

Surgical Management of Giant Multilevel Aneurysmal Bone Cyst of Cervical Spine in a 10-Year-Old Child: Case Report with Review of Literature (Evid Based Spine Care J 2012; 3(4):55–59)

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In response to the excellent case report and summary on aneurysmal bone cysts by Gurjar et al¹ in the November 2012 edition of *EBSJ*, we felt that some additional points should be brought to the attention of the readers. In the management of these difficult but benign lesions, many good points were raised by the authors, but minimally invasive sclerotherapy was omitted. This procedure involves percutaneous puncturing, often repeated injections of a sclerosing agent, traditionally polidocanol and more recently ethanol, owing to complications reported with the former and not seen with the latter. It is a curious omission of the authors because one of the better articles considering sclerotherapy—"Is Sclerotherapy Better than Intralesional Excision for Treating Aneurysmal Bone Cysts?" by Varshney in *CORR* 2010—is from one of the author's institution, the All India Institute of Medical Sciences.² In this Level II study, 94 patients were prospectively randomized into two treatment groups receiving either repetitive sclerotherapy using polidocanol or an intralesional extended curettage with autograft. With an average follow-up of more than 3 years, 93% achieved the group's criteria for healing versus 85% in the curettage control group, yet with a much more favorable complication profile. This injection-based treatment option has also been left unnoticed by other centers—as much as can be gleaned from the literature—as this form of therapy is likely not available in some institutions without more advanced interventional radiology departments.³ There is concern, however, regarding cervical aneurysmal bone cysts (ABCs) and injection of Ethibloc (polidocanol) following a case report resulting in death. This was felt to be related to tumor involvement with the vertebral artery.⁴ As noted by Gurjar, preoperative angiography, and if possible, embolization, are requisite studies.

In our experience, sclerotherapy has been a valuable tool in the treatment of this disease. For tumors with significant three-column involvement over multiple areas, it may not be possible to remove the lesion in its entirety even though a macroscopic intralesional resection may seem complete. Residual or "recurrent" disease involving one or both vertebral arteries may be seen on MRI in the setting of a solid incorporating fusion and graft. We have found sclerotherapy to be very helpful in those cases to try to get a jump on early "recurrence." Extension into the bone graft could precipitate implant loosening and pseudarthrosis, which are difficult issues, especially in children. Within the last year, we had five large cervical and two thoracic ABCs with multicolumn and multilevel involvement; four were resected and instrumented, thus far without recurrences. Another was resected without reconstruction and is doing well. Two children have had resection and circumferential-instrumented fusions but have recurrent tumors being managed with sclerotherapy and serial MRI as the adjuvant. As a standalone treatment, sclerotherapy has not been helpful for the large multilevel, multicolumn tumors, but it is a helpful adjuvant, especially in early recurrence or smaller tumors where resection and reconstruction are unnecessary as discussed by Varshney et al.²

Also worth mentioning is the concept of spinal instability in neoplastic conditions, which does not directly equate with traditional methods of assessing stability in trauma. Destruction by tumor does not usually involve the additional loss of the ligamentous and soft tissue secondary stabilizers, making the assessment of stability much more difficult. Fournier and Gokaslan as well as the Spine Oncology Study Group have illustrated these unique differences to neoplasia.^{5,6} With respect to the case described by Gurjar et al and its

three-column involvement with encasement of the vertebral artery, sclerotherapy may not have been the best option and this may be the reason it was not included in the article. One should, however, not forget that percutaneous sclerotherapy merits inclusion as a neoadjuvant treatment modality, especially for children with aneurysmal bone cysts presenting with larger cases typically seen in tertiary centers.

Absent from the review portion in the presented article is the neoplastic status of the tumor which has been nicely documented by recurrent t(16;17)(q22;p13) translocations and USP6 rearrangements in many articles such as Panoutsakopoulos et al.⁷ and Pietschmann et al.⁸ Evidence is growing that supports the concept that aneurysmal bone cysts are clonal neoplasms rather than the previously held theory that they represent vascular derangements. Many would challenge this group's declarative nonneoplastic designation, but this is an area of some controversy. Certainly, they exhibit all the characteristics of Enneking class 2 or 3, benign aggressive neoplasm.

Radiation is not a reasonable option in the child unless the tumor cannot be controlled after resection. Concerns over growth and the risk of radiation-associated malignancy make this a poor choice except in desperate cases.

Please take these points as additions to an otherwise excellent article on a difficult subject in the treatment of children with complex spine problems!

References

- 1 Gurjar HK, Sarkari A, Chandra PS. Surgical management of giant multilevel aneurysmal bone cyst of cervical spine in a ten-year-old child: case report with review of literature. *Evid Based Spine Care J* 2012;3(4):55–59
- 2 Varshney MK, Rastogi S, Khan SA, Trikha V. Is sclerotherapy better than intralesional excision for treating aneurysmal bone cysts? *Clin Orthop Relat Res* 2010;468(6):1649–1659
- 3 Rastogi S, Varshney MK, Trikha V, Khan SA, Choudhury B, Safaya R. Treatment of aneurysmal bone cysts with percutaneous sclerotherapy using polidocanol. A review of 72 cases with long-term follow-up. *J Bone Joint Surg Br* 2006;88(9):1212–1216
- 4 Peraud A, Drake JM, Armstrong D, Hedden D, Babyn P, Wilson G. Fatal Ethibloc embolization of vertebrobasilar system following percutaneous injection into aneurysmal bone cyst of the second cervical vertebra. *AJNR Am J Neuroradiol* 2004;25(6):1116–1120
- 5 Fourney DR, Frangou EM, Ryken TC, et al. Spinal instability neoplastic score: an analysis of reliability and validity from the spine oncology study group. *J Clin Oncol* 2011;29(22):3072–3077
- 6 Fourney DR, Gokaslan ZL. Spinal instability and deformity due to neoplastic conditions. *Neurosurg Focus* 2003;14(1):e8
- 7 Panoutsakopoulos G, Pandis N, Kyriazoglou I, Gustafson P, Mertens F, Mandahl N. Recurrent t(16;17)(q22;p13) in aneurysmal bone cysts. *Genes Chromosomes Cancer* 1999;26(3):265–266
- 8 Pietschmann MF, Oliveira AM, Chou MM, et al. Aneurysmal bone cysts of soft tissue represent true neoplasms: a report of two cases. *J Bone Joint Surg Am* 2011;93(9):e45