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GIANT NONOSSIFYING FIBROMA OF THE DISTAL TIBIA IN ADOLESCENTS TREATED BY CURETTAGE AND INTRALESIONAL CEMENTATION

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Abstract

Background. Fibrous cortical defects and nonossifying fibromas are the most common benign non-neoplastic bone lesions occurring in the metaphyses of long bones in children and adolescents. Giant nonossifying fibromas (NOF) are not asymptomatic but usually present with pain and/or pathological fractures due to increased stress.

Materials and methods. 20 adolescent patients, 14 males and 6 females, with mean age 18 years and 6 months (range, from 16 to < 21 years); presented to the National Institute of Neuromotor system, Egypt, between September 2007 and September 2009, with giant nonossifying fibromas of the distal tibia. Diagnosis was made by clinical examination, plain radiographs, magnetic resonance imaging, and histopathological reports. Treatment was achieved by curettage without bone graft, but with intralesional filling with bone cement. Evaluation concerning pain, functional activity using MSTS scoring, pathological fracture, and local recurrence were done over a mean follow-up period of 6 years and 2 months (range, 5 to 7 years).

Results. Pain and functional activity improved in the twenty patients with mean MSTS score of 29.2 (range, 25 to 30). There was no pathological fracture, no local recurrence, no change in the cement-bone interface, and no arthrogenic problems over the follow-up period. The p value was <0.05.

Conclusion. Giant nonossifying fibromas can be treated simply and effectively by curettage and intralesional cementation with excellent functional results.

Key words: nonossifying fibromas, fibrous cortical defects, benign bone lesions.

Introduction

Nonossifying fibromas (NOF) are developmental lesions which commonly affect children and adolescents, mainly in the lower extremity [1, 2, 5, 9]. Most of the cases are asymptomatic and accidentally discovered. However, giant osseous lesions (>30 mm in diameter, or encroaching to >50% of the affected bone) can be alarming, being presenting with pain and possible pathological fracture [3].

The aim of the study was to diagnose symptomatic cases of giant nonossifying fibromas in the distal tibia of adolescents, and evaluate the results of curettage of the lesions without autogenous bone grafting, but with intralesional filling with bone cement, on the improvement of pain, the functional activity, the protection from pathological fractures, and the recurrence of the lesions.

Materials and methods

A retrospective review was prepared collecting a group of twenty patients who presented to the National Institute of Neuromotor system (Egypt) between September 2007 and September 2009 with giant nonossifying fibromas of the distal tibia treated by curettage without bone graft, but with intralesional filling with bone cement. The institute provides health service to the skeletally disabled and handicapped children from all over Egypt that explains the relatively large number of this lesion collected from one centre. The results in this group after at least five years and up to more than seven years of follow-up were reported in September 2014 using previous medical records, history, physical examination, plain radiography, MRI, and histopathological reports. The study fulfills the Egyptian's Ethics code of researches.

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Inclusion criteria. Giant nonossifying fibromas, in the distal tibia of adolescents, between 16 years to <21 years of age; as evidenced by plain radiography, magnetic resonance imaging, and histopathology, without pathological fractures, were chosen.

Exclusion criteria. Other giant intraosseous lesions in the distal tibia as unicameral bone cysts, aneurismal bone cysts, chondromyxoid fibromas, giant cell tumours, and infections; or lesions with pathological fractures, were excluded.

Preoperative clinical evaluation, original disease, and treatment. The mean age of the patients at the time of surgery was 18.5 years ranging from 16 to <21 years. They were 14 male and 6 female patients. The right side was affected in 8 male and 4 female patients; whereas the left side was affected in 6 male and 2 female patients. There was no bilateral affection (Table 1). Lower leg and ankle pain were present in all patients for an average of 2 months (range, 1 to 3 months), but not in other areas of the body, and were attributed to activity as labor, sports, or sprains. No patient had given history of leg bone fracture, nor received any form of immobilization treatment before they were thoroughly examined clinically and by imaging, and admitted for surgery.

Table 1

Age group, gender, and side

Age group, years		Gender		Total
		Males	Females	
16 to < 17		2	1	3
17 to < 18		2	1	3
18 to < 19		2	0	2
19 to < 20		4	2	6
20 to < 21		4	2	6
Total		14	6	20
Side	right	8	4	12
	left	6	2	8
	bilateral	0	0	0

The preoperative clinical examination protocol involved the presence of lower leg swelling, eliciting of local tenderness over the lower leg by palpation, and examination of the ankle and foot motion. All had mild lower leg swelling, local tenderness, but painless satisfactory ankle/foot motion (Fig. 1).

Preoperative plain radiographic evaluation. The suspicion of the nonossifying fibromas was made on the basis of the radiographic findings. Jaffe (in 1942) stated that plain radiography is sufficient to make a diagnosis of nonossifying fibrommas [10]. A.M. Schwartz, R.M. Ramos [14] and M.H. Klein et al. [12] considered the plain radiographic diagnosis of nonossifying fibromas to be 100% accurate. Standard anteroposterior and lateral radiographs were made. All of the 20 patients showed a large (>30 mm in diameter, encroaching to >50% of the distal tibial medullary diameter), radiolucent, eccentric, ovoid, multiloculated lesion, with surrounding sclerosis, with its long axis parallel to the long axis of the bone, with thinned out cortices, within the distal tibial metaphysis in both views (Fig. 2). The lesion mean anteroposterior dimension was 65×70 mm and mean lateral dimension 60×70 mm (range, 60 mm to 100 mm). The volume of the lesion was calculated according to the formula: $(0.5 \times D \times d^2)$ and the mean volume was 80 cm^3 (range, 60 to 120 cm^3). The mean ratio of lesion/bone diameter was 60% (range, 50% to 70%).

Preoperative magnetic resonance imaging evaluation. The exact location, extent, and nature of the lesions within the distal tibia were confirmed by MRI imaging. All lesions showed low signal intensity in T_1 weighted due to abundant fibrous tissues, and high signal intensity in T_2 weighted due to abundant foamy histiocytes [11] (Fig. 3). There was no evidence of bone destruction, marrow oedema, or soft-tissue mass.

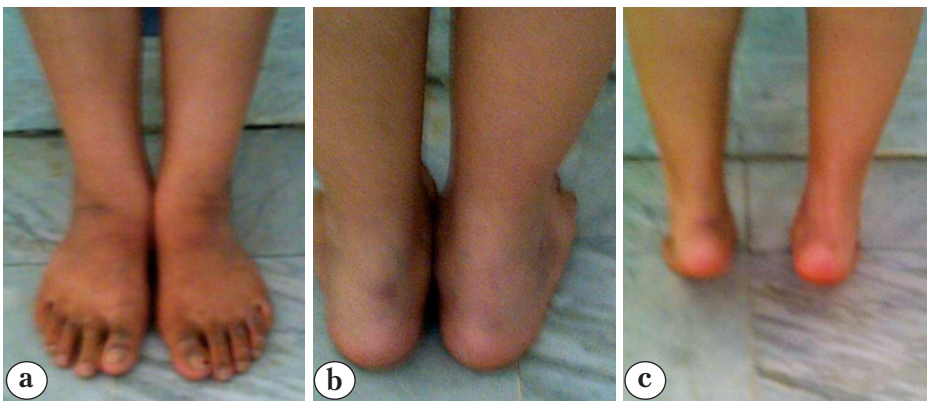

Fig. 1. Mild swelling in the medial *(a)*, and posterior *(b)* aspects of the right lower leg, but with full range of ankle plantar-flexion *(c)*

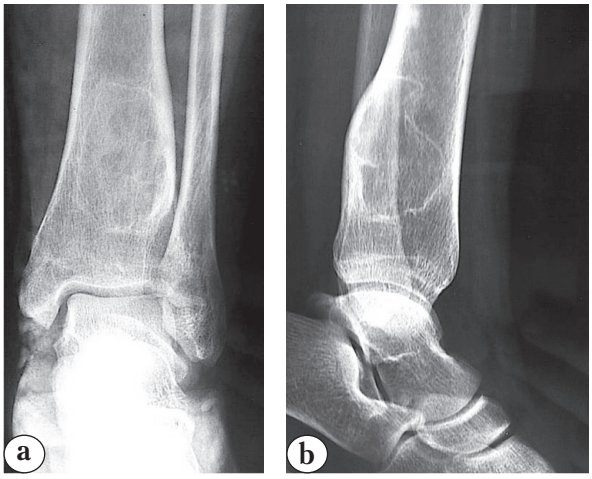

Fig. 2. Preoperative plain radiographs, anteroposterior *(a)* and lateral *(b)* views showing the huge intraosseous lesion, encroaching to >50% of the distal tibia, with surrounding sclerosis

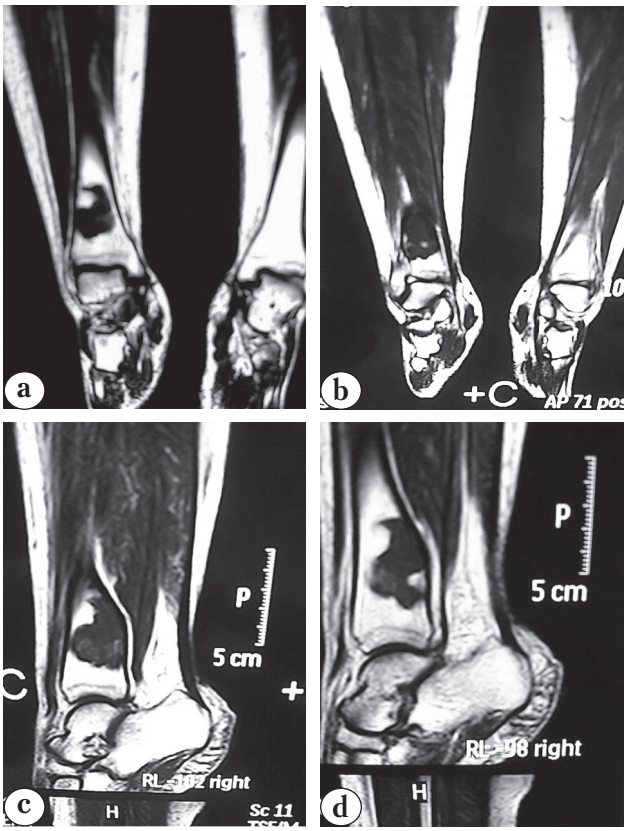

Fig. 3. Preoperative magnetic resonance imaging, T_1 coronal *(a)*, T_2 coronal *(b)*, T_1 sagittal *(c)*, and T_2 sagittal *(d)* showing the location, extent and nature of the lesion with low signal intensity in T_1 weighted and high signal intensity in T_2 weighted

Preoperative fine needle aspiration cytology: This non-invasive measure, though having false negative results, revealed a mixture of fibroblasts and histiocytes, in favour of a benign fibrous lesion.

Operative procedure: Owing to the triangular configuration of the distal tibia, having three borders (anterior or shin, medial, and lateral) and three surfaces (medial or subcutaneous, lateral, and posterior), the approach was through a longitudinal incision either on the anterior or the posterior aspect of the lower leg depending on the extent of the lesion. The anterior approach was used in lesions causing thinning out of the anterior, medial, or lateral cortex of the distal tibia, whereas preserving the posterior cortex. It was performed between the tibialis anterior tendon which was retracted medially, and the extensor hallucis longus which was retracted laterally protecting the anterior neurovascular bundle. The shin, medial and lateral surfaces can be exposed through it. The posterior approach was used in lesions extending posteriorly causing thinning out of the posterior cortex. It was performed between the flexor hallucis longus which was retracted medially protecting the posterior neurovascular bundle, and the peroneal tendons which were retracted laterally (Fig. 4). The thinned tibial cortex was exposed and a window was removed by a sharp osteotome connecting multiple circumferential drill holes in the surrounding healthy bone, as a safety margin, to inspect the interior of the lesion (Fig. 5).

The interior of the lesion was thoroughly curetted with a curette spoon; and was found to be yellowish brown material (Fig. 6) with fibrous consistency.

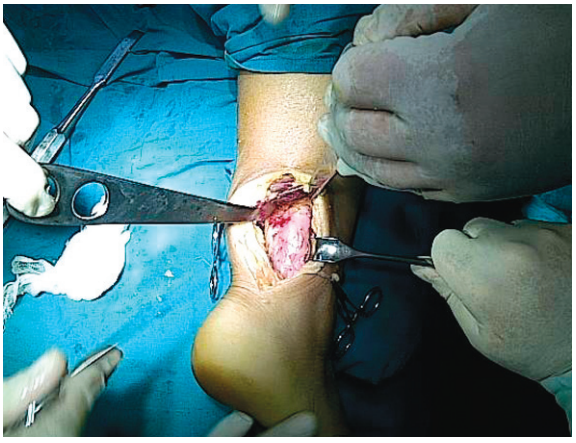

Fig. 4. Posterior exposure of the lesion between the flexor hallucis longus medially and the peroneal tendons laterally

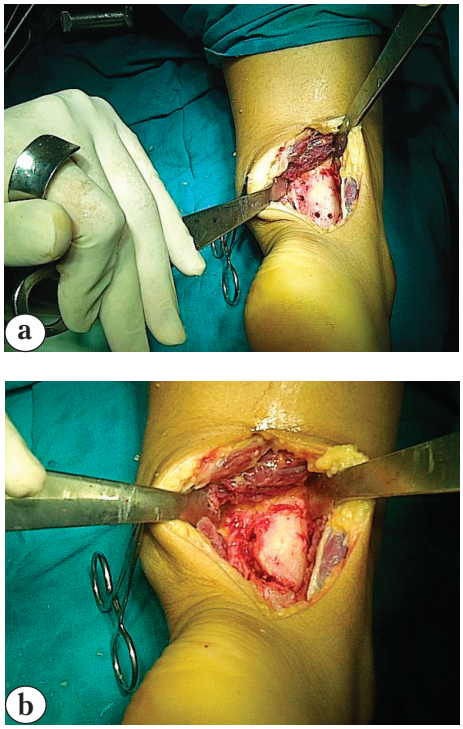

Fig. 5. Multiple circumferential drill holes in the surrounding healthy bone as a safety margin *(a)* to remove a window *(b)*

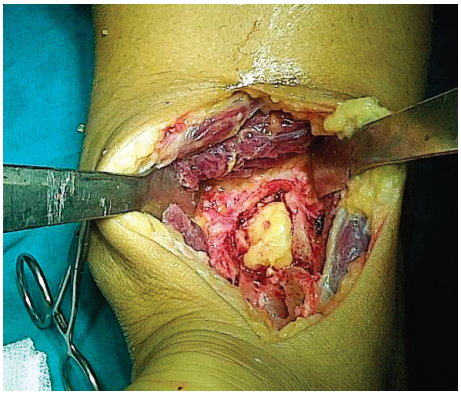

Fig. 6. The interior of the lesion after removal of the window

The curettage process continued till the other healthy cortices were clearly visualized. The cavity was filled with aqueous solution of 80% phenol with compression on the wall of the cavity, for a few minutes, and then rinsed in pure alcohol; and the procedure was repeated three times. No liquid nitrogen cryosurgery was used. Cement in the form of high molecular weight polymethylmethacrylate (acrylic) was submerged to fill the cavity by polymerization (Fig. 7). No bone graft was used. The wound was closed in layers. A sterile dressing was applied without immobilization.

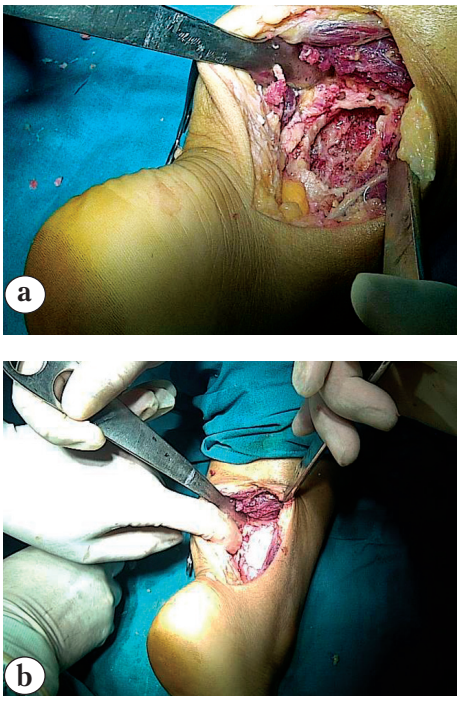

Fig. 7. The interior of the lesion before *(a)*, and after insertion of the cement *(b)*

Postoperative care. Active and passive movements through the ranges of motion of the ankle and toes started the day after the operation. The sutures were removed two weeks postoperatively; at which time partial weightbearing was allowed. Full weightbearing was gradually allowed after the next two weeks.

Postoperative evaluation: Sequential plain radiographs were made until clinical healing of the bone defect which usually took two to three months; then every 2 months for one year, every six months for 2 years, and yearly for 5 years to detect local recurrence. All histopathological reports concluded the presence of nonossifying fibroma showing abundant fibrous and areolar tissue, dense spindle-shaped fibroblast, histiocytes, but no atypia or pleomorphism (Fig. 8).

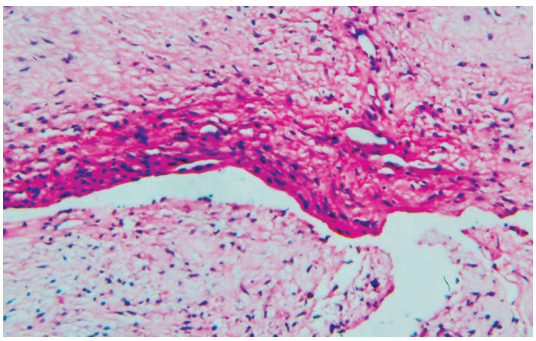

Fig. 8. Postoperative histopathology showing abundant fibrous and areolar tissue, dense spindle-shaped fibroblast, histiocytes, but no atypia or pleomorphism

Results

The twenty patients were assessed both clinically and radiographically in a mean follow-up period of 6 years and 2 months (range, 5 to 7 years) as regards to the presence or absence of pain, the functional activity, the occurrence of pathological fracture, the cement/bone interface zone, and the recurrence of the lesion.

Pain and functional activity were studied using the musculoskeletal tumour society (MSTS) score of the lower extremity which provided numerical values (0 to 5) assigned for six categories (pain, function, emotion, support, walking, and gait), giving a total score between 0 and 30, with 0 indicating poor and 30 excellent (Table 2) [6].

Pain. Pain disappeared completely in 16 out of the twenty patients who got a score of 5; was intermediate between modest non-disabling and absent in 2 patients (no. 3 & 15) who got a score of 4; and was modest non-disabling in 2 patients (no. 9 & 13) who got a score of 3. No patient had intermediate between moderate disabling and modest non-disabling pain (score 2); moderate disabling (score 1); and severe disabling (score 0).

Function. Function was not restricted in 16 out of the twenty patients who got a score of 5; was intermediate between recreational restriction and no restriction in 2 patients (no. 3 & 15) who got a score of 4; and was recreational restricted in 2 patients (no. 9 & 13) who got a score of 3. No patient had intermediate between partial restriction and recreational restriction (score 2); partial restriction (score 1); and total restriction (score 0).

Emotion. Emotion was enthused in 16 out of the twenty patients who got a score of 5; and was intermediate between satisfied and enthused in 4 patients (no. 3, 9, 13, & 15) who got a score of 4. No patient had satisfied (score 3); intermediate between accept and satisfied (score 2); accept (score 1); and dislike (score 0).

Support: There was no support in all of the 20 patients who got a score of 5.

Walking. Walking was unlimited in all of the 20 patients who got a score of 5.

Gait. Gait was normal with no limp in all of the 20 patients who got a score of 5.

The mean MSTS score was 29.2 (range, 25 to 30). Sixteen patients got a score of 30, two patients (no. 3 & 15) a score of 27, and two patients (no. 9 & 13) a score of 25 (Table 3).

There was no local recurrence, pathological fracture, or change in the cement-bone interface in all of the twenty patients as evidenced by the sequential radiographic imaging throughout the follow-up period (Fig. 9).

There was no correlation of the results with the size or extent of the lesion, or the gender of the patient.

Statistical analysis. The clinical and radiographic results before and after surgery were compared; and the differences were analyzed and defined as significant if the p value was < 0.05.

Intra-observer and inter-observer variability were studied. The 20 legs were examined and scored independently by four observers. On a separate occasion, two of the observers repeated the assessments of the same legs in the absence of information from the initial observations. The chance corrected and weighted kappa statistics for observer agreement, both for inter-observer and intra-observer variability demonstrated satisfactory repeatability of the MSTS scoring system. The overall intra-observer mean weighted kappa was $\chi w = +0.45$ (range SE $\chi = 0.015–0.055$) and the overall inter-observer mean weighted kappa was $\chi w = +0.34$ (range SE $\chi = 0.008–0.040$).

Table 2

Musculoskeletal tumour society (MSTS) score (lower extremity)

Score	Pain	Function	Emotion	Support	Walking	Gait
5	No pain	No restriction	Enthused	None	Unlimited	Normal
4	Intermediate	Intermediate	Intermediate	Intermediate	Intermediate	Intermediate
3	Modest/ non-disabling	Recreational restriction	Satisfied	Brace	Limited	Minor cosmetic
2	Intermediate	Intermediate	Intermediate	Intermediate	Intermediate	Intermediate
1	Moderate/ disabling	Partial restriction	Accept	One crutch	Inside only	Major cosmetic
0	Severe disabling	Total restriction	Dislike	Two crutches	Not independent	Major handicapped

Table 3

The final score of the patients

Case No	Pain	Function	Emotion	Support	Walking	Gait	Total score
1	No pain (5)	No restriction (5)	Enthused (5)	No support (5)	Unlimited (5)	Normal (5)	30
2	No pain (5)	No restriction (5)	Enthused (5)	No support (5)	Unlimited (5)	Normal (5)	30
3	Intermediate (4)	Intermediate restriction (4)	Intermediate (4)	No support (5)	Unlimited (5)	Normal (5)	27
4	No pain (5)	No restriction (5)	Enthused (5)	No support (5)	Unlimited (5)	Normal (5)	30
5	No pain (5)	No restriction (5)	Enthused (5)	No support (5)	Unlimited (5)	Normal (5)	30
6	No pain (5)	No restriction (5)	Enthused (5)	No support (5)	Unlimited (5)	Normal (5)	30
7	No pain (5)	No restriction (5)	Enthused (5)	No support (5)	Unlimited (5)	Normal (5)	30
8	No pain (5)	No restriction (5)	Enthused (5)	No support (5)	Unlimited (5)	Normal (5)	30
9	Non disabling (3)	Recreational restriction (3)	Intermediate (4)	No support (5)	Unlimited (5)	Normal (5)	25
10	No pain (5)	No restriction (5)	Enthused (5)	No support (5)	Unlimited (5)	Normal (5)	30
11	No pain (5)	No restriction (5)	Enthused (5)	No support (5)	Unlimited (5)	Normal (5)	30
12	No pain (5)	No restriction (5)	Enthused (5)	No support (5)	Unlimited (5)	Normal (5)	30
13	Non disabling (3)	Recreational restriction (3)	Intermediate (4)	No support (5)	Unlimited (5)	Normal (5)	25
14	No pain (5)	No restriction	Enthused (5)	No support (5)	Unlimited (5)	Normal (5)	30
15	Intermediate (4)	Intermediate restriction (4)	Intermediate (4)	No support (5)	Unlimited (5)	Normal (5)	27
16	No pain (5)	No restriction (5)	Enthused (5)	No support (5)	Unlimited (5)	Normal (5)	30
17	No pain (5)	No restriction (5)	Enthused (5)	No support (5)	Unlimited (5)	Normal (5)	30
18	No pain (5)	No restriction (5)	Enthused (5)	No support (5)	Unlimited (5)	Normal (5)	30
19	No pain (5)	No restriction (5)	Enthused (5)	No support (5)	Unlimited (5)	Normal (5)	30
20	No pain (5)	No restriction (5)	Enthused (5)	No support (5)	Unlimited (5)	Normal (5)	30

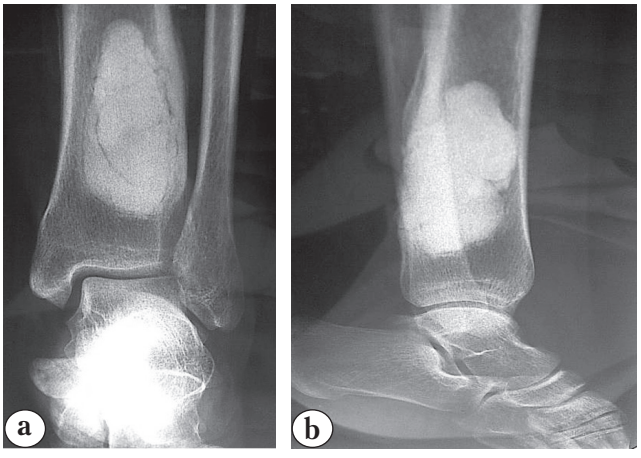

Fig. 9. Postoperative plain radiographs after 5 years, anteroposterior *(a)* and lateral *(b)* views showing intact cement-bone interface, no pathological fracture, and no local recurrence

Discussion

Fibrous cortical defects and nonossifying fibromas are the most common benign non-neoplastic bone lesions occurring in the metaphyses of long bones, most often in the distal femur, proximal and distal tibia, fibula, and humerus, in 30% to 40% of individuals between 8 to 20 years of age [5]. They are histologically identical but classified separately depending on their intraosseous involvement. Fibrous cortical defects are small, intracortical lesions that are asymptomatic and accidentally discovered in radiography for other reasons; whereas nonossifying fibromas are larger, actively growing lesions that involve the medullary cavity [1, 2]. Collectively, the two lesions are referred to as fibroxanthomas or histiocytic xanthogranulomas [9]. Huge or giant nonossifying fibroma is described as a lesion which is >30 mm in diameter or occupying >50% of the affected bone diameter [3]. Giant lesions are not asymptomatic; but usually present with pain and/or pathological fractures due to increased stress.

Imaging of giant nonossifying fibromas using plain radiography alone is possible due to the distinct characteristics of the lesion [10, 12, 14]. The possible differential diagnoses based on radiograph are unicameral bone cysts, aneurismal bone cysts, chondromyxoid fibromas, giant cell tumours, and

infections [15]. Magnetic resonance imaging defines the exact location, extent, and nature of the lesion. Fine needle aspiration cytology is a non-invasive diagnostic modality. Nonetheless, if the diagnosis is not clear, then wide surgical curettage acts as a biopsy and the definitive diagnosis can be made.

Curettage of the lesion and packing with autogenous bone graft were performed in most reported cases of nonossifying fibromas [4]. The technique of bone grafting involves operative harvest from a donor site having many complications, including painful scar, infection, hematoma, fracture, and gait disturbances [8] and does not guarantee local recurrence of the lesion.

J. Vidal et al. [16] was the first to describe curettage and cementation in treatment of benign bone lesions since then, the technique was adopted by many surgeons. It has the advantage of increasing the mechanical stability of the bone after curettage of the lesions, and minimizing the incidence of their local recurrence by the cytotoxic effect of the cement. However, there was no published literature describing the intralesional cementation in giant nonossifying fibromas.

M.M. Malawer et al. [13] used liquid nitrogen cryosurgery to minimize recurrence of giant cell tumour; and reported 2 to 8% recurrence. N. Fraquet et al. [7] used curettage, adjuvant chemical substance (aqueous solution of 80% phenol), and intralesional cementation to minimize recurrence of giant cell tumour; and reported 30% recurrence and arthrogenic problems due to the proximity of the cement to the articular cartilage.

In the current study, the work was concentrated on nonossifying fibromas only in the distal tibias of adolescents using curettage, adjuvant chemical substance (phenol), and intralesional cementation without autogenous bone graft, and without osteosynthesis. There was much improvement of pain and functional activity as studied by MSTS score (mean 29.2), no local recurrence (i.e. 0%), no widening of the cement-bone interface, and no pathological fracture.

In conclusion, giant nonossifying fibromas can be treated simply and effectively by curettage and intralesional cementation with excellent functional results. There was no arthrogenic effect of the cement being not in close proximity to the articular cartilage; even when used for filling huge cavities. No patients treated with curettage and cementation suffered from secondary joint problems. The mechanical effect of the cement improved the bone stability against pathological fractures; and its cytotoxic properties prevents local recurrence.

Conflict of interest: none.

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ЛЕЧЕНИЕ ГИГАНТСКОЙ НЕОССИФИЦИРУЮЩЕЙСЯ ФИБРОМЫ ДИСТАЛЬНОГО ОТДЕЛА БОЛЬШЕБЕРЦОВОЙ КОСТИ ПУТЕМ КЮРЕТАЖА С ПОСЛЕДУЮЩИМ ЗАПОЛНЕНИЕМ ДЕФЕКТА КОСТНЫМ ЦЕМЕНТОМ

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Реферат

Введение. Фиброзные кортикальные дефекты и неоссифицирующиеся фибромы (НФ) являются наиболее распространенными доброкачественными поражениями костей неопухолевого генеза, локализующимися в метафизах длинных костей у детей и подростков. Гигантские НФ сопровождаются болевым синдромом и/или патологическими переломами вследствие повышенного стресса.

Материал и методы. 20 пациентов подросткового возраста (14 мальчиков и 6 девочек), средний возраст которых составил 18,5 лет (от 16 до <21 лет), поступили на лечение в Национальный институт нейромоторной системы (Египет) в период с сентября 2007 по сентябрь 2009 года с диагнозом: гигантская НФ дистального отдела голени. Диагноз был поставлен на основе результатов клинического осмотра, рентгенографического исследования, магнитно-резонансной томографии и гистологического исследования. Лечение состояло в кюретаже с заполнением очага костном цементом без использования костных трансплантатов. Выраженность болевого синдрома и степень функциональной активности по шкале MSTS, наличие патологических переломов и местных рецидивов оценивались в средние сроки наблюдения 6 лет и 2 месяца (от 5 до 7 лет).

Результаты. Показатели боли и функциональной активности улучшились у всех пациентов и составили в среднем 29,2 балла по шкале MSTS (от 25 до 30). Ни у одного из пациентов не было патологических переломов и местных рецидивов, не наблюдалось никаких изменений на границе цемента с костной тканью, а также проблем артрогенного характера за весь период наблюдения. Статистически значимыми считали различия при значении $p < 0,05$.

Заключение. Лечение пациентов с гигантскими неоссифицирующимися фибромами путем кюретажа и внутриочагового цементирования является простым, эффективным и дает отличные функциональные результаты.

Ключевые слова: гигантская неоссифицирующаяся фиброма, хирургическое лечение, кюретаж.

Conflict of interest: none.

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