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EXPERT'S COMMENT

Expert's comment concerning Grand Rounds case entitled "Aneurysmal bone cyst of C2 treated with novel anterior reconstruction and stabilization" by S. Rajasekaran et al. (Eur Spine J; 2016: DOI 10.1007/s00586-016-4518-0)

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Aneurysmal bone cyst (ABC) is a benign bone tumor. Only recently, the neoplastic nature of this disease has been demonstrated [1] concluding speculations about a presumptive hyperplastic nature of this disease, based on slow growing cases, sometimes self-limited cases [2], reported as well as cases of fast and locally aggressive (but never infiltrative) behavior [3].

Aneurysmal bone cyst is well known to be sensitive to radiotherapy [4, 5] and the rate of local recurrence has been reported to be very low after complete intralesional excision [6].

In the historical series, the limit was represented by the difficulty to perform complete intralesional excisions due to typical profuse bleeding, particularly risky in sites like the cervical spine, where it could be very difficult to control. Selective arterial embolization (SAE) became soon precious in reducing the intraoperative blood loss, making it possible to perform complete excision and, therefore, contemporarily achieving the double target to reduce the surgical morbidity and to reduce the local recurrence rate. In our department by chance—as it happens sometimes in scientific discoveries—due to some surgeries not performed for some reason even if SAE had been performed, the positive evolution of the ABC was realized after SAE alone. The literature is abundant [7–11]; the results can be extremely satisfactory even in case of local instability and mild neurological impingement (Fig. 1).

But SAE-alone sequential strategy has some limits: it is not always feasible due to microvascularity, or due to anastomoses with cord vascularity, sometimes many procedures are needed and the patient is exposed to a non-negligible amount of ionizing radiations.

In recent years, more and more therapeutic proposals have been published. They include local injection of ethanol [12, 13], acrylic glue [14], PMMA [15, 16], bioactive glass [17], osteoconductive materials [18], doxycycline [19, 20], calcitonin and steroid [21–24], autologous bone marrow centrifugate [25, 26] (Figs. 2, 3), local treatments like CT-guided radiofrequency ablation [27], percutaneous cryoablation [28, 29] and systemic treatment with bisphosphonates [30], zoledronic acid [31], and denosumab [32–36].

All these strategies appear extremely interesting, due to the low morbidity profile and the high success rate, even in case of presumed instability or moderate neurological compromise.

On the opposite side, many reports and case reviews of ABC are found in the literature about very aggressive surgery, including en bloc resection [3, 37] and highly morbid approaches [38–42], as it would be appropriate for a malignant tumor. In many of these papers, the surgical technique, the staged approaches, the reconstruction required,

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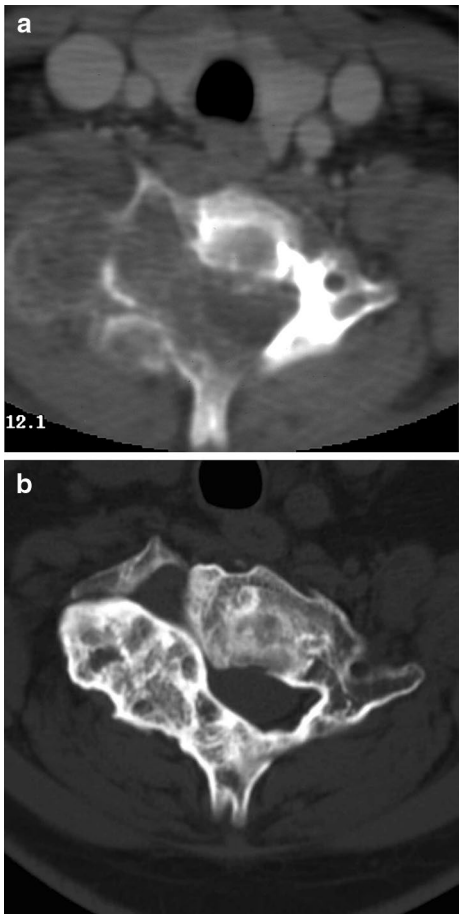

Fig. 1 **a** 14-year-old girl; ABC of C5. Diagnosis achieved by CT-guided trocar biopsy. The amount of the erosion suggests a consistent risk of fracture and she complains of pain and loss of motor function at the right upper limb. After the first SAE, pain started to subside and motor function gradually recovered. **b** CT scan after seven SEA: the reconstruction is completed and motor function is normal. 7 years of clinical follow-up: no evidence of local recurrence

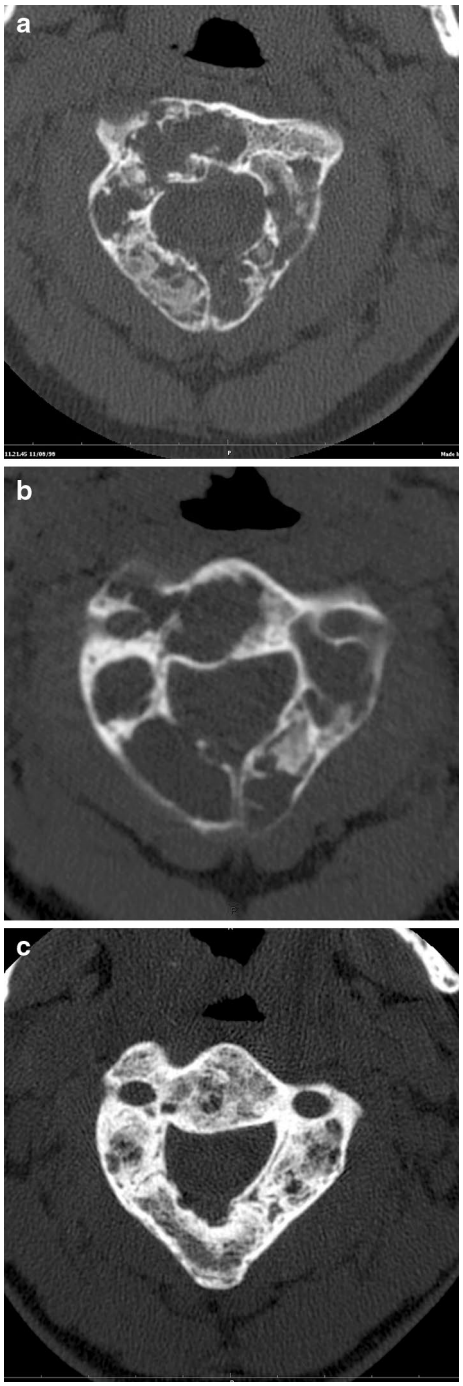

Fig. 2 **a** Trocar biopsy confirmed C2 ABC in a 15-year-old boy. Lytic changes with progressive swelling of the vertebra outline. Moderate pain. **b** SAE partially effective due to microvascularization by deep cervical artery and vertebral artery. After 10 months of observation, the result is satisfactory from a clinical stand point (no pain, limited reduction of the ROM), but the reconstruction was considered not acceptable and one injection of centrifugate autologous (from iliac crest) bone marrow was performed. **c** CT scan control 31 months after the CBM injection and 41 months after the diagnosis. One local CBM injection allowed to achieve an excellent local reconstruction

and obviously, the related morbidity can be expected to be the same as the operated tumor was a chordoma or an osteosarcoma.

This is the objective of this comment. It reports on an excellent surgical technique ending in a complete intralesional excision and requiring a very complex reconstruction [43].

The authors deserve the highest respect for the surgical skill, but I think that a comment could be made on the decision-making process and on the risk to benefit ratio.

When everything—like in this case—goes smooth, everybody is happy (the patients and the relatives first), but I think that the authors of this paper will agree that the morbidity of this procedure is quite consistent: what can happen if another surgeon performs the same surgery and something goes wrong? What can happen if the patients and their relatives (and their lawyer) read in the literature

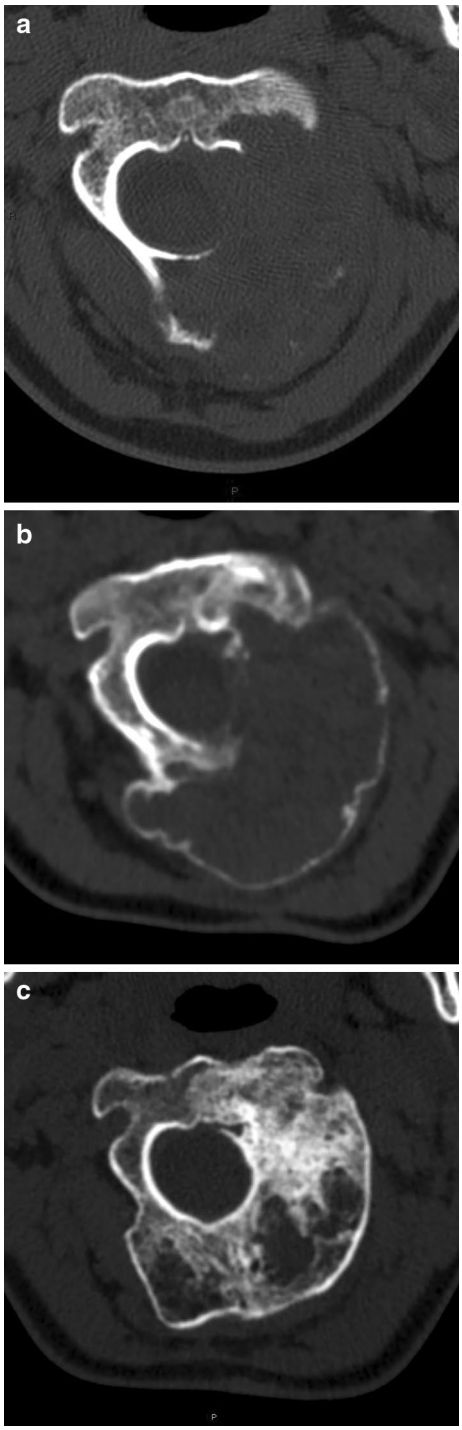

Fig. 3 **a** Trocar biopsy confirmed C2 ABC in a 14-year-old girl. Lytic change of the whole left hemivertebra. Pain and clinical instability. No neurological symptom. **b** SEA with closure of the vertebral artery was not sufficient to stop tumor growth. Protocol of centrifugate bone marrow (CBM) was started. **c** After three CBM injections, reconstruction is achieved. Ossification and tumor mass shrinking. CT scan control 3 years after the diagnosis

that ABC seems to respond extremely well to a number of local and systemic treatments associated with a much lower morbidity?

I think that in front of such a broad spectrum of therapeutic options, before deciding an aggressive and morbid surgical approach, the decision-making process should be based on a very careful discussion including all the other options.

This is particularly relevant as in large cohorts and in the literature reviews [6] no treatment seems to be superior to another as evaluated on the recurrence rate, which is in any case very low, while the morbidity of major surgery (like double approach in higher cervical spine) is obviously significant [44, 45].

Maybe, 20 years ago, we could decide only between surgery and radiotherapy (or a combination of both), but today the number of therapeutic solutions is increasing day by day, supported by positive results. If we can agree that repeated SAE exposes the patient (frequently very young) to high amount of radiation both from the procedure itself and the need of frequent controls, local injection of ethanol, acrylic glue, PMMA, bioactive glass, osteoconductive materials, doxycycline, calcitonin and steroid, autologous bone marrow centrifugate, local treatments like CT-guided radiofrequency ablation, percutaneous cryoablation and systemic treatment with bisphosphonates, zoledronic acid, and denosumab are associated with a negligible morbidity and can be a good solution even in case of low risk of fracture or minor neurological compromise due to the fast local effect. Further, such a wide spectrum of opportunities allows to change from one strategy to another until some result is obtained, leaving excisional surgery as the last resource.

The authors of this article stress on the fact that logistic and patient attitude reason (distant location, reject of halo immobilization) did condition the choice, but I think we should always remember (it is a daily experience in our hospitals) that as soon as something goes wrong all these considerations are vanishing.

I think that a more specific medico-legal opinion on this matter would be helpful, maybe concluding on the need to submit to the patient and/or his/her parents an informed consensus including all the possible treatments, their possible applications in the specific case and their specific morbidity.

I conclude my comment to this article remarking once more the excellent surgical procedure reported, but suggesting the suspect of theoretical overtreating strategy in front of many possible less aggressive alternatives. This aggressive surgery should be performed in case of failure of less morbid solution, and this point should be duly stressed.

Compliance with ethical standards

Conflict of interest The author has no conflict of interest.

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