



Original Article

Can ethanol be used as an adjuvant to extended curettage in order to reduce the recurrence rate of aneurysmal bone cyst?☆

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ABSTRACT

Objective: The best treatment of aneurysmal bone cyst (ABC) is still unclear. This study aimed to evaluate the usefulness of extended curettage and ethanol as an adjuvant to reduce local recurrence of ABCs.

Methods: Retrospectively, 68 cases treated for primary and secondary ABCs caused by benign tumors from 2003 to 2013 were enrolled to a follow-up visit between one to ten years after the surgery. The treatment protocol was en-bloc resection, biopsy and curettage, extended curettage consisted of curettage, high-speed burring, ethanol 96%, and electrocauterization (combined four-step alcohol-using approach) followed by defect filling, consecutively.

Results: Among 36 patients with primary ABCs (16 male, 20 female, mean age of 16 years, range 3–46 years), 29 cases were treated with the combined four-step alcohol-using approach, four patients with resection, and three with biopsy and curettage. Thirty-two cases had secondary ABCs on benign lesions (17 male, 15 female). The recurrence rate was 5.88 in all primary and secondary ABC cases; two recurrences among 29 patients with primary ABCs (6.9%) and one recurrence among the 22 cases with secondary ABCs (4.5%).

Conclusions: It could be suggested that the combined four-step alcohol-using approach may result in a very low recurrence rate of primary and secondary ABC lesions.

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O etanol pode ser usado como adjuvante na curetagem ampla a fim de reduzir a taxa de reincidência de cisto ósseo aneurismático?

RESUMO

Objetivo: Ainda não se sabe qual o melhor tratamento para cistos ósseos aneurismáticos (COA). Este estudo teve como objetivo avaliar a utilidade da curetagem estendida e do etanol como adjuvante para reduzir a reincidência local de COAs.

Palavras-chave:

Cisto ósseo aneurismático

Etanol

☆ Study was carried out in Bone and Joint Diseases Research Center, Shiraz University of Medical Sciences, Shiraz, Iran.

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Álcool
Neoplasmas

Métodos: Retrospectivamente, 68 casos que receberam tratamento para COAs primários e secundários causados por tumores benignos entre 2003 e 2013 foram chamados para uma consulta de seguimento, em um intervalo entre um e dez anos após a cirurgia. O protocolo de tratamento foi ressecção em bloco, biópsia e curetagem; a curetagem estendida consistiu em curetagem, broqueamento em alta velocidade, etanol 96% e eletrocauterização (abordagem combinada em quatro etapas usando álcool), seguida do preenchimento do defeito, de forma consecutiva.

Resultados: Entre os 36 pacientes com COAs primárias (16 do sexo masculino, 20 do sexo feminino, idade média de 16 anos, intervalo 3-46 anos), 29 casos foram tratados com a abordagem combinada em quatro etapas usando álcool, quatro pacientes com ressecção e três com biópsia e curetagem. Trinta e dois casos apresentavam COAs secundárias em lesões benignas (17 do sexo masculino, 15 do sexo feminino). A taxa de reincidência foi de 5,88 em todos os casos de COAs primárias e secundárias; duas reincidências foram observadas entre 29 pacientes com COAs primária (6,9%) e uma reincidência entre os 22 casos (4,5%) de COAs secundária.

Conclusão: Sugere-se que a abordagem combinada em quatro etapas usando álcool pode resultar em uma taxa de reincidência muito baixa em lesões COAs primárias e secundárias.

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Introduction

Aneurysmal bone cyst (ABC), a rare benign and locally aggressive bone lesion, is a blood-filled cavity within an expanded region of the bone with thinning of the surrounding cortex.¹ It was first described by Jaffe and Lichtenstein² in 1942. It may present as a primary bone cyst or a secondary lesion arising from other osseous conditions like giant cell tumor, chondroblastoma, fibrous dysplasia, osteoblastoma, non-ossifying fibroma, telangiectatic osteosarcoma.^{3,4} Although precise pathogenesis of ABC remains unclear, multiple theories have been proposed. They have been described that specific translocational events on chromosome 16 and 17 as the main etiology of primary ABCs,^{5,6} and intraosseous or subperiosteal hemorrhage because of abnormal venous circulation as a cause of secondary ABCs.⁷ ABC tumors are frequently seen in the first two decades of life with slight female predominance. It may occur in all bones, but metaphysis of the long bones and dorsal elements of the vertebrae are the most common sites.^{8,9}

Treatment of ABC lesions in the long bones commonly is extended curettage with bone grafting or wide en-bloc resection of tumor.^{3,8,10} Some authors have used adjuvants inducing hydrogen peroxide,⁸ phenol,¹¹ polymethylmethacrylate bone cement,¹² liquid nitrogen,¹³ argon beam,¹⁴ and high-speed burring¹⁵ in order to decrease the recurrence rate of ABC lesions.¹ The main purpose of this study is to evaluate the usefulness of ethanol as an adjuvant in a combined four-step procedure including curettage, high-speed burring, ethanol 96% and electrocauterization followed by grafting to reduce local recurrence of primary and secondary ABCs developed on benign tumors.

Methods and materials

After approval of the study by the ethic committee of our university, a retrospective review of medical clinical records

was performed on cases treated for ABC from 2003 to 2013 by the senior author at the main orthopedic center of south of Iran. After exclusion of cases with secondary ABCs arising from malignant tumors, 68 patients, with range of follow-up visit of 1 to 10 years, signed the prepared written consent form. They consisted of 36 cases with primary ABCs and 32 patients with secondary ABCs on benign tumors. Demographic data (age at the time of the surgery, gender), primary symptom of the patient, exact location of ABC lesion, pathologic reports, and further surgeries were reviewed according to the medical records and available images. The most recent taken X-ray radiographs were considered to evaluate curing or recurrence.

Suspicious lesions were treated with biopsy and curettage to determine the exact pathology. En-bloc resection was carried out in ABC lesions in expandable parts of the bones like proximal fibula. Our approach, called the combined four-step alcohol-using approach, consisted of extended curettage, high-speed burring, ethanol 96%, and electrocauterization of the lesion, consecutively. Extended curettage was performed to remove all abnormal tissues. After using high-speed burr on the walls of the lesion, the defect was irrigated by normal saline. In the third step, ethanol 96% was carefully poured in the lesion with syringe to fill it completely. Any possible spill of ethanol in the surrounding tissues was suctioned immediately. After one minute, ethanol was evacuated by suction tube followed by irrigation with normal saline. This cycle was repeated three times. Following irrigation to eliminate the risk of explosion, electrocauterization of the lesion with monopolar coagulation diathermy set to 50 W was performed on the whole wall of the lesion. The protocol of combined four-step alcohol-using approach was carried out in lesions of any bone of the body irrespective of size or location except in the spinal region. Finally, the void would be filled by autograft, allograft, or bone cement. We routinely used autograft from iliac crest to fill the defect but allograft should be used for larger defects in the children. Also, bone cement was used to fill the

Table 1 – Distribution of ABC lesions by anatomic site.

	Femur	Tibia	Humerus	Radius	Foot	Fibula	Hand	Ulna	Clavicle	Pelvis	Patella
Primary ABC	11	6	5	3	3	2	2	1	1	1	1
Secondly ABC	13	9	5	–	1	1	1	1	1	1	–

Table 2 – Treatment method and recurrence for ABC lesions.

Treatment	Primary ABC		Secondary ABC	
	Number of subjects	Number of recurrences	Number of subjects	Number of recurrences
Biopsy & Curettage	3	0	1	0
Extended Curettage + Autograft	18	1	7	1
Extended Curettage + Allograft	10	1	6	0
Extended Curettage + Cement	1	0	9	0
Resection + Autograft	1	0	7	0
Resection + Allograft	3	0	2	0

void of periarticular lesions and secondary ABCs on giant cell tumors. Treating of pathological fractures was a little different; after extended curettage, the fracture was reduced and fixed. After covering the periphery of the fracture by multiple gauzes and using suction tube to reduce the risk of ethanol leakage, ethanol was spilled in the lesion. After electrocauterization, bone graft was impacted in the defect.

After any recurrence found on X-ray, other modalities such as CT scan or magnetic resonance imaging was requested, based on the characteristics of the lesion. If it was big with possibility of fracture in the future, surgery was done according to the combined four-step alcohol-using approach.

Results

Primary ABC

Thirty six patients with histologically confirmed primary ABC were treated. There were 16 male (44%) and 20 female (56%) with a mean age of 16 years (range 3–46 years) at the time of surgery. Half of the subjects were between 10 and 20 years of age (19% of cases <10 years of age, 31% of cases >20 years of age). Twenty one cases (58%) had lesion on the right-sided limbs of the body and 15 had ABCs of left limbs (42%). [Table 1](#) shows distribution of ABC lesions by the anatomic site. On presentation, 12 subjects referred with pathologic fracture, 10 cases came with mass and 11 had pain. ABC lesions were found in three cases incidentally. Treatment method and recurrence rate are described in [Table 2](#).

Totally, we had two recurrent ABCs in the series treated with combined four-step alcohol-using approach (6.9%). The first case was a six-year-old boy with ABC of distal of right radius treated with extended curettage and allograft insertion. He developed local recurrence of lesion one year after surgery. The second subject was a three-year-old girl underwent extended curettage and autograft for ABC of distal radius of left upper limb.

Secondary ABC

Thirty two cases had secondary ABCs on benign lesions (male: 17, female: 15). The mean age of the patients was 29 years,

with a range of 7–51 years. Two subjects (6%) had age of less than 10 years, 12 patients (38%) were between 10 and 20 years of age, and 18 cases (56%) were older than 20 years of age. The anatomical locations for the secondary ABC lesions are listed in [Table 1](#). The complaints of the patients at the time of initial visit were pain in 23 cases, mass in 6 subjects, pathologic fracture in 2 patients, and incidental finding in one. Primary lesions were giant cell tumor in 13 cases (40%), fibrous dysplasia in 7 subjects (22%), chondroblastoma in 6 (19%), non-ossifying fibroma in 5 (16%) and histiocytoma in one patient (3%). Nine out of 13 cases with giant cell tumor were cured with extended curettage and cement without recurrence, as shown in [Table 2](#). Only one case among 13 treated with combined four-step alcohol-using approach had recurrence of the lesion. This patient was a case of secondary ABC on non-ossifying fibroma. Frozen-section biopsy during the second surgery approved recurrence of ABC on healed non-ossifying fibroma; so combined four-step alcohol-using approach was carried out.

Discussion

The optimal method of treatment for ABC is still unclear. Although different material injections and percutaneous sclerotherapy have been introduced as an efficient treatment,^{16–18} extended curettage with or without bone grafting is the most classically accepted mode of treatment in the long bones. Wide en-bloc resection is an excellent option to prevent any recurrence but complex resection of tumor must be limited to expandable bones like distal ulna or proximal fibula. Our results showed 100% local control of ABC lesions after complete resection of tumors followed by grafting. Using extensive surgery of en-bloc resection associated with neurovascular damage, considerable morbidity, and complete reconstructive surgeries are not practical for many locations in the skeletal system of human being.

Other than wide resection, curettage and bone graft showed failure rates of less than 30%.^{1,3,8,10,19,20} Using different adjuvants in order to reduce the recurrence rate have been associated with controversial results. Keçeci et al.²¹ did not report any significant difference in terms of local recurrence between patients treated with curettage and bone grafting

without adjuvants and patients treated with curettage and bone grafting beside burring or phenol usage. On the other hand, using curettage, burring, and bone grafting resulted in lower recurrence rates.^{15,22} Also adding electrocautery and phenol in addition to burring have been suggested by the others.²³ Shifting from phenol to ethanol appears rational due to its corrosive effects, chemical burns, neurovascular damages, mucosal damages of the respiratory system, paralysis of peripheral nerve endings, and even systemic poisoning.²⁴

Nowadays intracystic injection of alcohol (polidocanol) showed good results with a low rate of complications.²² Even it is a technique of choice in some centers.¹ Ethanol is a harmless and easily-available material in most operation rooms. The efficacies of ethanol to treat giant cell tumor,²⁵ osteoid osteoma,²⁶ skeletal metastasis,²⁷ and other bone lesions²⁸ have been described in the literature. The efficacy of ethanol has not been described yet to reduce recurrence rate of ABC lesions. We could suggest using alcohol as an adjuvant to treat primary and secondary ABC lesions with recurrence rate of about 6%. We had just one recurrence among 22 cases (4.5%) with secondary ABC lesions and two among 29 patients with primary ABC (6.9%).

We acknowledge the limited number of our patients and minimum follow-up duration. But it is obvious that a minimum of two years follow-up is required to diagnose most of recurrences of ABC lesions, because most of them occur within 12–18 months.²⁹ In the presented study, all patients treated with four-step alcohol-using approach were evaluated 2 years post-operatively. So, our results could be reliable. Just two patients with en-bloc resection were assessed one year after surgery.

Conclusion

It could be suggested that four-step alcohol-using approach can reduce recurrence rate in patients with primary and secondary ABC lesions. Moreover, the recurrence rate is lower than others reported after using other adjuvants in the literature.

Conflicts of interest

The authors declare no conflicts of interest.

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