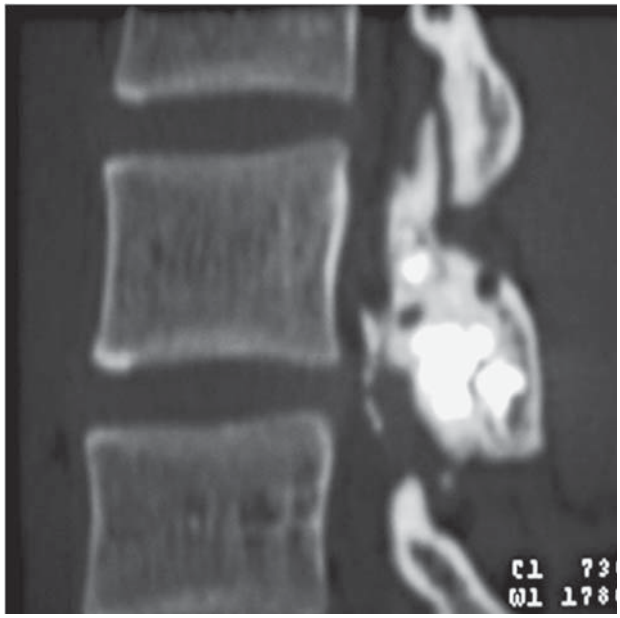

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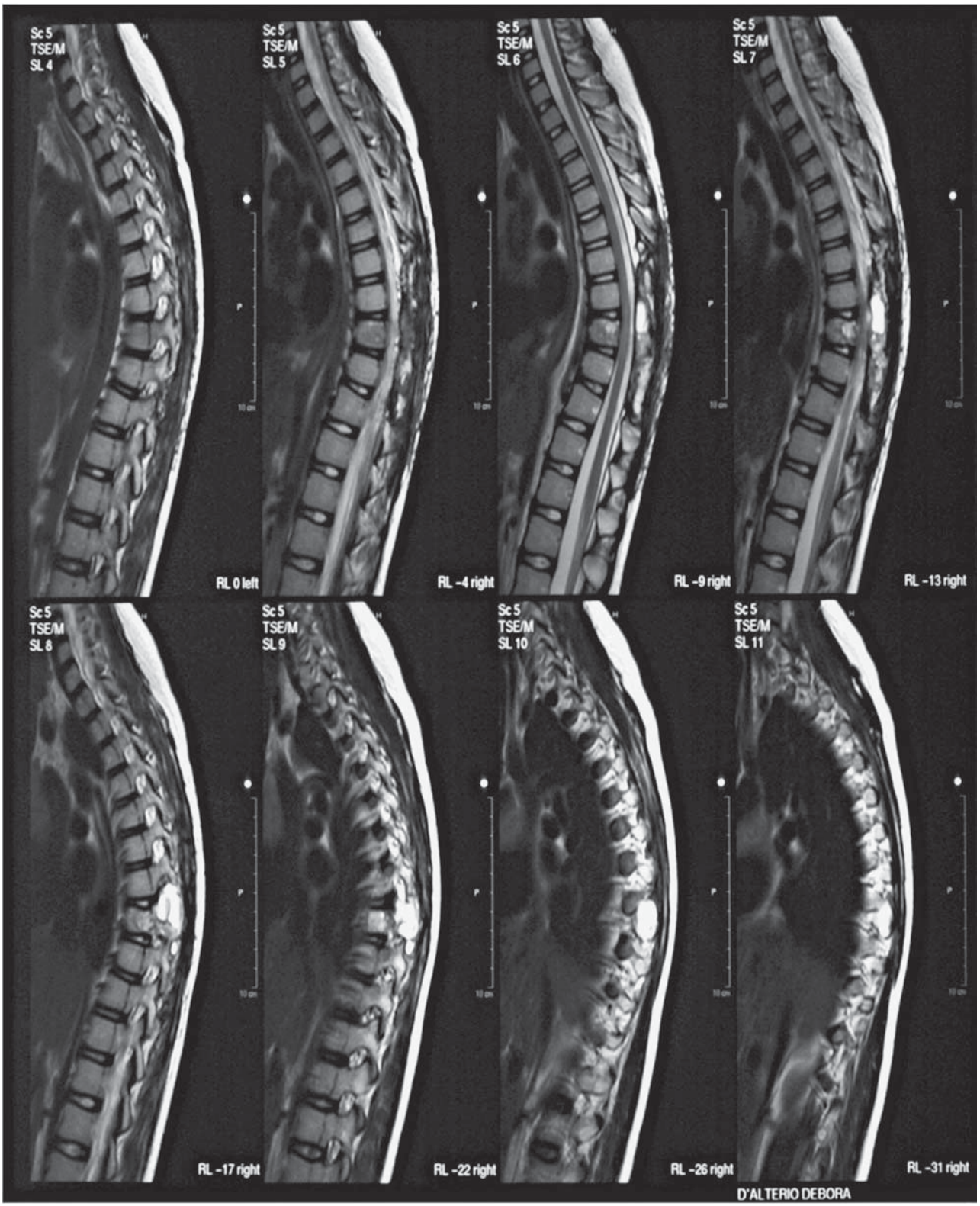
Figure 2 20-year-old male with lumbar pain more localized at L4 level. A,B) MRI axial and sagittal T1W + contrast shows a lytic lesion with blood level inside the posterior arch of L4. C) Direct sclerotherapy under CT guidance D) Sagittal MPR-MDCT reconstruction after direct sclerotherapy. E) Sagittal MPR-MDCT reconstruction after 1 year follow-up shows a stable result.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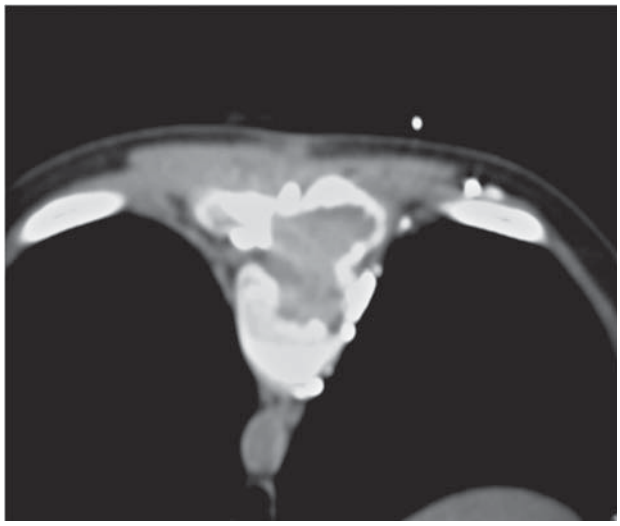

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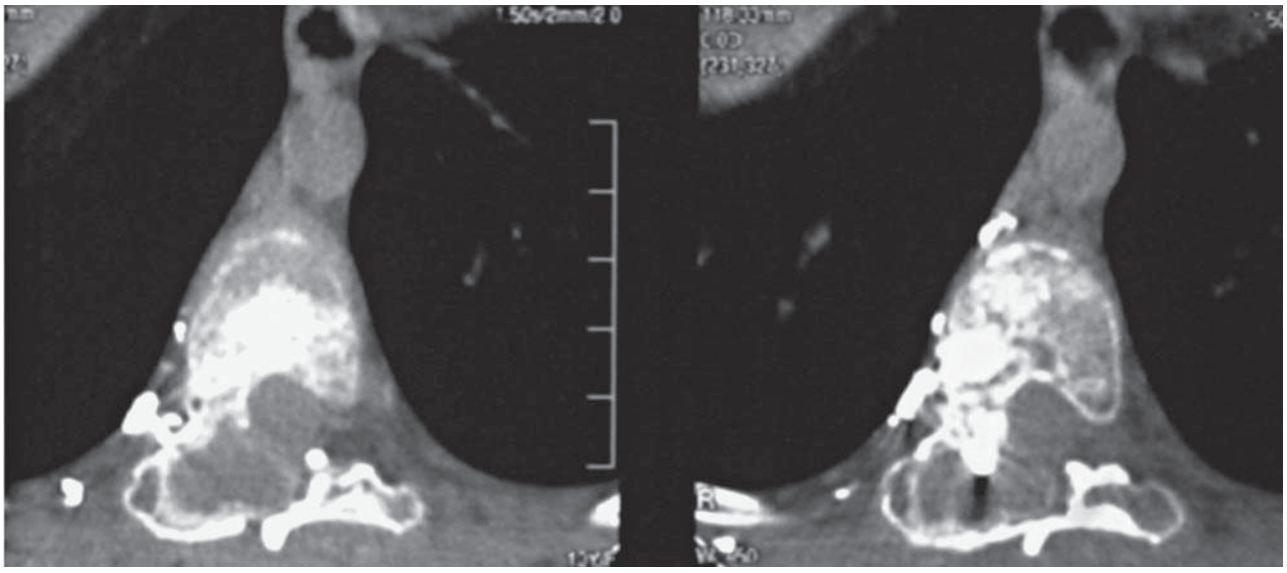
Figure 3 9-year-old female admitted in emergency for acute paraparesis. Partial decompressive laminectomy was performed on admission. The patient improved and an endovascular and percutaneous treatment was suggested. A) Sagittal T2W MRI shows a lytic lesion at T9. B) Axial CT shows a large expansive lytic lesion at T9. C) CT control after direct sclerotherapy of the lytic lesion. D) Sagittal MPR-MDCT reconstruction: final control after percutaneous sclerotherapy.



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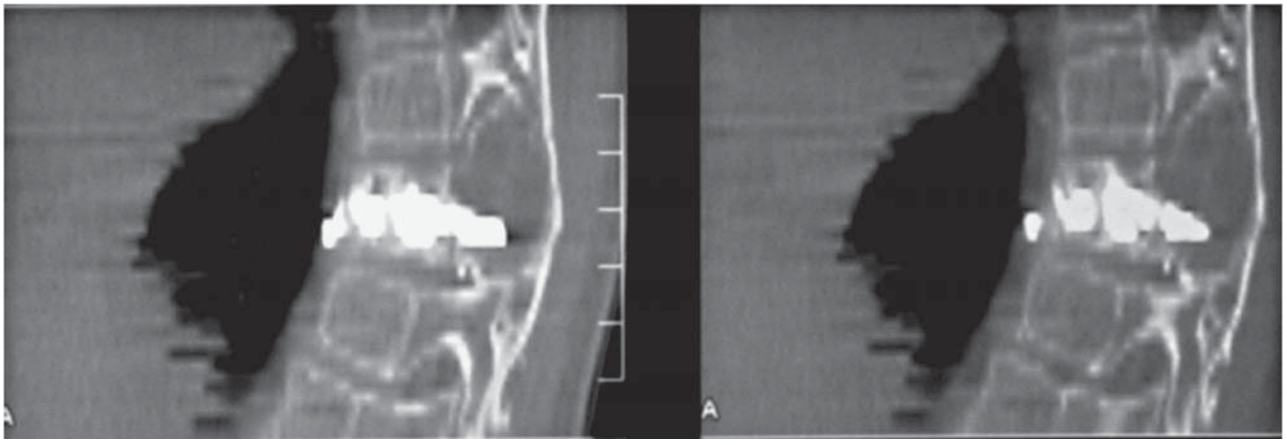

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show increased uptake of tracers in ABCs, and selective angiography can sometimes reveal the blood supply to these vascular lesions and may also occasionally reveal arteriovenous shunts [2].

In 1942, Jaffe and Lichtenstein first described ABC as a distinct nosologic entity in two young men (17 and 18 years old) with lesions located in the pubic bone and at the level of C2. They believed that many cases of ABC might have been misdiagnosed as either benign or malignant bone lesions [14].

The true aetiology of ABC remains controversial [2]. However it is believed that the ABCs may be primary, if no underlying lesion is present, or secondary to be distinguished from similar bone tumours such as fibrous dysplasia, chondroblastoma, giant cell tumour or osteosarcoma [5]. The lesions are more common in patients in the first two decades of life rather than in later years and seem to be slightly more frequent in females than males. ABCs have an overall incidence of approximately 1% of primary bone tumours [4,22].

The most common sites are the metaphysis of the long bones and the spine [23]. Within the spinal column, almost 70% of cysts are seen in the thoracolumbar region, and less than 25% are seen within the cervical spine [22]. More than one vertebral level is often affected. Among aneurysmal bone cysts, 60% occur in the pedicles, laminae and spinous processes [8].

The bone cyst can cause destruction of bone and isolated symptoms at the site of the lesion. The most common problem that a bone cyst will cause is weakening of the bone. This may lead to increased susceptibility to fracture at that location. Others symptoms are pain, swelling and onset of neurological problems, while sometimes the ABCs are asymptomatic [7]. Spontaneous regression of the lesions is uncommon [24-25].

For vertebral lesions, the choice of treatment must take into consideration the risk of neurologic and vascular lesions, and it must preserve spine stability and, if possible, spine mobility [7]. Treatment is controversial. Many treatments have been described including surgical procedures (*curettage and wide resection with intralesional excision, or "en bloc" excision*), radiation therapy, selective arterial embolization, and sclerotherapy. Sometime these treatments can be combined.

Surgical treatment of spinal aneurysmal bone cysts, which can be highly vascular, is technically considered to be the treatment of choice for these lesions including resection and curettage and spinal fixation. These procedures carry the risk of significant blood loss, postoperative spinal deformity, axial deformity, postoperative haemorrhage [26]. Some authors proposed a simple intralesional excision with bone grafting, while others proposed "en bloc" excision of the vertebra involved as the only treatment free from the risk of local recurrence [7].

Marcove et Al and Cole reported a post-surgical recurrence rate of more than 50% after curettage with or without bone grafting, while Schreuder et Al found 30.8% after curettage and bone grafting [27-30].

Radiotherapy has been used in the past, but this treatment is generally contraindicated because of the risk of sarcoma induction, gonadal damage and growth-plate disruption. Much risk is associated with treating a benign lesion with a therapy that can have damaging adverse effects, although radiation therapy is still occasionally used at low doses to treat surgically inaccessible lesions [11].

Another option is selective arterial embolization with a recurrence rate not statistically different from that after intralesional excision. The risk of spinal cord ischaemia caused by arterial embolization remains controversial especially when ABC is located at thoracic level. Some have proposed a preoperative embolixation followed by surgical excision with bone grafting, while De Kleuver et Al consider embolization an insufficient treatment [17]. De Cristofaro et Al reported a recurrence rate of 10.5% after superselective embolization [31].

Recently many percutaneous mini-invasive procedures with different sclerosing agents have been developed with good results for the treatment of ABCs and obviating the potentional functional disabilities produced by surgery or radiotherapy. However, same have reported major local and general complications, such as pulmonary and vertebrobasilar embolization, leakage of fibrosing agent in soft tissue, aseptic fistulization and bone destruction [16, 32] caused by sclerosing agents. Many sclerosing agents can be used by mini-invasive direct percutaneous injection: polidocanol, ethibloc or glubaran can be combined with supersective embolization usiing Onyx. The rationale of these elements is to produce an obstruction at the venous side of the multiple parietal arteriolar afferents of the ABC acting by direct damage to the endothelial lining, triggering a coagulation cascade and thrombotic occlusion of blood vessels [27].

Polidocanol 3% (hydroxypolyaethoxydodecan) is used in the treatment of varicose veins and

venous malformations of the head-neck. Rastogi et Al reported a good outcome in patients after treatment with a mean clinical response of 84.5% with a low recurrence rate (2%) including hypopigmentation, necrosis at site of the injection in the case of extravasation, pulmonary embolism, osteomyelitis, and allergic reaction [27].

Ethibloc is a fibrosing and sclerosing agent containing a hydroalcoholic radiopaque solution of zein, a contrast agent, oleum papaverim and propylene glycol. Falappa et Al and Adamsbaum et Al reported a good outcome in A long-term follow-up of patients treated by direct percutaneous injection of ethibloc demonstrating it is a safe and efficient mini-invasive method [33-34]. However, aseptic bone necrosis, venous leakage in soft tissue painful inflammatory reactions have been described, usually when the agent has leaked into the surrounding soft tissue [33-34].

Our experience is based on combined endovascular and direct percutaneous treatment as therapy without any major or minor complications: three by direct percutaneous sclerotherapy, and three by percutaneous sclerotherapy combined with endovascular treatment. Our therapeutic choice was based on the morphology and vascular distribution of the lesion on MR and MDCT imaging and on angiographic study.

Injection of Glubran® is safe and was performed very slowly controlling the flow direction under CT or fluoroscopic guidance without any complication caused by the rapid polymerization proprieties of Glubran® on contact with blood. No inflammatory reaction was observed after treatment by Glubran® injection alone or combined methods. However long-standing experience is recommended for Glubran® injection Onyx® (Onyx Liquid Embolic System) was also injected under fluoroscopic guidance when it was combined with direct percutaneous treatment. It is a biocompatible liquid polymer that precipitates and solidifies in contact with blood, thus forming a soft spongy embolus. The injected material is thick enough to fill vessels but does not adhere to the catheter. Three concentrations are available to permit a range of precipitation rates. A drawback of Onyx is the angiotoxicity of dimethyl-sulphoxide.

All patients in our small series had a complete healing of the cyst. No major complications were noted either during direct percutaneous Glubran injection, or when percutaneous treatment was combined with endovascular embolization. One patient with ABC at C7 level had a recurrence of ABC at three month follow-up and at four months had a car accident and underwent surgical spine stabilization.

Strong patient selection combined with a good understanding of vascular anatomy are recommended for the right choice of treatment.

## Conclusions

In summary, combined endovascular and percutaneous treatment or direct percutaneous sclerotherapy with glue alone offer important and minimally invasive therapeutic options for symptomatic aneurysmal bone cyst with several advantages over other forms of therapy if accurate and correct patient selection are ensured.

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