

“Three-in-One minimally invasive approach to surgical treatment of pediatric pathological fractures with wide bone loss through bone cysts: ESIN, curettage and packing with injectable HA bone substitute. A retrospective series of 116 cases.”



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ABSTRACT

Introduction: The outcome of pathological fracture due to large aggressive benign stage 3 Dormans and Flynn lesions [6] is often unsatisfactory and the rate of recurrence is high. No single technique has been considered safe and successful. Many Authors suggested curettage and bone grafting as the unique effective treatment in cases of large defect but, because of the invasive and complex nature of the operation (it needs a double-step procedure), it is not preferred. The purpose of this study is to examine the effectiveness of a minimally invasive treatment in one step through ESIN, curettage and packing with self-setting calcium phosphate cement.

Patients and Methods: This is a retrospective study of 116 children admitted at the Division of Pediatric Orthopaedics Surgery of Santobono Children Hospital between 2006 and 2014 with a diagnosis of pathological fracture due to large aggressive osteolytic benign lesions (stage 3 Dormans and Flynn). The size of bone loss was measured on the AP and ML radiographs and all the cysts with a caudo cranial extension from 5 to 8 cm and with a medio lateral extension from 3 to 5 cm were included. Mean time follow up 24 months was performed.

Results: The three-in-one procedure was applied in all 116 patients. After two years of follow up, 113 patients were classified as healed and just 3 required 3 years to complete heal. No severe life threatening adverse effects or complications associated with the use of ESIN and injectable HA were recorded during the follow up period of 24–36 months. Fracture healing occurred in all cases within 4–6 weeks with adequate periosteal and endosteal callus formation. No second pathological fractures occurred in our series as well as no cysts reoccurred. Patients with humeral localizations showed a more rapid regain of muscular function and reestablishment of a complete range of motion.

Conclusions: The proposed three-in-one procedure has shown to be efficient, cost-effective, associated to high rates of definitive bone healing and low incidence of adverse effects.

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Introduction

Bone cysts are generally benign tumoral lesions which may be associated with pathologic fractures due to the increased fragility of the involved bone which is lacking the viscoelastic properties and mechanical strength of normal bone [1,2].

These lesions can show different behavior according to their aggressiveness. Stage 1, or latent benign lesions, are generally asymptomatic, discovered accidentally and can heal spontaneously or remain static. They are rarely associated with pathological fractures. Stage 2, or active benign lesions, which represent the widest group, tend to grow steadily, are limited by natural barriers and they can be symptomatic. Stage 3, or aggressive benign lesions, are generally symptomatic and tender, grow rapidly and are frequently associated with pathologic fractures. This stage 3 lesions often have a more aggressive natural history and include

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Aneurysmal bone cysts (ABCs) and Aggressive Unicameral Bone Cysts (UBCs) together with other malignant tumors [3,4].

This study focuses on pathological fractures in ABCs and aggressive UBCs in children and on clinical management through a new approach described by the Authors.

When pathological fractures occur in stage 3 lesions there are two simultaneous challenges: the treatment of the fracture sometimes involving the growth plate and the management of the condition responsible for the fracture [2].

Several surgical techniques are available to treat bone cysts lesions with an inconstant rate of success [5–7].

Traditionally simple curettage combined with bone grafting is the most performed surgical procedure. Open curettage and bone grafting with autogenous or allograft bone has been associated with a recurrence risk ranging from 12% to 45% in the UBCs, while in ABCs the recurrence rate is higher [5,8–11].

Intracystic injections of methylprednisolone (Scaglietti's procedure) have induced a partial response in 50% to 80% of patients, but this treatment option has been limited by the need for repeated injections and a significant rate of incomplete healing [12–16].

First reports with elastic Ender nails were described by Santori et al. in 1986. They noticed healing in all their cases and suggested that relieving the internal pressure of the cyst could promote its healing. These results were also confirmed by de Sanctis and Andreacchio in their study [17–20].

Selective arterial embolisation can be used for treating ABCs but it is not always possible to perform because the vessels which feed the lesion do not guarantee success [21,22].

Adjuvants such as phenol, alcohol and cryotherapy with liquid nitrogen have also been used in the treatment of UBCs and ABCs without optimal results [23–27].

An endoscopic approach has been proposed but larger lesions were difficult to dominate [28].

A variety of grafting materials, with osteoconductive and osteoinductive properties, have been used with minimally invasive grafting techniques. Pellets made of radiopaque, medical grade calcium sulfate can be placed percutaneously in bone cysts [29–34]. But these materials show poor mechanical resistance so weight bearing is forbidden. Among osteoconductive stimulants, demineralized bone matrix has been used [35].

Finally we present our experience in managing pathological fractures with wide bone loss in ABCs and aggressive UBCs (stage 3 according to Dormans and Flynn classification). We combined a three-in-one minimally invasive approach: stabilization of the fracture-decompression of cystic areas with Titanium Elastic Stable Intramedullary Nailing (ESIN/TEN), mechanical curettage and packing with injectable calcium phosphate through the same minimally invasive surgical access.

We believe that the combination of fracture stabilization, intramedullary decompression, mechanical curettage and bone substitute packing together will be both effective and with the advantages of Minimally Invasive Surgery (MIS), safe allowing the outpatient to return to full activities in a shorter period of time.

To our knowledge there are no other descriptions in the literature for this specific treatment of pathological fractures involving stage 3 bone cysts.

Patients and methods

This is a retrospective study of 116 children who were consecutively admitted at the Surgical Science Department Division of Pediatric Orthopaedics Surgery of Santobono Children Hospital between 2006 and 2014 with a diagnosis of pathological fracture due to large aggressive osteolytic benign lesions (stage 3 Dormans and Flynn)

The series was composed of 83 males and 33 females, with a mean age of 9.2 years (range 2–16 years). Type of underlying lesions were Aneurysmal Bone Cysts (ABC) in 74 patients and Unicameral Bone Cysts (UBC) in 42 patients. (Table 1) Follow up 24 months by surgery.

The localization of the lesions was humerus (epiphysis and metaphysis) in 79 patients; 9 of them showed invasion of the physis. Femur instead (epiphysis and metaphysis) was involved in 37 patients. (Table 2)

Inclusion criteria were: (a) the size of the bone loss and pathological fracture, (b) the large extension of thinner cortices, (c) the very low density of osteolytic areas at x-rays, (d) the large width of lesion. In our series the size of bone loss was measured on the AP and ML radiographs and all the cysts with a caudo cranial extension from 5 to 8 cm and with a medio-lateral extension from 3 to 5 cm were included. (Tables 3 and 4)

In all the patients we performed same technique: a three-in-one minimally invasive approach. During the same operatory session we firstly performed the stabilization of the pathological fracture by means of intramedullary retrograde nailing with two elastic titanium nails (Elastic Stable Intramedullary Nailing)

We followed a classic retrograde nailing surgical approach as reported by Metaizeau et al. [36].

Displaced pathological fractures were first reduced. The diameter and length of the nails were selected on the basis of measurements made with a tape on the preoperative anteroposterior (AP) radiograph. The diameter of the nails was selected such that two nails would occupy approximately two thirds of the medullary canal, and their tips can pass the growth plate of the bones.

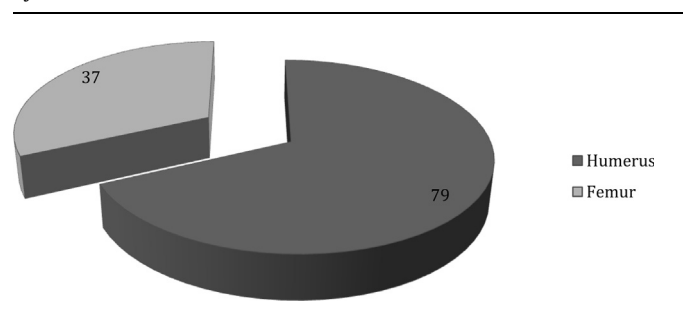
For the humerus the access was through two supracondylar 1.5 cm miniportals on the medial and on the lateral aspect of the humerus or twice lateral. In those cases in which cystic lesions were abutting the proximal growth plate of the humerus maximum of 2.5 mm nails could be driven across the physis (Fig. 1).

For the femur the approach was through two supracondylar 2.5–3 cm miniportals on the medial and on the lateral aspect of the

Table 1
Sample features.

Observational period	2006–2014
Cases	116
Age	2–16
Sex	83 Males 33 Females
Mean age of onset	9.2
Underlying lesion	74 ABCs 42 UBCs
Follow up	24 months

Table 2
Cysts localization.



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