

Solid aneurysmal bone cyst on the cervical spine of a young child

L. Casabianca · A. Journé · G. Mirouse ·
M. Zerah · D. Moulies · C. Glorion ·
T. Odent

Received: 29 September 2014/Revised: 9 February 2015/Accepted: 11 February 2015/Published online: 18 March 2015
© Springer-Verlag Berlin Heidelberg 2015

Abstract



Introduction Solid aneurysmal bone cyst (S-ABC) is a variant of aneurysmal bone cyst (ABC), an uncommon benign bone tumor. There are few cases described in the cervical spine in kids up today. We treated a recurrent case with neurological involvement that needed multiple surgical procedures and radiotherapy.

Case presentation We report a case of C4 located S-ABC concerning a 2-year-old boy treated surgically by anterior and posterior approach. Three months after the initial procedure appearance of a tetraparesis led to diagnose a local recurrence treated by sclerotherapy and a second surgery. The patient had a full neurological recovery. Three months later, a follow-up CT scan showed a second recurrence requiring a new surgical revision by anterior approach and radiotherapy.

Outcome At 6-year follow-up after four surgical procedures, sclerotherapy and radiotherapy, the aneurysmal bone cyst has been healed. Patient had neurological impairment after a local recurrence but had full recovered after final revision surgery.

Keywords Aneurysmal bone cyst · Cervical cyst · Tetraparesis · Recurrence · Surgery

Case presentation

A 2-year-old boy presented in consultation with a neck pain and torticollis. The neurological examination was normal. No specific history of injury or trauma was reported. This patient had no personal or familial antecedent history of medical or surgical diseases. Laboratory findings were normal except for a small isolated hypothyroidism.

Diagnostic imaging section

We made the diagnosis of a lesion of 50 mm, an osteolytic lesion of the vertebral body of C4 (Fig. 1). There was no spinal compression. A surgical biopsy performed by a posterior approach concludes to an aneurysmal bone cyst.

L. Casabianca (✉) · A. Journé · G. Mirouse · M. Zerah ·
C. Glorion · T. Odent
Service d'orthopédie infantile, Hôpital Universitaire Necker
Enfants Malades, Assistance Publique Hôpitaux de Paris,
149 Rue de Sèvres, 75015 Paris, France
e-mail: laurent.casabianca@hotmail.fr

L. Casabianca · A. Journé · G. Mirouse · M. Zerah ·
C. Glorion · T. Odent
Faculté de médecine Paris Descartes, Université Sorbonne Paris
Cité, Paris, France

D. Moulies
Service d'orthopédie pédiatrique, CHU Limoges, 2, avenue
Martin Luther King, 87042 Limoges, France

Fig. 1 Initial X-rays with antero-posterior and lateral view of the lesion



Historical review

Aneurysmal bone cyst (ABC) is a pseudotumoral lesion of unknown etiology; it was first described by Jaffe and Lichtenstein in 1942 like a blood-filled lesion with a blow-out radiographic appearance, and it represents 15 % of all primary bone tumors [1–4]. However, there is a distinct variant of ABC, the so-called solid variant (S-ABC), which was first described by Sanerkin et al. in 1983 [5].

Incidence and localization

Primary ABC represents 1.4 % of primary bone tumors from the vertebral column [6, 7]; the cervical spine is involved in 30–41 %. Some of these tumors are solid tumors, whereas others are fluid cysts [4, 5]. S-ABC are rare lesions, accounting for 3.4–7.5 % of all ABC and almost 15 cases occurring in the spine have been reported [8, 9]. According to the literature, the age of the patients ranged from 6 to 17 years, with a sex ratio of 1:1.5 [9].

Diagnosis and differential diagnosis

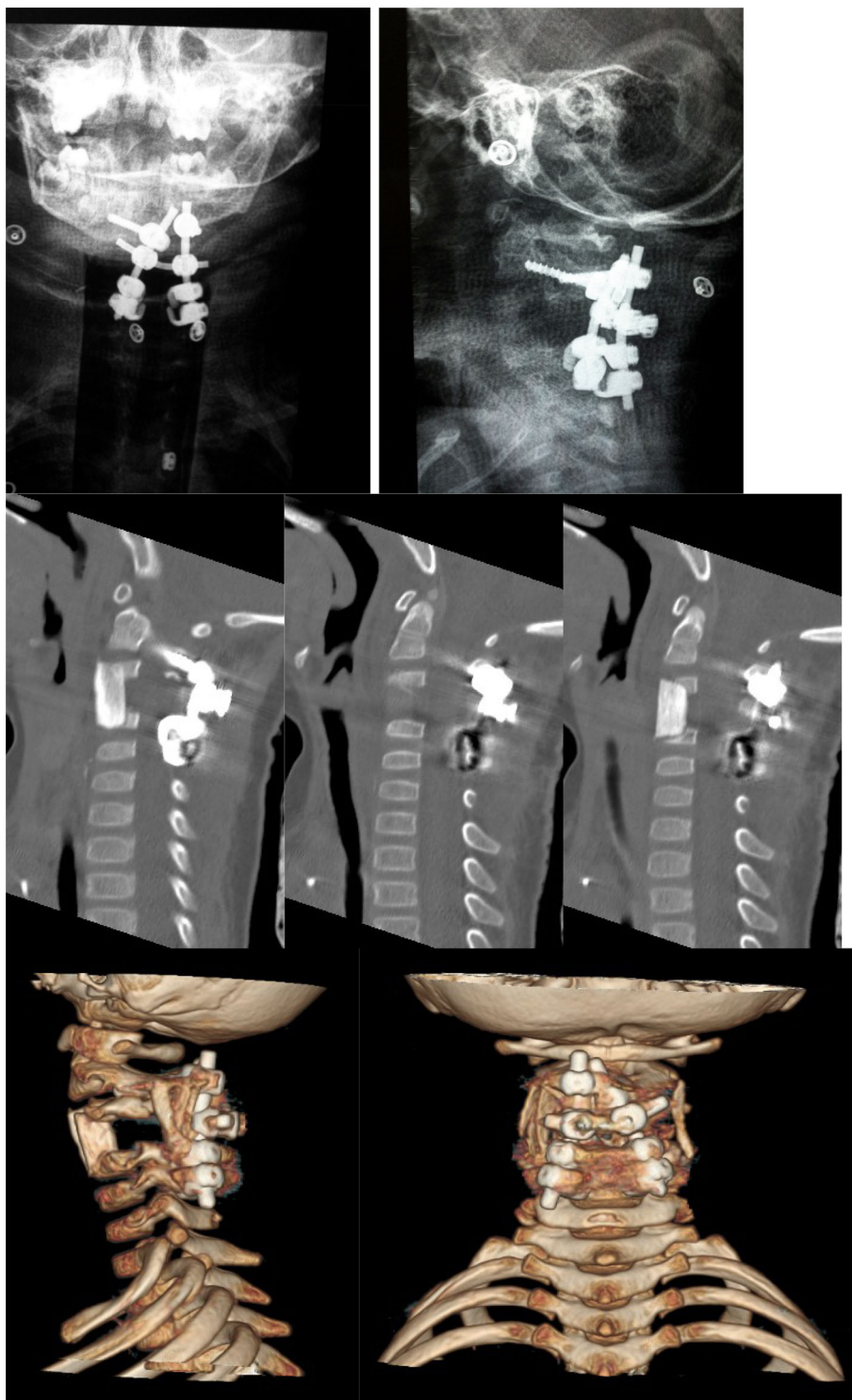
The diagnosis of S-ABC was partly based on the X-ray and CT aspects of the lesion: an osteolytic and expansive lesion [8, 10] that is indistinguishable from conventional ABC. On MRI, we found homogeneous low intensity on T1-weighted images [10] and heterogeneous low signal intensity with scattered high signal intensity areas on T2-weighted images [10, 11], the mass showed heterogeneous

high signal intensity on Gd-enhanced images [8]. The solid variant of ABC can be easily mistaken for a spindle cell neoplasm, especially telangiectatic osteosarcoma [9] or differential diagnosis of any lytic expansile destructive lesion of the spine: essential cyst, hemangioma, chondroblastoma, osteoblastoma and giant cell tumor. Histologically, ABC is typically characterized by cavernous channels surrounded by a spindle cell stroma with osteoclast-like giant cells and osteoid or bone production. In the solid variant described by Sanerkin et al. [5], we usually find a florid fibroblastic or fibrohistiocytic proliferation without any cellular or nuclear pleomorphism, areas with osteoclast-like giant cells, osteoblastic differentiation with osteoid production [5], fibromyxoid tissues component [8]. We performed a biopsy that found fibroblasts and giant osteoclastic cells associated with immature osteoid substance. We also realized a karyotype, which showed no abnormal clone.

Rationale for treatment and evidence-based literature

There are many existing treatment concerning S-ABC in literature [11–14] and treatment of ABC still controversial. For long bone ABC, it was simpler: curettage or en bloc excision and bone grafting is the mainstay of treatment because the lesion is easily accessible and surgical excision feasible without sequelae. On spine localizations treatment options are more complex, especially in the cervical spine due to the proximity of noble structures. When en bloc excision is technically difficult, we may consider other options of treatment as complete curettage, adjuvant selective arterial embolization, radiotherapy, intralesional

Fig. 2 X-rays and CT scan of the first intervention: excision, C5–C2 osteosynthesis and anterior strut bone grafting



injection of sclerosing agent, cryotherapy or embolization [12–14].

En bloc excision

Although en bloc removal of the tumor has the lowest rate of local recurrence, it can be technically challenging or

impossible particularly in the cervical spine due to the infiltration of the vertebral artery and the nerve roots [13, 14]. Surgery has many risks such as paralysis, intraoperative bleeding, neurological complications, and scarring complications. In addition to the operative risks, children have the highest incidence of post laminectomy kyphosis [15, 16]. Yasouka et al. [17] postulated that the deformity

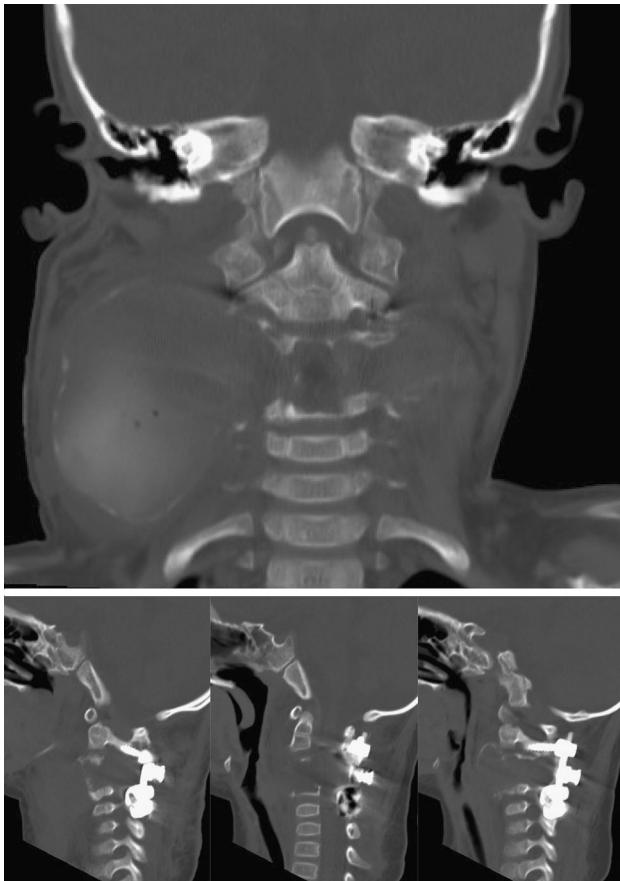


Fig. 3 CT scan showing ABC recurrence with complete bone graft osteolysis, extension of the lesion from C3 to C5 and to the surrounding soft tissue, and C2 screw loosening

is due to a wedging change in the cartilaginous portion of the vertebral body and to the viscoelasticity of ligaments in children. When treatment of this complication becomes necessary, anterior fusion may be effective in arresting progression.

Curettage

The curettage is often done during biopsy and could be sufficient to heal the cyst but may also result in recurrence, which will be problematic in an area where a second surgery would be difficult. In a series of ABC treated by curettage, 22 % required a second operation and 8.6 % required a third operation for recurrences or structural problems related to failed systems [1]. Intralesional excision is followed by a 30 % incidence of recurrence and a higher recurrence [6]; therefore, a complete marginal resection should be the goal.

Curopsy is a new biopsy technique introduced by Reddy Ki [18]. Curopsy technique is a percutaneous limited curettage biopsy, targeting the lining membrane from various quadrants of the cyst, leading then to consolidation.

This technique wasn't appropriate for the treatment of solid form of ABC.

Embolization

Selective arterial embolization has been used successfully as a sole treatment modality, and is also often recommended before surgery to reduce potential bleeding during surgery [19]. Embolization of the cyst may be a good solution in the management of ABC without destabilization of the spine [20] when you have a histological diagnosis. But embolization could cause cord ischemia in patients with cervicothoracic involvement. For this reason, its application to tumors lower than the T6 level is expected to be more successful. Moreover, this technique may be insufficient in cases where the lesion is very large [20, 21].

Radiotherapy

Postoperative radiotherapy treatment is useful for preventing local recurrences, especially in intralesionary cases. Radiation therapy is contraindicated in young patients near the spine because of the risk of myelopathy [22] radiation-induced spinal deformity [23] and secondary sarcomas. Radiation therapy should be reserved for patients with inoperable lesions or a recurrence that compromise the spinal cord.

Procedure

In this case, we opted initially for a complete surgical excision of the lesion by dual approach. This surgical option was chosen because of the size of the lesion (50 mm) and its dangerous location. It was threatening the stability of the cervical spine due to the large bone destruction (not easily accessible to single embolization). Due to the proximity to the spinal cord and the vertebral artery, cryotherapy or massive embolization were too risky.

The patient underwent the surgery at the age of 2 years and 3 months and the intervention has been performed by anterior and posterior approach. We realized a posterior mid-line skin incision focused on C2–C6 with tumorectomy, resection of the C4 lamina, spinous process and the right side part of C5. We released the vertebral arteries and the spinal cord. The spine was instrumented by two C2 pedicle screws and two hooks sus and sub laminar on C5 and C6 restoring stability of the spine with bone graft taken from the posterior ridge. Secondary, we did a right cervicotomy with tumorectomy and bone grafting. We noticed that the ABCs destroyed the totality of the vertebral body of C4. We achieved C4 corporectomy and C3–C4 and C4–C5 discectomy. Then, we realized a cortical bone strut graft

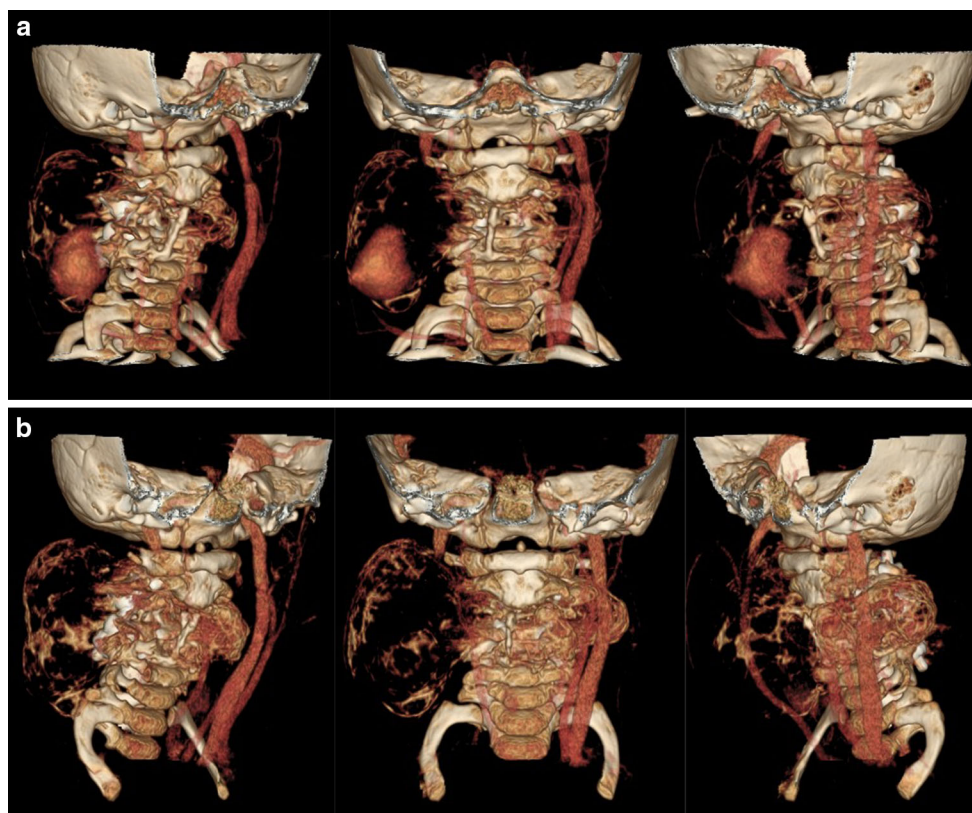


Fig. 4 a, b CT angiography. a Connection between the lesion and the vertebral artery. On the *right side*, the blocked vertebral artery and the perfusion of the lesion, b decrease of the lesion perfusion after selective embolisation

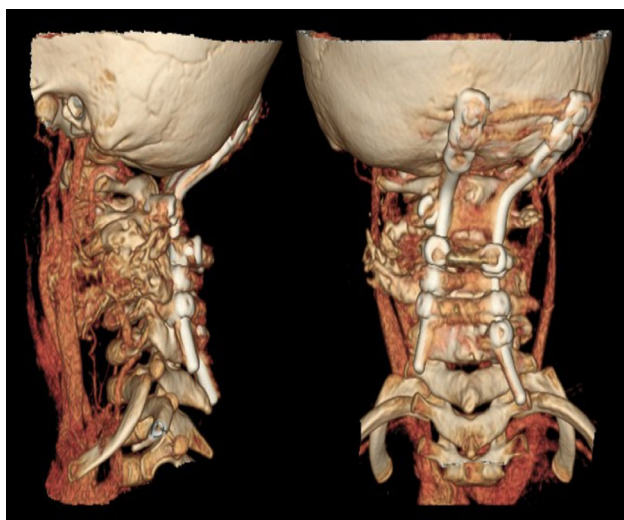


Fig. 5 CT-scan 3D reconstruction: extension of the posterior osteosynthesis to the occiput after the second surgery

from the right tibia in-between C3 and C5 vertebral body (Fig. 2). There was no major issue in the postoperative period after a quick stay of 4 days in intensive care unit. A brace maintained the cervical spine during 3 months. The final histology report concluded in S-ABC.

Follow-up

Three months after the procedure appeared a tetraparesis syndrome with a predominant deficit on the right arm. The CT found a huge tumor regrowth predominant on the right side with a complete osteolysis from C3 to C5 and of the autologous graft (Fig. 3). We first decided to perform a sclerotherapy which contributed in reducing the deficit signs on the lower limb and the volume of the lesion (Fig. 4). We have proceeded a second surgery to decompress the spinal cord by a posterior approach. We found a full destruction of the right pedicle of C2 associated with a loosening of the right screw. We resected the right component of the recurrence, release the spinal cord and roots and modified the osteosynthesis by extending the fusion to the occiput with hooks and bone graft (Fig. 5). Evolution was initially positive with disappearance of tetraparesis but a persistent deficit in the right arm. A second look surgery was performed 15 days after to remove the left posterolateral component of the lesion. Blood loss was two blood mass during these procedures. A fourth surgery has been done by anterior approach finally to remove the anterior component of the lesion with an osteosynthesis consisting of a screw inserted between vertebral bodies of C2 and C5 to stabilize the anterior spine.



Fig. 6 X-ray and CT-scan at last follow-up (6 years). Bone consolidation achieved without local recurrence

After 1 month, we realized radiotherapy (40 Gy) to reduce the risk of local recurrence.

Outcome

At 6-year follow-up, we have not observed local recurrence; the osteosynthesis achieved a stable spine. The child has been reviewed in consultation; he showed no after-effects of surgery, normal levels of mobility to four members, no paresthesias, no neurological symptoms or cervical complains.

Growth chart was normal and he didn't have any kyphotic deformation. Bone consolidation was achieved on the CT scan without any local recurrence (Fig. 6).

This case illustrates a solid ABC of the cervical spine, which is a benign bone tumor whose risk of recurrence is variable and depends on many factors [3]. This recurrence was observed in a very aggressive fashion and multiples surgeries were necessary to control this recurrence, resulting in a significant bone loss and an extensive osteosynthesis of the cervical spine. Fortunately, the patient had a full neurological recovery.

In this pathology particularly in children and the cervical spine, it is important to consider all treatment options before suggesting surgery which must remain a last intention option and remains at high risk of complications particularly recurrences.

Conflict of interest None of the authors have any potential conflict of interest.

References

- Mankin HJ, Hornicek FJ, Ortiz-Cruz E, Villafuerte J, Gebhardt MC (2005) Aneurysmal bone cyst: a review of 150 patients. *J Clin Oncol* 23(27):6756–6762
- Cottalorda J, Kohler R, Sales de Gauzy J, Chotel F, Mazda K, Lefort G, Louahem D, Bourelle S, Dimeglio A (2004) Epidemiology of aneurysmal bone cyst in children: a multicenter study and literature review. *J Pediatr Orthop B* 13(6):389–394
- Saccomanni B (2008) Aneurysmal bone cyst of spine: a review of literature. *Arch Orthop Trauma Surg* 128(10):1145–1147 (Epub 2007 Oct 9)
- Jaffe HL, Lichtenstein L (1942) Solitary unicameral bone cyst: with emphasis on the roentgen picture, the pathologic appearance and the pathogenesis. *Arch Surg* 44:1004–1025
- Sanerkin NG, Mott MG, Roylance J (1983) An unusual intraosseous lesion with fibroblastic, osteoclastic, osteoblastic, aneurysmal and fibromyxoid elements. “Solid” variant of aneurysmal bone cyst. *Cancer* 51(12):2278–2286
- Hay MC, Paterson D, Taylor TK (1978) Aneurysmal bone cysts of the spine. *J Bone Joint Surg Br* 60-B(3):406–411
- Papagelopoulos PJ, Currier BL, Shaughnessy WJ, Sim FH, Ebersold MJ, Bond JR, Unni KK (1998) Aneurysmal bone cyst of the spine. Management and outcome. *Spine (Phila Pa 1976)* 23(5):621–628
- Suzuki M, Satoh T, Nishida J, Kato S, Toba T, Honda T, Masuda T (2004) Solid variant of aneurysmal bone cyst of the cervical spine. *Spine (Phila Pa 1976)* 29(17):E376–E381
- Bertoni F, Bacchini P, Capanna R, Ruggieri P, Biagini R, Ferruzzi A, Bettelli G, Picci P, Campanacci M (1993) Solid variant of aneurysmal bone cyst. *Cancer* 71(3):729–734
- Al-Shamy G, Relyea K, Adesina A, Whitehead WE, Curry DJ, Luerssen TG, Jea A (2011) Solid variant of aneurysmal bone cyst of the thoracic spine: a case report. *J Med Case Rep.* 30(5):261. doi:10.1186/1752-1947-5-261
- Karampalis C, Lenthall R, Boszczyk B (2012) Solid variant of aneurysmal bone cyst on the cervical spine of a child: case report, differential diagnosis and treatment rationale. *Eur Spine J* 22(3):523–531. doi:10.1007/s00586-012-2548-9 (Epub 2012 Oct 31)
- Zileli M, Isik HS, Ogut FE, Is M, Cagli S, Calli C (2013) Aneurysmal bone cysts of the spine. *Eur Spine J* 22(3):593–601. doi:10.1007/s00586-012-2510-x (Epub 2012 Oct 1)
- Gladden ML Jr, Gillingham BL, Hennrikus W, Vaughan LM (2000) Aneurysmal bone cyst of the first cervical vertebrae in a child treated with percutaneous intralesional injection of calcitonin and methylprednisolone. A case report. *Spine (Phila Pa 1976)* 25(4):527–530
- Rai AT, Collins JJ (2005) Percutaneous treatment of pediatric aneurysmal bone cyst at C1: a minimally invasive alternative: a case report. *AJNR Am J Neuroradiol* 26(1):30–33
- Bell DF, Walker JL, O'Connor G, Tibshirani R (1994) Spinal deformity after multiple-level cervical laminectomy in children. *Spine (Phila Pa 1976)* 19(4):406–411
- Aronson DD, Kahn RH, Canady A, Bollinger RO, Towbin R (1991) Instability of the cervical spine after decompression in patients who have Arnold-Chiari malformation. *J Bone Joint Surg Am* 73(6):898–906
- Yasuoka S, Peterson HA, Laws ER Jr, MacCarty CS (1981) Pathogenesis and prophylaxis of postlaminectomy deformity of the spine after multiple level laminectomy: difference between children and adults. *Neurosurgery* 9(2):145–152
- Reddy KI, Sinnaeve F, Gaston CL, Grimer RJ, Carter SR (2014) Aneurysmal bone cysts: do simple treatments work? *Clin Orthop Relat Res* 472(6):1901–1910. doi:10.1007/s11999-014-3513-1
- Konya A, Szendroi M (1992) Aneurysmal bone cysts treated by superselective embolization. *Skeletal Radiol* 21:167–172
- Amendola L, Simonetti L, Simoes CE, Bandiera S, De Iure F, Boriani S (2013) Aneurysmal bone cyst of the mobile spine: the therapeutic role of embolization. *Eur Spine J* 22(3):533–541
- Ameli NO, Abbassioun K, Saleh H, Eslamdoost A (1985) Aneurysmal bone cysts of the spine. Report of 17 cases. *J Neurosurg* 63(5):685–690
- Palmer JJ (1972) Radiation myelopathy. *Brain* 95(1):109–122
- Mayfield JK, Riseborough EJ, Jaffe N, Nehme ME (1981) Spinal deformity in children treated for neuroblastoma. *J Bone Joint Surg Am* 63(2):183–193