CASE REPORT

RJME
Romanian Journal of Morphology & Embryology
http://www.rjme.ro/

Unicameral bone cyst of the calcaneus – minimally invasive endoscopic surgical treatment. Case report

IOAN CRISTIAN STOICA[1,2], DOINA MIHAELA POP[1,2], FLORIN GROSU[3]

[1] *Department of Orthopedics and Trauma, "Carol Davila" University of Medicine and Pharmacy, Bucharest, Romania*

[2] *"Foişor" Orthopedics Hospital, Bucharest, Romania*

[3] *Department of Histology, "Victor Papilian" Faculty of Medicine, "Lucian Blaga" University of Sibiu, Romania*

Abstract

The role of arthroscopic surgery for the treatment of various orthopedic pathologies has greatly improved during the last years. Recent publications showed that benign bone lesion may benefit from this minimally invasive surgical method, in order to minimize the invasiveness and the period of immobilization and to increase visualization. Unicameral bone cysts may be adequately treated by minimally invasive endoscopic surgery. The purpose of the current paper is to present the case report of a patient with a unicameral bone cyst of the calcaneus that underwent endoscopically assisted treatment with curettage and bone grafting with allograft from a bone bank, with emphasis on the surgical technique. Unicameral bone cyst is a benign bone lesion, which can be adequately treated by endoscopic curettage and percutaneous injection of morselized bone allograft in symptomatic patients.

Keywords: unicameral bone cyst, endoscopic procedure, curettage, bone allograft.

Introduction

Primary bone cysts are rare tumor lesions, commonly occurring in men, the female/male ratio being 1/2.5 [1, 2]. They mainly affect the long bones, especially the proximal extremity of the humerus and femur; still, they may occur in any bone. Even though they are benign lesions, various studies showed that these may turn into malignant tumors [3, 4]. These lesions are most frequently diagnosed in children and young people. It is considered that 3/4 of these lesions occur in the first two life decades and almost 95% are diagnosed in the first life decades [4, 5].

There are two types of bone cysts: simple or unicameral bone cysts and aneurysm bone cysts. The etiology of these lesions is still unknown.

Unicameral bone cyst is a benign fluid-filled bone lesion, present more frequently in long bones. Although it affects more frequently the humerus and the femur, in a minority of cases, the lesion may occur at the level of the calcaneus [6–8]. There is a consensus that surgery is indicated for patients with persistent pain or for patients with increased risk of pathological fractures [6, 7, 9, 10]. Traditionally, open curettage with bone grafting of the lesion has been the procedure of choice for unicameral bone cyst of the calcaneus with excellent clinical results [6, 7, 9, 11]. Recently, less invasive surgical procedures have been used for the treatment of these cases with earlier return to normal activities. Unicameral bone cysts may be adequately treated by minimally invasive endoscopic surgery.

The role of arthroscopic surgery for the treatment of various orthopedic pathologies has greatly improved during the last years. Recent publications demonstrated that benign bone tumors may benefit from this minimally invasive surgical method, in order to minimize the invasiveness and the period of immobilization and to increase visualization [12].

The purpose of the current paper is to present the value of endoscopically assisted treatment with curettage and bone grafting with allograft from a bone bank for patients with benign bone tumors and tumor-like lesions, by presenting the case report of a patient with unicameral bone cyst of the calcaneus, emphasizing the importance of correlating clinical, imagistic and histopathological data.

Case presentation

A 20-year-old male (SVA) presented in the Department of Orthopedics and Trauma, "Foişor" Orthopedics Hospital, Bucharest, Romania, with the main complaint of pain at hindfoot level during the last six months, without significant improvement with conservative treatment. Pain was of mild to moderate intensity; it was partially relieved by anti-inflammatory medication and rest and aggravated by activity. There was no personal or family history of trauma or other pathological conditions at the level of the affected foot. The general clinical exam showed no pathological characteristics. On physical examination, localized pain on palpation was identified on the lateral side of the hindfoot. Lab tests were within the normal range. Radiographs (antero-posterior, lateral and axial views of the calcaneus) revealed a centrally located, well-delimitated lytic lesion, with no cortex penetration or peri-osteal reaction (Figure 1, A–C). Computed tomography (CT) and magnetic resonance imaging (MRI) demonstrated

the typical imagistic features of this type of bone lesion, without any sign of stress fracture. Surgery was indicated for the treatment. The patient was informed that the case data would be submitted for publication, and we obtained the written consent. The Ethics Committee of the "Foişor" Orthopedics Hospital approved this case report for publication.

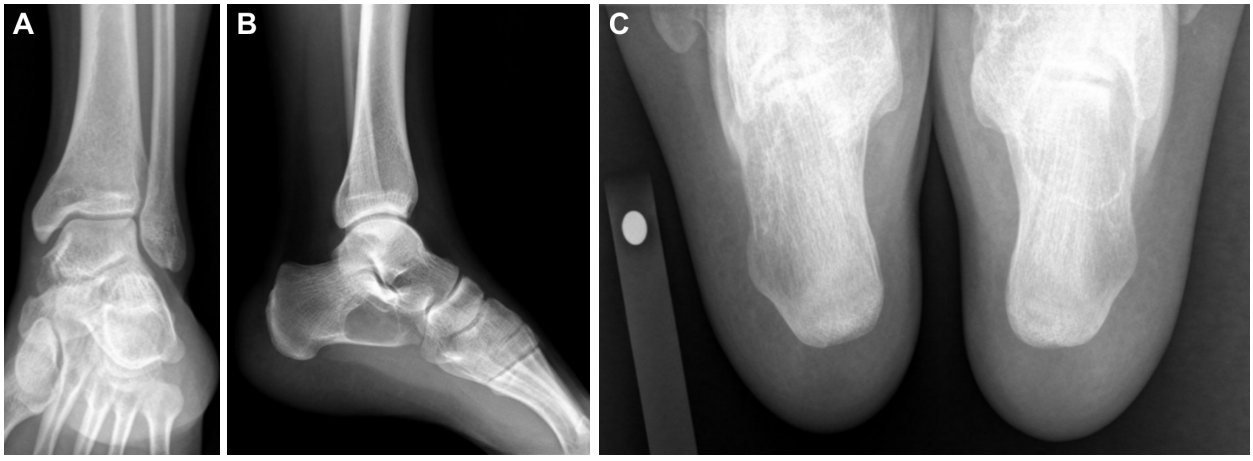

Figure 1 – ***Preoperative antero-posterior (A) and lateral view (B) of the ankle and axial view (C) of the calcaneus.***

The surgical treatment consisted of biopsy with intra-operative histopathological exam, curettage and bone grafting with femoral head allograft from bank. Endoscopy was used to reduce the invasiveness of the surgical procedure and the period of postoperative immobilization and to increase visualization during surgery.

In the current case, curetted fragments were taken from the thinned cortex surrounding the cyst membrane.

Surgical technique

Surgery was performed under spinal anesthesia. The patient was placed in lateral decubitus and with application of a tourniquet at the level of the thigh (Figure 2). The configuration of the calcaneal unicameral bone cyst was determined under image intensifier and marked on the lateral aspect of the hindfoot (Figure 3).

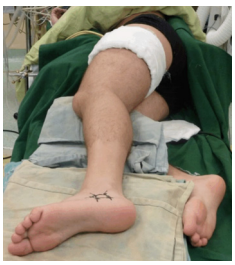

Figure 2 – ***Patient positioning.***

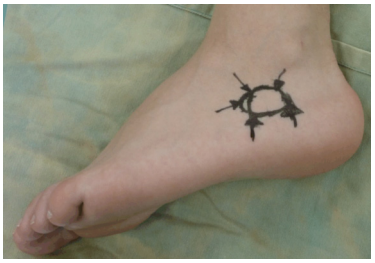

Figure 3 – ***The configuration of the calcaneal bone cyst marked on the lateral aspect of the hindfoot.***

We inserted a guide wire at the center of the bone cyst and we introduced successively trocars of progressively increasing diameter in order to create a bone window to allow aspiration of the intra-cystic fluid and adequate access for the endoscopic procedure (Figure 4).

We performed aspiration of the fluid and a detailed inspection of the bone cyst *via* endoscopic view. Successively, we obtained a biopsy sample for histological examination in order to have a final diagnosis. After histological confirmation of the diagnosis, reduction of the endoscopic blind area was obtained by resecting the inner bony septum with a suction shaver. Under the resulting optimal endoscopic visualization, we used a small curette to perform circumferential resection of the inner fibrous tissue with care to prevent excessive cortical thinning, which may increase the risk of postoperative iatrogenic fracture (Figure 5). After irrigation, morselized bone allograft from the bank we injected percutaneously through the previously created cortical window and impacted (Figure 6A). Radiographic confirmation of the adequate tumor cavity bone grafting was obtained intra-operatively (Figure 6B).

Histopathological data

Fresh fragments were evaluated by intra-operatory frozen sections and was negative for neoplastic lesion. Afterwards, the whole bioptic material was histologically processed to paraffin blocks. Thin 4 μm sections were obtained and stained with Hematoxylin and Eosin (HE) by standard procedures. Histopathological examination showed that it was an inactive, old lesion that had a thicker mesenchymal membrane, cholesterol clefts and ribbon-like fibrinous material (Figures 7 and 8). However, in our case, histopathological features of the bone cysts was polymorphic, showing some active aspects of the lesion, relatively nonspecific, with the thin membrane composed of mesenchymal cells, capillaries, scattered osteoclasts and little hemosiderin (Figure 9). New bone formation was also seen, in which the osteoblastic active rimming was prominent, suggesting a reparative postfractural process (Figure 10).

Follow-up included clinical and radiological examination at six weeks, three, six and 12 months, and yearly thereafter until the latest postoperative visit at five years after surgery. No pain and good functional results were present at any of the follow-up intervals. No radiological sign of recurrent unicameral bone cyst could be detected, correlated with progressive integration of the bone graft as demonstrated by the presence of normal trabecular bone structure (Figure 11).

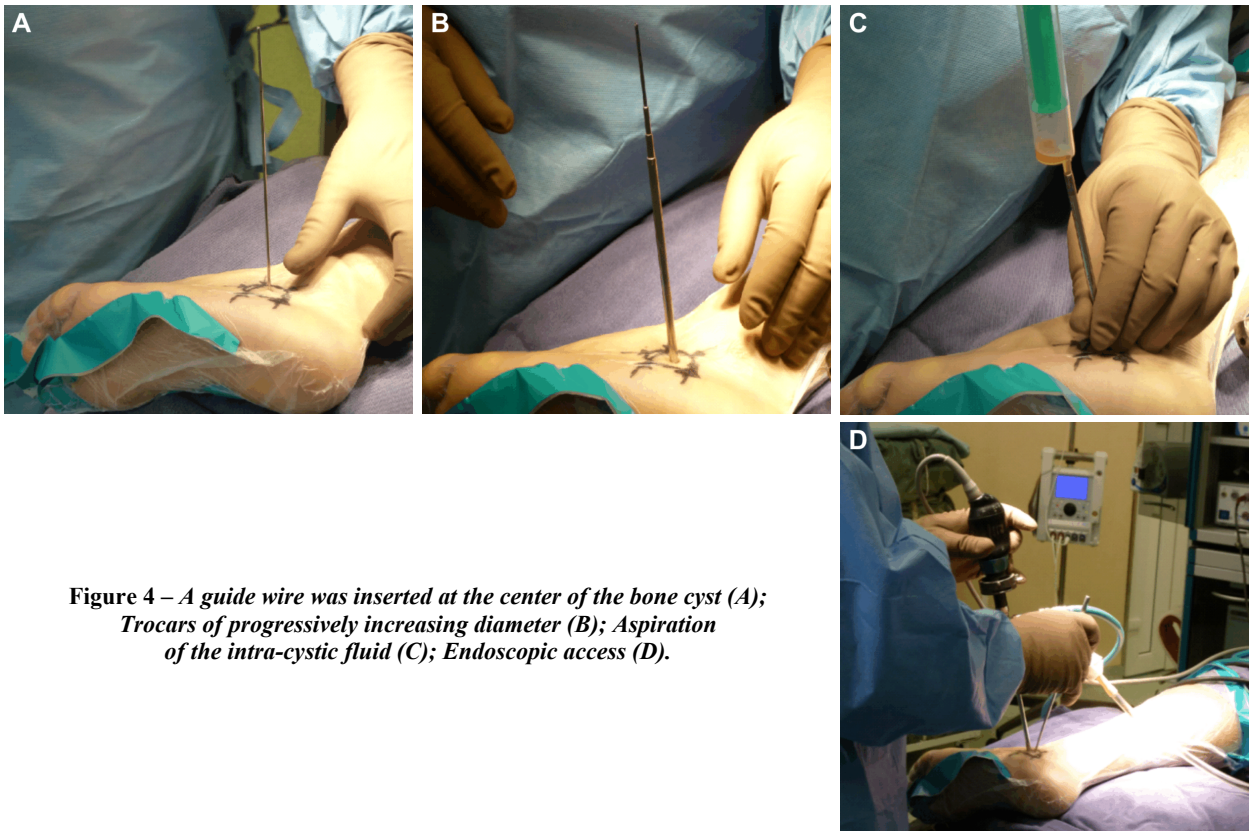

Figure 4 – *A guide wire was inserted at the center of the bone cyst (A); Trocars of progressively increasing diameter (B); Aspiration of the intra-cystic fluid (C); Endoscopic access (D).*

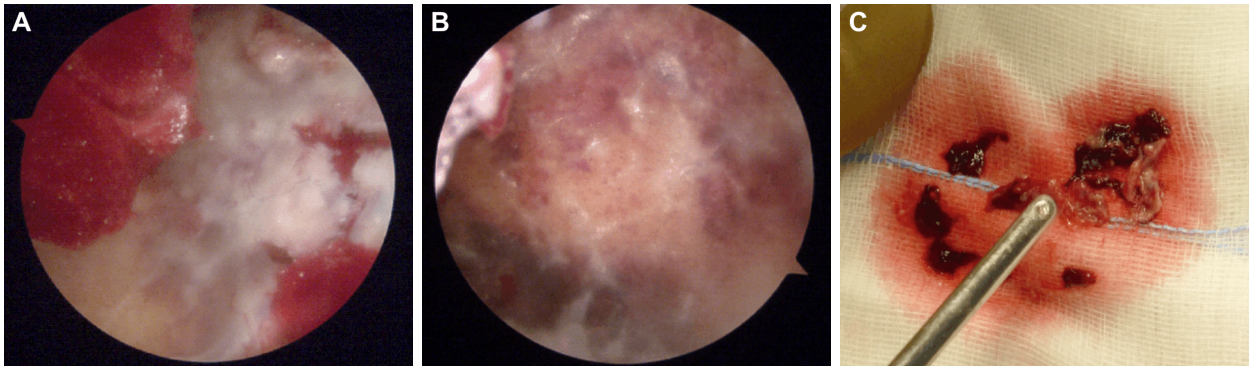

Figure 5 – *Endoscopic view of the bone cyst before resection (A) and after resection (B); Macroscopic aspect of the resected tissue (C).*

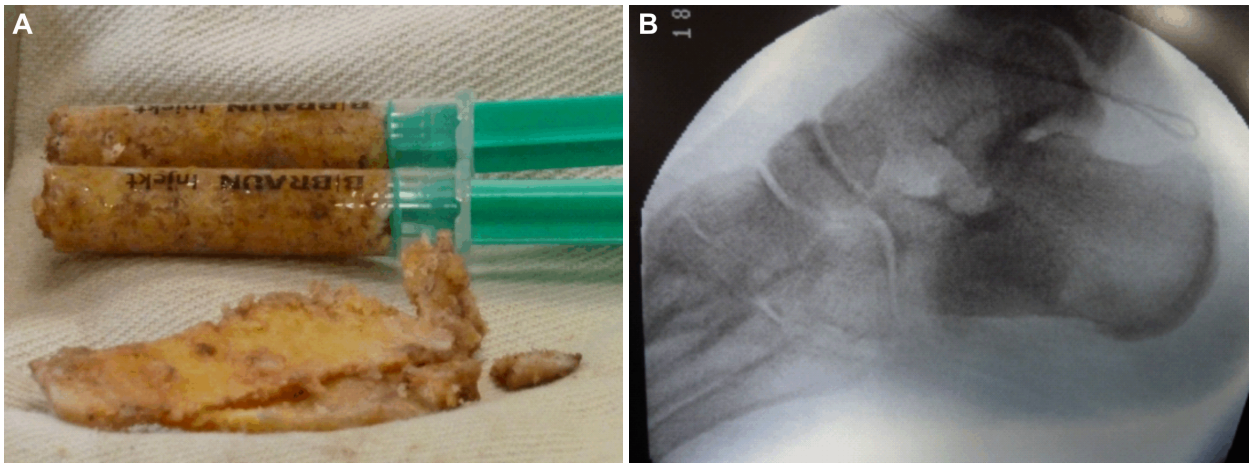

Figure 6 – *Morselized bone allograft prepared for injection in the cyst cavity (A) and intraoperative radiographic confirmation of adequate bone grafting (B).*

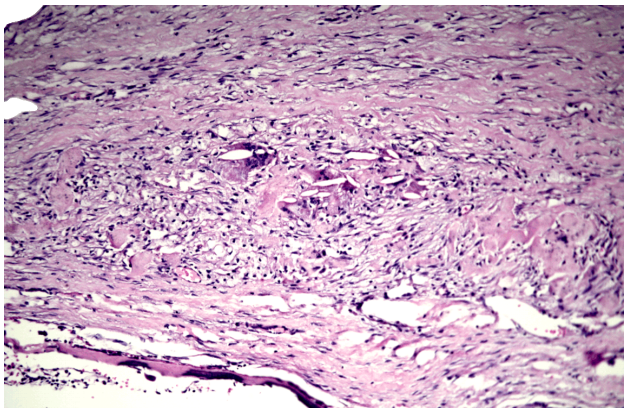

Figure 7 – ***Pink cementum-like material representing fibrin in old unicameral cyst, mesenchymal membrane and cholesterol clefts (HE staining, ×200).***

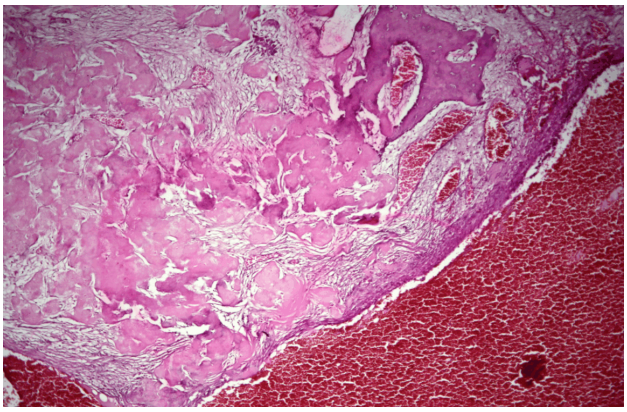

Figure 8 – ***Aneurysmal bone cyst like features in bone simple cyst with large blood filled spaces lined by connective tissue, cementum-like material and new bone formation (HE staining, ×100).***

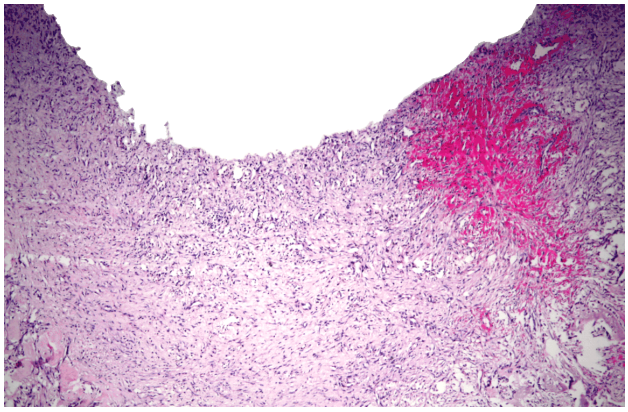

Figure 9 – ***Active aspects of simple bone cyst composed of mesenchymal cells, capillaries and a few scattered inflammatory cells (HE staining, ×100).***

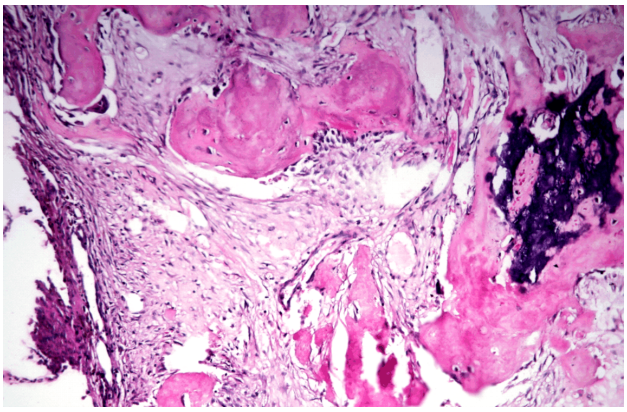

Figure 10 – ***New bone formation, calcifications and hemosiderin deposits in bone simple cyst (HE staining, ×200).***

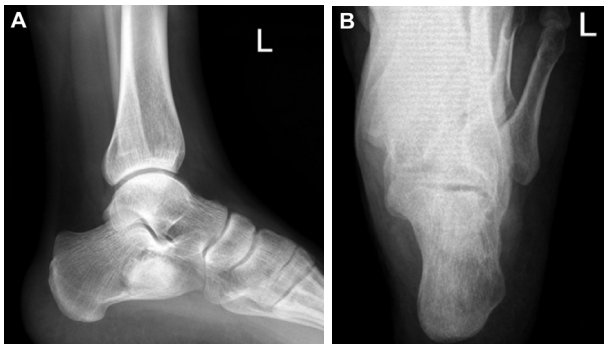

Figure 11 – ***(A and B) Postoperative radiological exam at five years follow-up demonstrates integration of the bone graft, with no sign of recurrent tumor.***

Discussion

The unicameral bone cyst is a benign bone lesion with a high recurrence rate, representing 3% of all tumor bone lesions [13, 14].

It is common during the first two decades of life, being twice more frequent in male patients than in female patients. The most common locations in children are represented by the proximal humerus and femur, while in the adult population the ilium and the femur are typically affected. The lesions are active during the period of skeletal growth and generally spontaneous healing occurs at maturity.

Our patient was a young man who presented with pain at hindfoot level for six months, without improvement after conservative treatment. The radiological examination highlighted the lesions and benign characteristics. Most studies showed that the calcaneus localization of the bone cysts is quite rare [1, 12].

In most of the cases, unicameral bone cysts are asymptomatic, being an incidental discovery. However, in some cases, a pathological fracture may be possible. Plain radiographs reveal a centrally located, well-delimitated lytic lesion. The unicameral bone cyst never penetrates the cortex and no periosteal reaction is present [6, 11, 15–17]. The preoperative diagnosis is usually straightforward but may be difficult in the presence of reparative postfractural changes or if a secondary aneurysmal bone cyst is associated.

Small and asymptomatic lesions in the upper extremities, with low fracture risk, can be treated conservatively, with serial plain radiographs. Larger lesions or symptomatic lesions in the lower extremities, with a higher fracture risk, are usually treated with curettage with bone grafting or aspiration and corticosteroid, autogenous bone marrow or demineralized bone matrix injection. More than 90% of patients can be successfully treated by repeated aspiration and corticosteroid injection. Pathological fractures through the unicameral bone cyst in the lower extremity should be treated with curettage, bone grafting and internal fixation. However, pathological fractures in the upper

extremity can be treated conservatively because the fracture may initiate the cyst healing process [18–21].

These lesions are not neoplastic. Presumably, they represent a local disorder of development and bone growth or as an alternative pathogenesis a synovial origin [22]. Macroscopically, these lesions present as unilocular clear or straw-colored fluid-filled cavities within the medulla. As we have mentioned, histopathological features of the bone cysts (simple) are polymorphic, some especially in children are active lesions and relatively nonspecific with the thin membrane composed of mesenchymal cells, capillaries, scattered osteoclasts and little hemosiderin. New bone formation may be seen in which the osteoblastic active rimming is prominent. In any case, a secondary aneurysmal bone cyst could be associated as well as fracture callus formation furthermore complicating the pathological aspects [23, 24].

Endoscopically assisted treatment with curettage and bone grafting with allograft from a bone bank for patients with benign bone tumors and tumor-like lesions is a newly modern minimally invasive treatment option in the current practice. It is of great importance to correlate clinical, imagistic and histopathological data for improving the final clinical outcome.

Conclusions

The current case report demonstrated that unicameral bone cyst, which is a benign bone lesion, can be adequately treated by endoscopic curettage and percutaneous injection of morselized bone allograft in symptomatic patients. The current case was followed-up for five years, without any recurrence. This emphasizes that minimally invasive endoscopic treatment may not be associated with increased surgical complications or recurrence rate. However, a larger series of patients with longer follow-up is needed for confirming this data.

Conflict of interests

There is no conflict of interests.

References

[1] Mascard E, Gomez-Brouchet A, Lambot K. Bone cysts: unicameral and aneurysmal bone cyst. Orthop Traumatol Surg Res, 2015, 101(1 Suppl):S119–S127.

[2] Boude AB, Vásquez LG, Alvarado-Gomez F, Bedoya MC, Rodríguez-Múnera A, Morales-Saenz LC. A simple bone cyst in cervical vertebrae of an adolescent patient. Case Rep Orthop, 2017, 2017:8908216.

[3] Brindley GW, Greene JF Jr, Frankel LS. Case reports: malignant transformation of aneurysmal bone cysts. Clin Orthop Relat Res, 2005, 438:282–287.

[4] Kapoor C, Shah M, Soni R, Patwa J, Merh A, Golwala P. Aneurysmal bone cyst of the proximal femur and its management – a case report. Cureus, 2017, 9(1):e991.

[5] Bonakdarpour A, Levy WM, Aegerter E. Primary and secondary aneurysmal bone cyst: a radiological study of 75 cases. Radiology, 1978, 126(1):75–83.

[6] Smith RW, Smith CF. Solitary unicameral bone cyst of the calcaneus. A review of twenty cases. J Bone Joint Surg Am, 1974, 56(1):49–56.

[7] Pogoda P, Priemel M, Linhart W, Stork A, Adam G, Windolf J, Rueger JM, Amling M. Clinical relevance of calcaneal bone cysts: a study of 50 cysts in 47 patients. Clin Orthop Relat Res, 2004, (424):202–210.

[8] Wilkins RM. Unicameral bone cysts. J Am Acad Orthop Surg, 2000, 8(4):217–224.

[9] Glaser DL, Dormans JP, Stanton RP, Davidson RS. Surgical management of calcaneal unicameral bone cysts. Clin Orthop Relat Res, 1999, (360):231–237.

[10] Abdel-Wanis ME, Tsuchiya H, Uehara K, Tomita K. Minimal curettage, multiple drilling, and continuous decompression through a cannulated screw for treatment of calcaneal simple bone cysts in children. J Pediatr Orthop, 2002, 22(4):540–543.

[11] Moreau G, Letts M. Unicameral bone cyst of the calcaneus in children. J Pediatr Orthop, 1994, 14(1):101–104.

[12] Toepfer A, Lenze U, Gerdesmeyer L, Pohlig F, Harrasser N. Endoscopic resection and allografting for benign osteolytic lesions of the calcaneus. Springerplus, 2016, 5:427.

[13] Cohen J. Etiology of simple bone cyst. J Bone Joint Surg Am, 1970, 52(7):1493–1497.

[14] Rosario MS, Yamamoto N, Hayashi K, Takeuchi A, Kimura H, Miwa S, Higuchi T, Inatani H, Abe K, Taniguchi Y, Aiba H, Tsuchiya H. An unusual case of proximal humeral simple bone cyst in an adult from secondary cystic change. World J Surg Oncol, 2017, 15(1):102.

[15] Mik G, Arkader A, Manteghi A, Dormans JP. Results of a minimally invasive technique for treatment of unicameral bone cysts. Clin Orthop Relat Res, 2009, 467(11):2949–2954.

[16] Levy DM, Gross CE, Garras DN. Treatment of unicameral bone cysts of the calcaneus: a systematic review. J Foot Ankle Surg, 2015, 54(4):652–656.

[17] Chigira M, Maehara S, Arita S, Udagawa E. The aetiology and treatment of simple bone cysts. J Bone Joint Surg Br, 1983, 65(5):633–637.

[18] Innami K, Takao M, Miyamoto W, Abe S, Nishi H, Matsushita T. Endoscopic surgery for young athletes with symptomatic unicameral bone cyst of the calcaneus. Am J Sports Med, 2011, 39(3):575–581.

[19] Alvarez RG, Arnold JM. Technical tip: arthroscopic assistance in minimally invasive curettage and bone grafting of a calcaneal unicameral bone cyst. Foot Ankle Int, 2007, 28(11):1198–1199.

[20] Mainard D, Galois L. Treatment of a solitary calcaneal cyst with endoscopic curettage and percutaneous injection of calcium phosphate cement. J Foot Ankle Surg, 2006, 45(6): 436–440.

[21] Park IH, Micic ID, Jeon IH. A study of 23 unicameral bone cysts of the calcaneus: open chip allogeneic bone graft *versus* percutaneous injection of bone powder with autogenous bone marrow. Foot Ankle Int, 2008, 29(2):164–170.

[22] Mirra JM, Bernard GW, Bullough PG, Johnston W, Mink G. Cementum-like bone production in solitary bone cysts. (so-called "cementoma" of long bones). Report of three cases. Electron microscopic observations supporting a synovial origin to the simple bone cyst. Clin Orthop Relat Res, 1978, (135):295–307.

[23] Amling M, Werner M, Pösl M, Maas R, Korn U, Delling G. Calcifying solitary bone cyst: morphological aspects and differential diagnosis of sclerotic bone tumours. Virchows Arch, 1995, 426(3):235–242.

[24] Campanacci M, Capanna R, Picci P. Unicameral and aneurysmal bone cysts. Clin Orthop Relat Res, 1986, (204): 25–36.

Corresponding author

Ioan Cristian Stoica, Associate Professor, MD, PhD, Department of Orthopedics and Trauma, "Carol Davila" University of Medicine and Pharmacy, "Foişor" Orthopedics Hospital, 35–37 Ferdinand I Avenue, Sector 2, 021382 Bucharest, Romania; Phone +4021–252 13 87, Mobile +40722–292 770, e-mail: stoicacristianioan@gmail.com

Received: April 30, 2016 *Accepted: July 14, 2017*