Surgical Treatment of an Aneurysmal Bone Cyst with Avascular Bone Graft

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Rezumat

Tratamentul chirurgical al unui chist osos anevrismal cu grefon nevascularizat

Introducere: Chistul osos anevrismal este o formațiune tumorală osoasă solitară, expansivă și litică cel mai adesea întâlnită în a doua decadă de viață, mai frecvent la bărbați decât la femei (2:1). Acestea pot apărea la nivelul oricărai os, fiind cel mai des întâlnit la nivelul metafizei oaselor lungi ale membrelor inferioare. Deși este o formațiune tumorală benignă, chistul anevrismal poate avea o evoluție locală agresivă și poate cauza o scădere importantă a rezistenței osoase. Se poate prezenta pentru durere, apariția unei mase tumorale sau apariția unei fracturi pe os patologic. Tradițional aceste leziuni au fost tratate chirurgical (chiuretaj sau resecție și grefare osoasă) cu o rată de recidivă de aproximativ 20%. Deoarece resecția osoasă poate conduce la defecte osoase, deformări sau lezarea funcției membrului afectat în ultima vreme se utilizează scleroterapie percutană cu agenți fibrozații alcoolic. Prezentare de caz: Vă prezentăm cazul unui pacient de 14 ani, prezentat pentru durere și deformare la nivel 1/3 distal al antebrațului drept cu debut insidios și agravat în ultima perioadă. În urma investigațiilor clinice, paraclinice și histopatologice s-a stabilit diagnosticul de chist osos anevrismal cubitus drept. Deoarece scleroterapia nu este încă disponibilă în clinica noastră, inițial s-a practicat o biopsie excisională cu chuirearea leziunii. Tumora a avut în continuare o evoluție agresivă postoperator, astfel incât s-a decis resecția osoasă și reconstrucție cu grefon peronier nevascularizat. Concluzie: Pacientul prezinta o evoluție postoperatorie pe termen scurt și mediu favorabilă, cu dispariția durerilor și reluarea
功能受影响的区域。放射学上可以观察到骨移植物的整合，以及局部复发的缺乏。

**关键词**：骨性骨囊肿，活检，足背肌肉血管性骨移植物，骨重建

**摘要**

**背景**：骨性骨囊肿是一种孤立的骨肿瘤，扩展性和破坏性，通常在20岁出头的男性中多见，且在下肢髓腔中最为常见。虽然它是一个良性肿瘤的形成，骨性骨囊肿可能会有强烈的局部恶变，并导致显著的骨强度降低。患者可能会表现出局部疼痛，局部变形由于肿瘤的生成，或者是病理骨折的发生。传统上，这些病变被通过手术（刮除术或切除骨移植物）治疗，复发率约为20%。因为骨切除可能会导致骨缺陷、变形或损伤受影响肢的功能，近年来更倾向于使用硬化剂（如乙醇）进行血管和微创治疗。

**案例报告**：我们报道了一例14岁患者，因右前臂远端三分之二的疼痛和变形，病程看似缓慢，但近来加剧。根据临床调查、实验室和病理学检查，患者被诊断为右尺骨骨性骨囊肿。由于硬化剂治疗不可在我们的诊所进行，我们最初进行了刮除术和骨移植物刮除术。因为肿瘤仍然具有侵袭性的术后演变，我们决定进行骨切除和使用足背肌肉血管性骨移植物进行再造。术后，患者表现出短期和中期的良好结果，疼痛消失，受影响区段的功能恢复。放射学上可以观察到骨移植物的整合，且无局部复发的证据。

**结论**：虽然现代技术用于治疗骨性骨囊肿包括注射硬化剂或切除和血管性骨移植物，传统的由梅尔·达布吉尔提出的使用血管性骨移植物技术仍然有效，尤其是上肢。

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**背景**

骨性骨囊肿（ABC）是一种孤立的，扩展性，透光性生物体，通常位于长骨的骨化性区域。虽然它们的数量比单纯骨囊肿少，它们占所有主要骨肿瘤样本的1%。骨性骨囊肿最常见于青少年（80%）。骨性骨囊肿可以存在于骨骼的任何区域，最常见的位置是股骨、胫骨、脊柱、肱骨、骨盆和腓骨，大约一半的病例发生在四肢。

骨性骨囊肿可以作为一种原发性骨肿瘤或通过动脉-静脉性病变作为潜在性肿瘤的破坏性影响。骨性骨囊肿可能代表一个原发性骨肿瘤或一个次生反应，发生率约为20%。由于骨性骨囊肿可能会有强烈的局部恶变，并导致显著的骨强度降低。患者可能会表现出局部疼痛，局部变形由于肿瘤的生成，或者是病理骨折的发生。传统上，这些病变被通过手术（刮除术或切除骨移植物）治疗，复发率约为20%。因为骨切除可能会导致骨缺陷、变形或损伤受影响肢的功能，近年来更倾向于使用硬化剂（如乙醇）进行血管和微创治疗。

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association of aneurysmal cysts with other primary lesions, such as nonossifying fibromas, fibrous dysplasia, chondroblastomas, giant cell tumors and simple bone cysts. Over 30% of aneurysmal cysts are believed to be secondary to other lesions, thus once the diagnosis is considered, a thorough preoperative evaluation is necessary, and careful pathologic studies are needed to ensure whether it is a primary lesion or secondary to a different bone tumor (2).

Aneurysmal bone cysts spread rapidly, thus the clinical presentation includes localized pain of several weeks’ or months’ duration, tenderness. The lesions are usually located eccentrically and tend to break out of the bone and expand. This leads to palpable swellings and protuberances. If the cysts involve the spine, progressive enlargement may affect the spinal cord or nerve roots, resulting in neurologic deficits such as motor weakness or sensory disturbance. Spinal involvement mandates urgent intervention.

Radiologically, the aneurysmal bone cyst is an eccentrically located, metaphyseal osteolytic lesion with minimal marginal sclerosing or septum formation. The classic radiographic feature of aneurysmal bone cysts was described by Jaffe as a periostal “blowout” or ballooned-out lesion that is outlined by a thin shell of periosteal new bone formation. The periosteal reaction appears to be aggressive, and the lesion may be mistaken for an aggressive or malignant tumor.

Taking in consideration the location and radiographic findings the aneurysmal bone cyst should differentiated from giant cell tumor, simple bone cyst, but also from a teleangiectatic osteosarcoma.

Aneurysmal bone cysts can have an expansive and aggressive growth and can rapidly reach considerable size. Although spontaneous healing of aneurysmal bone cysts has been reported, it is very uncommon. As a rule, however, when the diagnosis of aneurysmal bone cyst is made, active treatment is recommended. A biopsy should always be performed before starting the surgical treatment as aneurysmal bone cysts can develop from another tumor (5). Management includes combinations of embolization, curettage with or without bone grafting, cementing of the cavity, reconstructive surgery- bone resection and grafting using vascular or avascular bone graft, and most recently sclerotherapy (4,6).

Curettage with or without bone grafting of aneurysmal cysts has been the standard treatment for many years. Unfortunately, this tumor has a high incidence of local recurrence (14% to 59%) after curettage and other treatments have been indicated (4,6).

En-bloc resection of the tumor, as it rarely extends to the epiphysis, followed by reconstruction using a vascular or avascular bone graft is usually preferable. Because bone resection may lead to bone defects, deformations or damage in the affected limb’s function, lately the preferred treatment percutaneous sclerotherapy using fibrosing alcoholic agents (5,6,7).

The use of polidocanol as an endovenous sclerosing agent to treat varicose veins dates from the 1960s, and has been recently shown to be effective in the treatment of ABCs. Sclerosants act by causing damage to the endothelium of vessels and starting a coagulation cascade that results in thrombosis. Overall, sclerotherapy has emerged as a promising treatment that could eventually replace previous methods, which may be associated with considerable morbidity. However, it is not widely accepted and its efficacy remains to be verified in large series of patients (8,9).

**Case report**

Patient A.R. aged 14 presented in our clinic for pain and deformation in the distal third of the right forearm. The symptoms had an insidious onset in the last few months, but were aggravated lately. A X-Ray of the forearm, in PA and L views, was performed and it revealed an osteolytical bone lesion in the distal third of the ulna that produced the thinning and deformation of the cortical bone (“blown out bone” aspect) (Fig. 1). Conservative treatment and careful monitoring was decided. Within a
month the tumor had grown significantly (Fig. 2), therefore an excisional bone biopsy with curettage of the lesion was performed. After the histopathological exam a final diagnosis was established: Aneurysmal bone cyst of the right ulna.

Because the tumor still had an aggressive postoperative evolution, illustrated by the growth of the lesion on the follow-up X-Rays (Figs. 3,4,5) and also by the clinical aspects, large deformation of the distal third of the right forearm (Figs. 6,7,8), a second surgery was decided upon.

A preoperative CT was performed and it revealed an oval osteolytical bone lesion distal third of the ulna, approximately 55 x 32 mm, presenting fluid density, with cortical bone lysis and that doesn’t expand beyond the physis into the epiphysis making more surgical options available (Fig. 9). A 3D reconstruction was performed, that highlighted the lytical and fluid aspects of the cyst (Fig. 10,11).

As sclerotherapy was not yet available in our clinic the selected treatment method was an en-bloc resection and reconstruction using an avascular peroneal bone graft. The tumor was
excised, distally right above the physis and proximally 3 cm above the lesion, and sent to the histology department. Special care was taken during the surgery as to conserve the growth plate and the periosteum. The reconstruction of the bone defect was performed using an avascular graft harvested from the ipsilateral peroneal bone. After performing the reconstruction the graft was fixed in place using a Kirschner wire and the forearm was placed in a cast for 1 month. A postoperative X-Ray was performed the next day after surgery and showed the excision of the cyst and the reconstruction of the bone defect (Fig. 12).

Within the next months the patient had constant follow-ups, consisting of careful clinical examination and forearm X-Rays, which revealed bone formation and graft integration (Fig. 13). The 6 month follow-up X-Ray showed complete integration of the graft and almost complete healing of the donor site (Fig. 14).

The one year postop X-Ray showed healing and integration of the graft in the distal ulna (Fig. 15) therefore the removal of the K wire was performed. Postoperative evolution was good, with normal function of the limb and no local pain.

An X-Ray was performed 3 months after the removal of the K wire, showing good local evolution (fig. 16).

Conclusion

In conclusion, the fact that postoperative, the patient presents a favourable short and medium term evolution, the disappearance of pain and resumed function of the affected
segment correlated with radiologically observed bone graft integration and with the absence of local recurrence demonstrates that although modern techniques for treating aneurysmal bone cyst include either injecting fibrosing alcoholic agents or reconstruction using vascular bone graft, the traditional technique described by Merle d’Aubigne which implies the usage of avascular bone graft is still helpful, leading to successful results especially in the upper limbs.

Conflicts of interest
No interest conflict to report.

References