Complete Intra-Lesional Resection for Aneurysmal Bone Cyst of the Spine. A Personal Experience of 15 Consecutive Cases.

Hossam Salah Eldin Taha MD, FRCS
Orthopaedic Department, Cairo University, Egypt.

Abstract

Background Data: Several methods of treatment have been reported for the management of aneurysmal bone cyst (ABC) of the spine. Incomplete excision has been associated with a high local recurrence rate.

Purpose: To review a personal case series of fifteen cases of ABC of the spine treated by complete intralesional resection and to present the clinical and radiological results.

Study Design: Retrospective review of the clinical and radiological data.

Patients and Methods: Fifteen patients, 8 males and seven females with mean age of 12.7 years underwent complete intralesional resection of ABC lesions of the spine with reconstruction. All patients presented with local axial spine pain, 60% with radicular pain, 47% with a neurological deficit, 13% with painful scoliosis and 13% had a palpable mass. Thirty three percent had previous attempts at surgical resection.

Results: Mean operative time was 6.4 hours and mean estimated blood loss was 1850 ml. Mean follow up was 52.9 months. Eighty six percent of those with a preoperative neurological deficit showed a complete recovery. All patients obtained a solid fusion or were radiologically healed at final follow up (52.9 months).

Conclusion: Complete intralesional excision of ABC lesions of the spine is an effective method of treatment. Attention to reconstruction of the spine is mandatory and should be individually tailored to each case. This surgical strategy eliminated local recurrence in this case series. (2012ESJ027)

Keywords: aneurysmal bone cyst, spine, intralesional resection

Introduction

Aneurysmal bone cyst (ABC) was initially described as a distinct clinicopathological entity by Jaffe and Lichtenstein in 1942.[9] Although these lesions are generally regarded as non-neoplastic in nature, they are expansile tumours containing thin-walled, blood-filled cystic cavities, and they frequently affect the paediatric population. Aneurysmal bone cysts comprise approximately 1% of all primary bone tumours and 15% of all primary spine tumours.[1,11,18,21] Approximately 10 to 30% of ABC cases involve the spine, most commonly in the thoracic and lumbar regions. In these cases, the lesions generally arise in the posterior

elements of the spine and can expand and extend into the pedicles, vertebral body, and spinal canal, resulting in pathological fracture and neurological compromise.[1,3,4,8]

Management of ABCs of the spine requires special consideration due to the relative inaccessibility of the lesions, the proximity of the lesion to the neural structures, and the potential for postoperative spinal instability and deformity.[1,7] A number of methods have been described for the treatment including surgery, radiotherapy, embolization and a combination of these. Simple curettage has been associated with a variable rate of local recurrence. Mankin et al[12] reviewed 150 cases of ABC treated by curettage. They reported 20 % rate of local recurrence. Papageloupolos et al[16] reported 10% local recurrence rate. Ozaki et al[15] reported that nine patients who underwent complete excision did not suffer a local recurrence, whereas two who underwent curettage alone experienced local recurrences. Hay et al[8] reported no recurrences after total excision and a 25% recurrence rate for partial excision. Total excision must include the entire cyst wall, all abnormal tissues that feel spongy, and bone surfaces that are lined with fragile and hypervascular membranes.

This study is a retrospective review of the author's case series of spinal ABC treated by complete intralesional excision and reconstruction. The clinical and radiological results are presented.

Patients and Methods

Fifteen patients with aneurysmal bone cyst of the spine treated between January 2001 and June 2007 were reviewed. There were 8 males and 7 females. The mean age at the time of surgery was 12.7 years (range 4-19 years). All patients presented with local axial spine pain. Nine patients (60%) complained of radicular pain, seven (47%) presented with a neurological deficit, two (13%) with painful scoliosis and two (13%) had a palpable mass. Five patients (33%) had previous attempts at surgical resection (up to four surgeries) and presented with a local recurrence.

All patients had plain radiographs, a CT scan and an MRI. Lesions were located in the lumbar spine in six patients (40%), thoracic spine in four (26.7%), in the cervical spine in four (26.7%) and in the sacrum in one (6.6%). Twelve patients (80%) showed fluid-fluid levels on the MRI. The local extent of the lesion was assessed using the Weinstein-Boriani-Biagini[2] (WBB) staging (Figure 1).

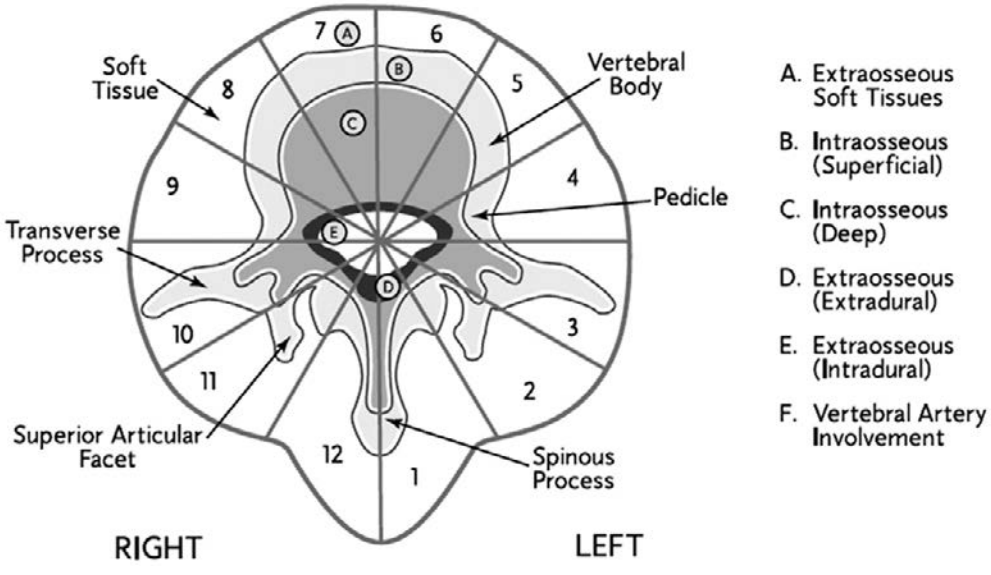

Figure 1. The WBB[2] staging better defines the local extent of the lesion. The cross section of the spine is divided into 12 zones numbered 1 to 12. The extent is then described from superficial to deep designated letters A to F.

Lesions were also staged according to Enneking[5], thirteen were grade 3 (benign aggressive) and two grade 2 (benign active). The patients' data are depicted in table (1).

Table 1. Database of our 15 Patients

Patient	Age/ Sex	Deformity Preop	DOS	ASIA Preop	Level	WBB stage	Enneking	Prev-Surg	Surg App	Follow up/ms	Out-come	ASIA Postop	Deformity Postop
1/YH	5/M	15°kyphosis	5/2001	D	C6/-C7	4-2 ABCDF	3	0	A+P	60	Fused	E	None
2/SA	17/F	10°kyphosis	5/2001	D	C4-C7	1-12 ABCDF	3	4	A+P	86	Fused	E	None
3/RG	11/M	20°kyphosis	7/2001	A	T5-T6	1-12 ABCD	3	1	P+A	66	Fused	A	12° kyphosis
4/MF	15/M	None	9/2001	D	L4	6-11 ABCD	3	0	P	76	Fused	E	None
5/SF	15/F	None	8/2002	E	L3 - L4	11-4 ABCD	3	0	P	66	Fused	E	None
6/AD	4/M	27°kyphosis	9/2002	C	C5-C7	1-12 ABCDF	3	2	P+A	60	Fused	E	0° kyphosis
7/KY	14/F	None	3/2004	D	T1	2-4 ABCD	2	0	P	60	Healed	E	None
8/NK	12/F	22°kyphosis	4/2004	C	T7-T9	9-2 ABCD	3	1	P	48	Fused	E	None
9/SK	12/F	28°scoliosis	5/2004	E	T8	12-3 ABC	3	0	P	36	Fused	E	15° scoliosis
10/SN	16/F	None	8/2004	E	L3	6-12 ABCD	3	0	P	60	Fused	E	None
11/AN	15/M	None	7/2005	E	C6	3-7 ABCDF	3	0	A+P	42	Fused	E	None
12/MH	10/F	None	8/2005	E	L2	8-1 ABCD	2	1	P	28	Fused	E	None
13/MG	17/M	None	2/2006	E	L3	9-2 ABCD	3	0	P	30	Fused	E	None
14/ MM	8/M	25°scoliosis	4/2006	E	L2	10-11 ABC	3	0	P	38	Fused	E	10° scoliosis
15/HS	19/M	None	6/2007	E	S1-S3	11-8 ABCD	3	0	P	38	Healed	E	None

Preop: preoperative, Postop: postoperative, ASIA: neurological status according ASIA classification, Enneking: Enneking staging, Surg App: surgical approach, DOS: date of surgery

In the five patients who had previous surgery, the pathology slides were reviewed by an experienced pathologist and in all five the diagnosis of primary ABC was confirmed. Nine patients had characteristic radiological features of ABC and in these an intra-operative frozen section confirmed the diagnosis. In one patient, the characteristic radiological features were lacking, a core needle biopsy was performed under CT guidance. Pathological features of ABC were found.

Ten patients underwent a posterior surgical approach for resection of the lesion and five had a combined anterior and posterior approach. Twelve patients had instrumented reconstruction and fusion after the resection because of the presence or the risk of spinal instability after the completion of the resection. Two patients had non-instrumented fusion; one (Case 7) had resection of one facet and pedicle at T1 with preservation of the contra lateral facet and underwent an uninstrumented fusion. The other (Case 6) was a four year old patient that had a previous attempt of resection and developed a local recurrence at C5-7. The anterior column was reconstructed with a strut of fibula without anterior plating, followed by posterior resection. Postoperatively the patient was placed in a Minerva plaster cast for three months. He showed an excellent recovery and when last seen 5 years post surgery, was completely asymptomatic with a solid bony fusion. The sacral lesion (Case 15) underwent resection without fusion.

The technique of resection involved a wide exposure attempting to surround the tumour initially without getting into the lesion. Instrumentation anchors were then inserted as needed. At this stage the resection of the lesion began, the walls

were excised as well as all the available tissue and septa within the lesion. The dural sac was identified and a fibrous layer commonly seen upon the dura is completely excised. The lesion surrounded the vertebral artery in four occasions, and in all these occasions the vertebral artery was formally exposed within the foramina transversaria anteriorly, posteriorly or both. This ensured a complete excision of the lesion. In three occasions the tumour extended laterally surrounding the nerve roots lateral to the spinal canal. Again the dissection exposed the involved nerve roots in these locations protecting them and ensuring a complete resection of the lesion (Figure 2). The operative time ranged from 2 to 11 hours (mean 6.4 hours). The estimated blood loss was 300 to 3800 ml (mean 1850 ml).

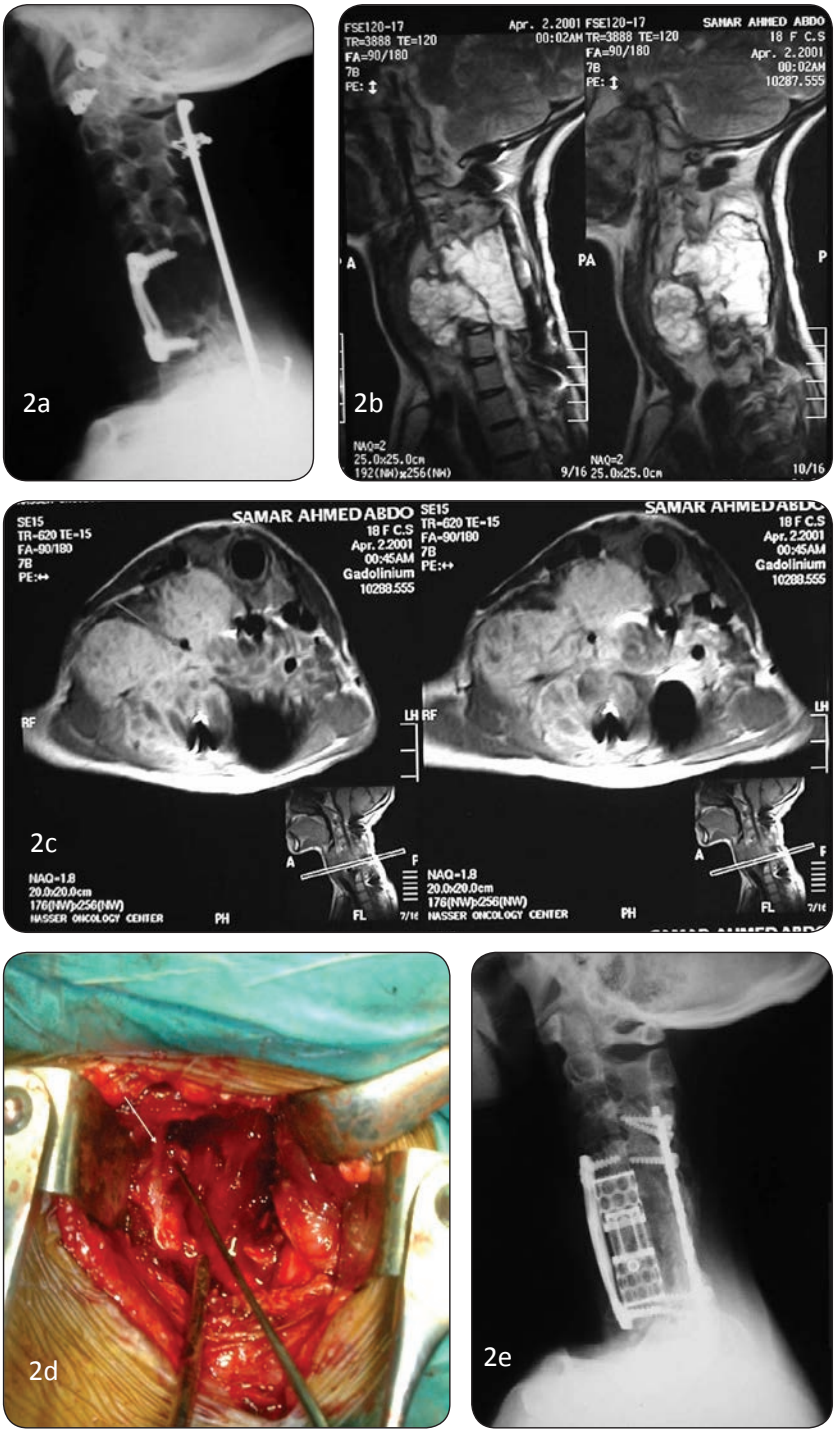

Figure 2.
Case 2: SA 17 year old female underwent 4 previous surgeries and presenting with ASIA D neurology, axial neck pain and right brachialgia.
(2a) plain x-ray lateral view showing destruction of C5,6 and 7 with inadequate anterior and posterior fixation.
(2b) Sagittal T2 MRI showing the extent of the lesion.
(2c) Axial contrast enhanced T1 MRI showing the extent of the lesion. Note the encasement of the vertebral artery by the tumour (red arrow)
(2d) intraoperative photograph showing the anterior exposure of the vertebral artery (white arrow) after excision of the surrounding tumour tissue.
(2e) 5 year postoperative plain x-ray lateral view showing solid bony fusion after anterior and posterior resection and reconstruction. Patient completely free of symptoms with no local recurrence.

Results

Patients were followed up for 28 to 86 months (mean 52.9 months). All patients reported improvement in their axial spine pain at their final follow up; ten reported complete resolution and five reported intermittent pain requiring occasional analgesia. All seven patients presenting with radicular pain had complete relief of their pain. Seven patients presented with a neurological deficit; six showed complete recovery of their deficit, while one patient remained unchanged at ASIA A with complete paraplegia. This was a patient who presented with a recurrent T5/T6 lesion with complete paraplegia (Case 3). No patient was downgraded neurologically by the surgery.

Radiological follow up included plain radiographs and occasionally CT scan and MRI. All patients showed bony fusion or complete healing without evidence of local recurrence at the final follow up (Figure 3).

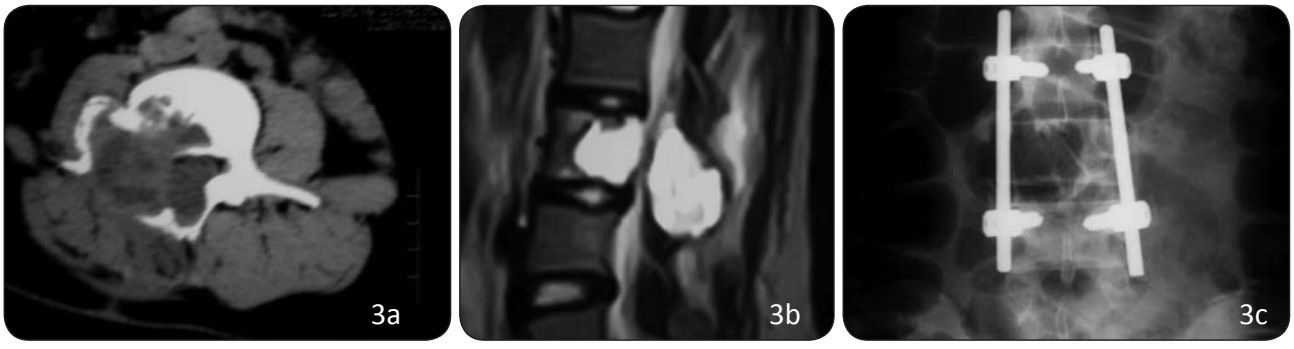

Figure 3. Case 10: SN 16 year old female presenting with back pain and right L3 radiculopathy. **(3a)** Axial CT of L3 showing the extent of the lesion. **(3b)** Sagittal T2 MRI showing anterior and posterior destruction by the lesion. **(3c)** Postoperative plain x-ray AP view showing stabilization and fusion from L2 to L4.

One patient developed a post-operative pneumonia that required an extended hospital stay of ten days and IV antibiotics. One patient developed a low grade infection of the posterior wound. This required a three week period of IV antibiotics. His wound eventually healed without consequences. There were no neurological, implant related complications or pseudoarthrosis found in this series.

Discussion

This case series have shown a wide local extent of the lesion and therefore has required more extensive reconstruction. This may be related to the aggressive behaviour of the lesions in this series and a delay in the diagnosis. Reconstruction has required individualized surgical planning and execution depending upon the site, the extent of the lesion and the age of the patient.

Nine patients (90%) of those with thoracic and lumbar lesions (N=10) had a posterior approach and one (10%) had a combined anterior and posterior approach. De kleuver et al[4] showed that the anterior approach had a significantly lower local recurrence than the posterior approach. We believe that this is not really related to the approach but rather to the completeness of the excision. Indeed, 60% of their patients who had incomplete excision suffered a local recurrence. Thus, in their series, the posterior approach represented an incomplete excision and therefore higher local recurrence. In this series, the posterior approach was an extensible one; following the lesion through the destructed pedicle into the vertebral body with thorough excision of the lesion and using a high speed burr. A costo-transversectomy was added to improve the exposure in two thoracic lesions (Cases 8, 9). An anterior approach was used, in addition, in one case; a case with a recurrent lesion, paraplegia and extensive destruction (Case 3). All of our cervical cases (N=4) had a combined anterior and posterior approach. Others have reported a similar technique.[10,17,19] In addition, an exposure of the vertebral artery either anteriorly, posteriorly or both was performed, depending upon the local extent of the lesion, that allowed a complete excision of the tumour.

Garg et al[7] described a surgical technique using a four-step approach of intralesional curettage, high-

speed bur, electrocautery, and bone grafting for treating spinal ABC. They reported elimination of recurrence (0/8 cases) using the four-step approach compared with traditional intralesional curettage and bone grafting (4/4 cases). The technique described in this series is slightly different; no electrocautery was used in this series, however, an aggressive curettage using a high speed burr and completely excising the walls back to normal looking healthy tissues has been used. Others have reported elimination of local recurrence with total excision.

Boriani et al[1] have suggested en-bloc excision of ABC lesions located posteriorly, WBB sectors 9 to 4. In our view, en bloc excision is just not warranted being an overtreatment for such a benign lesion but also can be difficult to perform because of the gross texture of the lesion. The cystic component and the friability of the walls make it difficult to handle it en bloc. Complete intralesional resection with complete excision of the wall back to healthy looking tissues has eliminated local recurrence in this series.

Five patients (33%) had one to four previous surgeries, where incomplete intralesional resections were performed. In a sense, this gives the rate of local recurrence after incomplete resection of these lesions. In addition, it reflects the aggressive behaviour of these lesions. The presence of previous surgery has added to the complexity of our surgery, however the goals remained the same; a complete intralesional resection with complete excision of the wall back to normal looking healthy tissue. In all five patients, no local recurrence of the lesion was observed at the latest follow up.

Boriani et al[1] believed that selective arterial embolization should be the first line of treatment for spinal ABC. However, patients with pathologic fractures, neurologic involvement and high cervical and low thoracic locations are not suitable for embolization and surgery is indicated. Thirteen patients (87%) of this series fell within these exclusion criteria and were therefore unsuitable for embolization.

Several authors have reported the use of preoperative embolization to decrease intraoperative bleeding.[1,13] No preoperative embolization was used in this series. Although intraoperative bleeding can be significant, this was controlled by the excision. The surgical strategy of surrounding the tumour and inserting implant anchors before attempting the excision had two values: first it helped with the identification of the walls of the lesion and therefore the completeness of the excision and second, the decrease of the blood loss by delaying the entry into the lesion. The swift excision decreased the bleeding; the more excision of the tumour the less the bleeding becomes. Fay et al[6] have reported intralesional injection of fibrin glue during surgery to decrease the bleeding. This seems to be an interesting idea although we have no experience with it.

Local adjuvant, like cryosurgery, has been reported to extend margins of resection in benign aggressive lesions. However, the risk of damage to neural structures is a concern when applying it to the spine.[20] This was the reason for avoiding it in this series.

Recently, an oncological origin for primary ABC has been suggested. An oncogene has been found in these lesions identifying the spindle cell as the neoplastic cell. This oncogene was absent in all other cells within the lesion as the giant and endothelial cells.[14] These spindle cells are found within the walls of the lesion. These findings may support the clinical experience here and with others that complete excision including the walls significantly decreases or eliminates local recurrence.

Conclusion

Complete intralesional excision is an effective treatment of ABC lesions of the spine. Completeness means excising all parts of the tumour including the walls back to normally looking healthy tissues. Reconstruction is individually tailored to each case and depends upon the location, extent and age of the patient. This surgical strategy obtained excellent results and eliminated local recurrence in this case series.

References

1. Boriani S, De lure F, Campanacci L, Gasbarrini A, Banderia S, Biagini R, et al: Aneurysmal bone cyst of the mobile spine. Spine 26:27–35, 2001
2. Chan P, Boriani S, Fourney DR, Biagini R, Dekutoski MB, Fehlings MG: An assessment of the reliability of the Enneking and Weinstein-Boriani-Biagini classifications for staging of primary spinal tumors by the Spine Oncology Study Group. Spine 34(4):384-91, 2009

3. Cottalorda J, Kohler R, Gauzy J, Chotel F, Mazda K, Lefort G, et al: Epidemiology of aneurysmal bone cyst in children: a multicenter study and literature review. J Pediatric Orthopaedics B 13:389–394, 2004
4. De Kleuver M, Van der Heul RO, Veraart BEEMJ: Aneurysmal bone cyst of the spine: 31 cases and the importance of the surgical approach. J Pediatr Orthop B7:286–292, 1998
5. Enneking WF: Aneurysmal bone cyst. In Musculoskeletal tumor surgery. New York, NY: Churchill Livingstone, 1983:1513–40
6. Fay LY, Wu JC, Huang WC, Shih YH, Cheng H: One-stage posterior resection is feasible for a holovertebral aneurysmal bone cyst of the axis: a case report and literature review. Surg Neurol 72(Suppl 2):S80-5, 2009
7. Garg S, Mehta S, Dormans JP: Modern surgical treatment of primary aneurysmal bone cyst of the spine in children and adolescents. J Pediatr Orthop 25(3):387-92, 2005
8. Hay MC, Paterson D, Taylor TKF: Aneurysmal bone cysts of the spine. J Bone Joint Surg Br 60B:406–411, 1978
9. Jaffe HL, Lichtenstein L: Solitary unicameral bone cyst with emphasis on roentgen picture, the pathological appearance and pathogenesis. Arch Surg 44:1025–1044, 1942
10. Khalil IM, Alaraj AM, Otrock ZK, Chamoun RB, Sabbagh AS, Skaf GS: Aneurysmal bone cyst of the cervical spine in a child: case report and review of the surgical role. Surg Neurol 65(3):298-303, 2006
11. Leithner A, Windhager R, Lang S, Haas O, Kainberger F, Kotz R: Aneurysmal bone cyst. A population based epidemiologic study and literature review. Clin Orthop 363:176–179, 1999
12. Mankin HJ, Hornicek FJ, Ortiz-Cruz E, Villafuerte J, Gebhardt MC: Aneurysmal bone cyst: a review of 150 patients. J Clin Oncol 20(23):6756-6762, 2005
13. Novais EN, Rose PS, Yaszemski MJ, Sim FH: Aneurysmal bone cyst of the cervical spine in children. J Bone Joint Surg Am 93(16):1534-43, 2011
14. Oliveira AM, Perez-Atayde AR, Inwards CY, Medeiros F, Derr V, His BL, Gebhardt MC, et al: USP6 and CDH11 oncogenes identify the neoplastic cell in primary aneurysmal bone cysts and are absent in so-called secondary aneurysmal bone cysts. Am J Pathology 165(5):1773-8, 2004
15. Ozaki T, Halm H, Hillmann A, Blasius S, Winkelmann W: Aneurysmal bone cysts of the spine. Arch Orthop Trauma Surg 119(3-4):159-62, 1999
16. Papagelopoulos PJ, Currier BL, Shaughnessy WJ, Sim FH, Ebsersold MJ, Bond JR, et al: Aneurysmal bone cyst of the spine. Management and outcome. Spine 23(5):621-8, 1998
17. Perlmutter DH, Campbell S, Rubery PT, Vates EG, Silberstein HJ: Aneurysmal bone cyst: surgical management in the pediatric cervical spine. Spine 34(1):E50-3, 2009
18. Ramirez AR, Stanton RP: Aneurysmal bone cyst in 29 children. J Pediatr Orthop B 22:533–539, 2002
19. Refai D, Holekamp T, Stewart TJ, Leonard J: Circumferential vertebrectomy with reconstruction for holocervical aneurysmal bone cyst at C4 in a 15-year-old girl. Spine 32(24):E725-9, 2007
20. Schreuder HWB, Veth RPH, Pruszczynski M, Lemmens JA, Koops HS, Molenaar WM: Aneurysmal bone cyst treated by curettage, cryotherapy, and bone grafting. J Bone Joint Surg 79B:20–5, 1997
21. Turker RJ, Mardjetko S, Lubicky J: Aneurysmal bone cysts of the spine: Excision and stabilization. J Pediatr Orthop 18:209–213, 1998

Address reprint request to:

Hossam Salah EldinTaha, MD
10 Hoda Shaarawy street, Bab Elluk, Cairo Egypt.
E-mail: hossammeister@gmail.com

الملخص العربي

الاستئصال الكامل من داخل الآفة لأكياس العظام الدموية بالعمود الفقري

البيانات الخلفية: توجد عدة طرق لعلاج أكياس العظام الدموية (ABC) في العمود الفقري . ارتبط الاستئصال غير كامل مع معدل عالي من الارتجاع المحلي.

الغرض: لاستعراض سلسلة من خمسة عشر حالة من أكياس العظام الدموية (ABC) في العمود الفقري تم علاجها بواسطة استئصال كامل داخل الآفة وتقديم النتائج السريرية والإشعاعية.

تصميم الدراسة: استعراض بأثر رجعي من البيانات السريرية والإشعاعية.

الأساليب: خضع ١٥ مريضا، ٨ ذكور و ٧ إناث مع متوسط عمر قدره ١٢.٧ سنة لاستئصال كامل من داخل الآفة لأكياس العظام الدموية بالعمود الفقري مع إعادة البناء. كان كل المرضى يعانون من آلام العمود الفقري المحوري ألم، و ٦٠٪ يعانون من ألم جذري، ٤٧٪ مع وجود عجز عصبي، و ١٣٪ مع الجنف المؤلم و ١٣٪ يعاني من كتلة واضحة. تعرض ٣٣٪ من المرضى لاستئصال جراحي سابق.

النتائج: كان متوسط وقت الجراحة ٦.٤ ساعة ومتوسط فقدان الدم يقدر بـ ١٨٥٠ مل. متوسط المتابعة كان ٥٢،٩ شهرا. وأظهر ٨٦٪ من الذين يعانون من عجز في جهازه العصبي قبل الجراحة تحسنا كاملا. حصل جميع المرضى على التحام صلب أو التئام كامل في المتابعة النهائية.

الخلاصة: الاستئصال الكامل من داخل الآفة لأكياس العظام الدموية بالعمود الفقري هو وسيلة فعالة للعلاج. يجب الانتباه إلى إعادة بناء العمود الفقري ويجب أن يكون مصمما بشكل فردي لكل حالة. اتباع هذه الاستراتيجية الجراحية منع الارتجاع المحلي في هذه السلسلة من الحالات.