Acta Orthopaedica et Traumatologica Turcica 52 (2018) 157–161

Contents lists available at ScienceDirect

Acta Orthopaedica et Traumatologica Turcica

journal homepage: https://www.elsevier.com/locate/aott

Epithelioid hemangioma of the scapula treated with chemoembolization and microwave ablation: A case report

Konstantinos Tsikopoulos [a, *], Evangelos Perdikakis [b], Dimitrios Georgiannos [a], Ilias Bisbinas [a]

[a] 1st Orthopaedic Department, 424 Army General Training Hospital, Thessaloniki, Greece
[b] Department of Interventional Radiology, 424 Army General Training Hospital, Thessaloniki, Greece

ARTICLE INFO

Article history:
Received 4 July 2016
Received in revised form
9 October 2016
Accepted 2 January 2017
Available online 31 January 2017

Keywords:
Epithelioid hemangioma
Chemoembolization
Microwave ablation
Scapula

ABSTRACT

Bone epithelioid hemangiomas are classified within benign vascular tumours but are commonly misdiagnosed as low-grade angiosarcomas or epithelioid hemangioendotheliomas. Current therapeutic interventions include various treatment options but local recurrence or distal lymph node involvement has been reported. We report a rare case of scapular epithelioid hemangioma that was initially treated using a combination of chemoembolization and microwave ablation. This combination has not been previously reported in the literature regarding the management of this tumour. A year after the first course of treatment, the tumour size has been reduced more than 70% and the patient has remarkable clinical improvement. Results reported in this case study demonstrated that combination of chemoembolization and microwave ablation is a feasible, safe and effective technique in the treatment of bone epithelioid hemangiomas. Even if the tumour is still present afterwards, a substantially smaller surgical excision will be needed.

Introduction

Bone Epithelioid Hemangiomas (EH) are classified within benign vascular tumours.[1] However, they are commonly misdiagnosed as low-grade angiosarcomas or malignant hemangioendothelioma.[1,2] The histopathology of EH reveals the presence of stromal inflammatory cells and endothelial cell proliferation.[1–4] Currently, various therapeutic interventions for the management of EH such as marginal or wide surgical excision, radiotherapy and embolization have been reported.[5] Local recurrence of HE or lymph node involvement is a rare complication after incomplete resection of the tumour, despite the fact that long term follow-up of patients after surgical resection of epithelioid hemangiomas has invariably failed to substantiate the existence of any malignant behaviour.[1–5]

We report a rare case of bone EH occurring in the scapula, which was treated with a combination of chemoembolization and microwave ablation. To the best of our knowledge, this is the first reported case of a EH in the scapula that was treated with the combined use of minimally invasive loco-regional interventional radiologic means.

Case report

The subject of this report is a white Caucasian 56-year-old man, who complained about deteriorating pain in his right dominant shoulder without history of trauma. His past medical history included coronary heart disease-aortic valve insufficiency and he had bypass and valve replacement surgery. He presented to the orthopaedic outpatient clinic of our hospital because of deteriorating pain during the last four months that did not settle after two corticosteroid injections in the shoulder performed by his General Practitioner. On clinical examination, there was increased tenderness on palpation and the patient kept holding his arm still avoiding any movement to reduce the pain in his affected shoulder. Laboratory examinations were unremarkable. Radiographs showed an osteolytic lesion in the patient's right scapula (Fig. 1a). Subsequent Computed Tomography (CT) showed a large tumour and a Tc^{99} bone scan demonstrated intense radiotracer uptake (Fig. 1b

* Corresponding author. Ring Road, N. Efkarpia, Thessaloniki, Greece.
E-mail addresses: kostastsikop@gmail.com, kostastsikop@yahoo.gr (K. Tsikopoulos).

Peer review under responsibility of Turkish Association of Orthopaedics and Traumatology.

http://dx.doi.org/10.1016/j.aott.2017.01.003

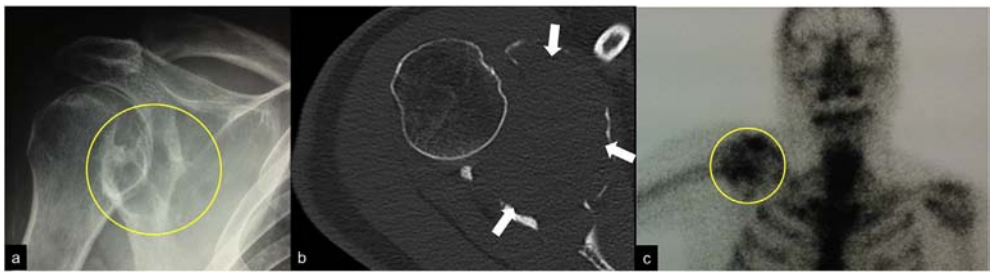

Fig. 1. (a) The plain radiograph shows a lytic lesion (area in circle) of the right scapula. (b) The axial multidetector computed tomography image verifies the lytic scapular lesion (arrows), and the scintigraphic study demonstrates intense tracer uptake (area in circle).

and c). Magnetic Resonance Imaging (MRI) scans confirmed the presence of a highly vascular tumour occupying most of the scapula, breaching the scapular cortices and infiltrating the surrounding soft tissues (Fig. 2). A tru-cut biopsy was performed using anterior approach and four biopsy cylinders of both soft tissue and bone were obtained from various sites of the lesion through the same skin portal.

Histopathologically, the lesion included vascular formations covered with endothelial cells and no atypia was present (Fig. 3a–d). Taking into account all the histopathological and immunohistochemical features, the patient was diagnosed with EH of the right scapula without any signs of malignancy.

Following discussion in the multidisciplinary meeting of the oncological team in the hospital, the patient was given the following options: a) a wide surgical excision, with probable extensive upper limb functional impairment because of location and size, and b) a minimally invasive treatment using a combination of chemoembolization and microwave ablation. The second option, which he finally chose, could be applied as either a single stage treatment or as an adjuvant treatment. The therapeutic interventions were performed in two cycles. A digital subtraction angiography (DSA) was performed during the first cycle. This angiography verified the highly vascular component of the tumour. Then, super-selective chemoembolization (mixture of 10 ml Lipiodol & 10 mg Bleomycin & 2 ml embolic microspheres 300 μm) of the tumour feeder vessels was carried out with the use of a microcatheter (Fig. 4a–d). The patient reported immediate pain relief and the second cycle of the treatment was scheduled 4 weeks later, after post embolization syndrome has subsided. The second treatment cycle consisted of super-selective embolization followed by a CT-guided microwave ablation. Ablation was performed with a 16G, 15 cm long microwave electrode needle. In order to avoid tumoral seeding during percutaneous treatment, meticulous measures such as coaxial catheter insertion and percutaneous tract ablation were applied. In total, 18 min of ablation were carried out (60 Watts/2450 MHz) in three different parts of the tumour. The aim was to cover the whole volume of the lesion (Fig. 5a and b). Nonsteroidal anti-inflammatory drugs (NSAIDS) were administered after each treatment for the management of post-intervention pain. The patient was followed-up every six months for a period of two years with clinical assessment, laboratory examinations, and MRI scans. Clinical outcomes and adverse effects were also recorded

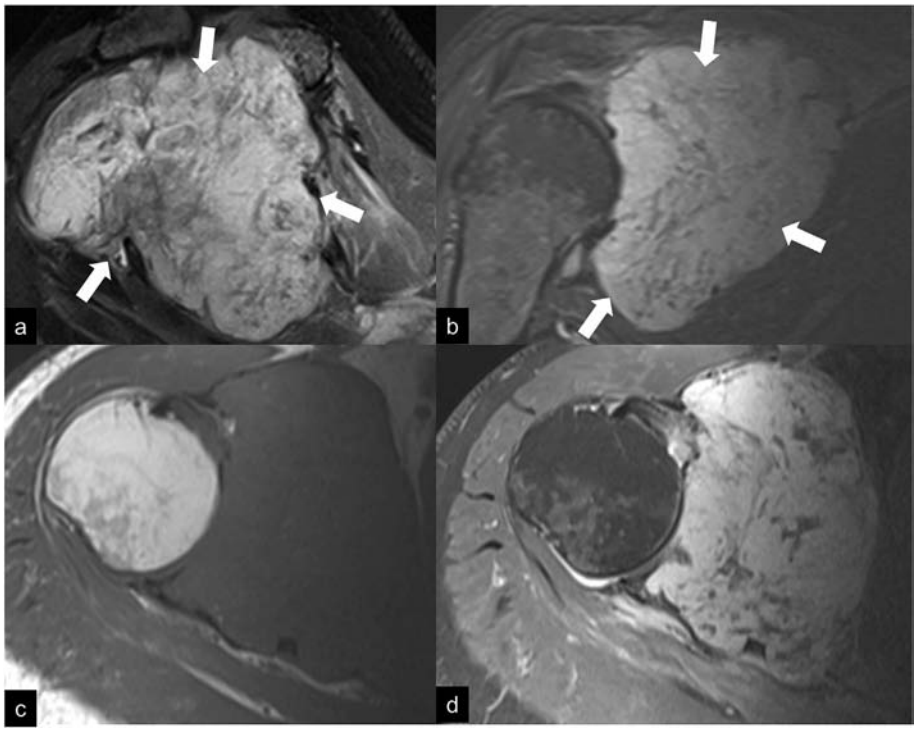

Fig. 2. The sagittal (a) and the coronal (b) T2 fat saturated MR images show the soft tissue mass infiltrating the right scapula (arrows). The axial (c) T1w and the axial (d) contrast enhanced fat saturated T1w MR images demonstrate intense enhancement of the tumour.

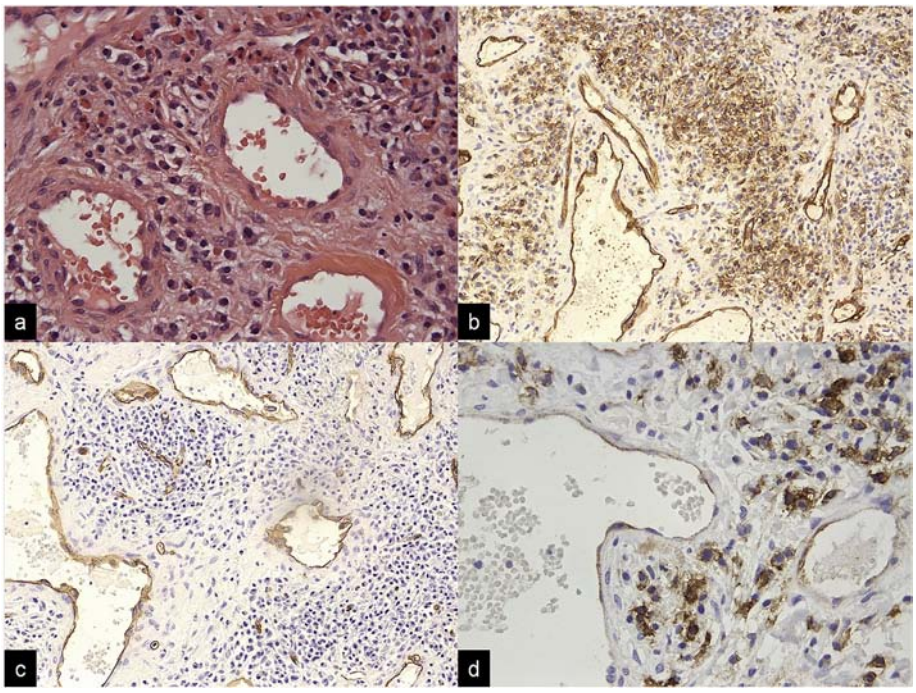

Fig. 3. Histopathologic images of epithelioid hemangioma. (a) Vascular channels with slightly prominent endothelial lining and inflammatory cells in the surrounding stroma (hematoxylin-eosin stain ×40 magnification view). (b) Immunohistochemistry stain CD31 (×20 magnification view). (c) Immunohistochemistry stain CD34 (×20 magnification view) (d) Immunohistochemistry stain epithelial membrane antigen (×40 magnification view).

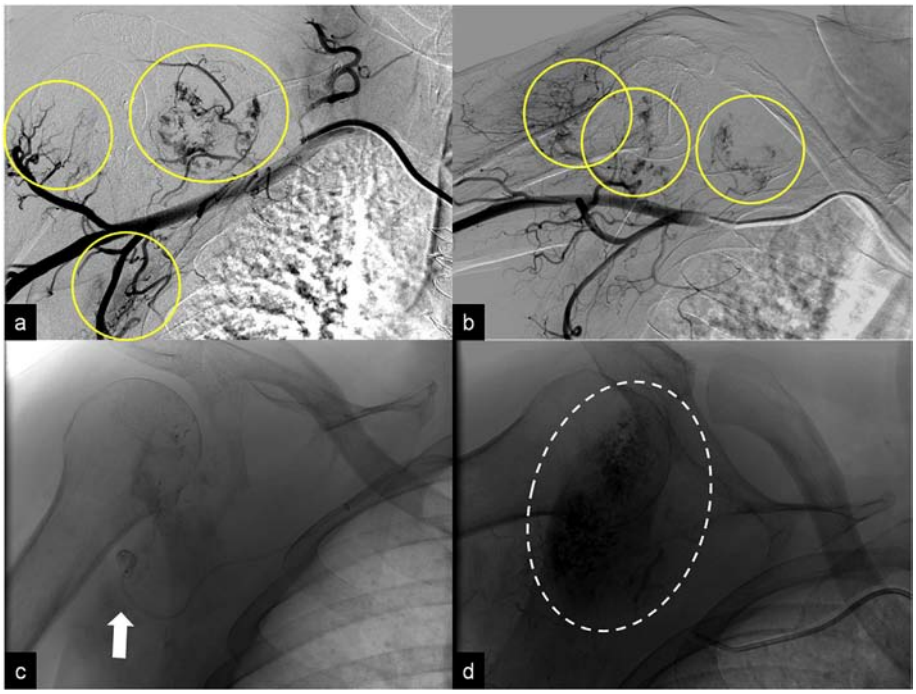

Fig. 4. The digital subtraction angiography images (a-b) show the abnormal vascular staining of the epithelioid hemangioma (areas in circle). (c) The Digital Subtraction angiographic image during selective embolization with a microcatheter (arrow). (d) Final result after chemoembolization shows satisfactory staining (area in dashed circle).

(Table 1). The clinical outcomes were assessed using: a) the Visual Analogue Scale (VAS) to measure pain intensity levels at rest, b) the shoulder pain and disability index (SPADI) measuring pain intensity and functional disability, and c) the active range of motion (AROM) of the affected shoulder.

Regarding the MRI scan assessment, the tumour size was reduced by more than 70% of its pre-treatment size at one year follow-up and the patient was remarkably satisfied from the clinical outcome. The adverse effects included a local skin rash and partial avascular necrosis (AVN) of the right humeral head. The skin

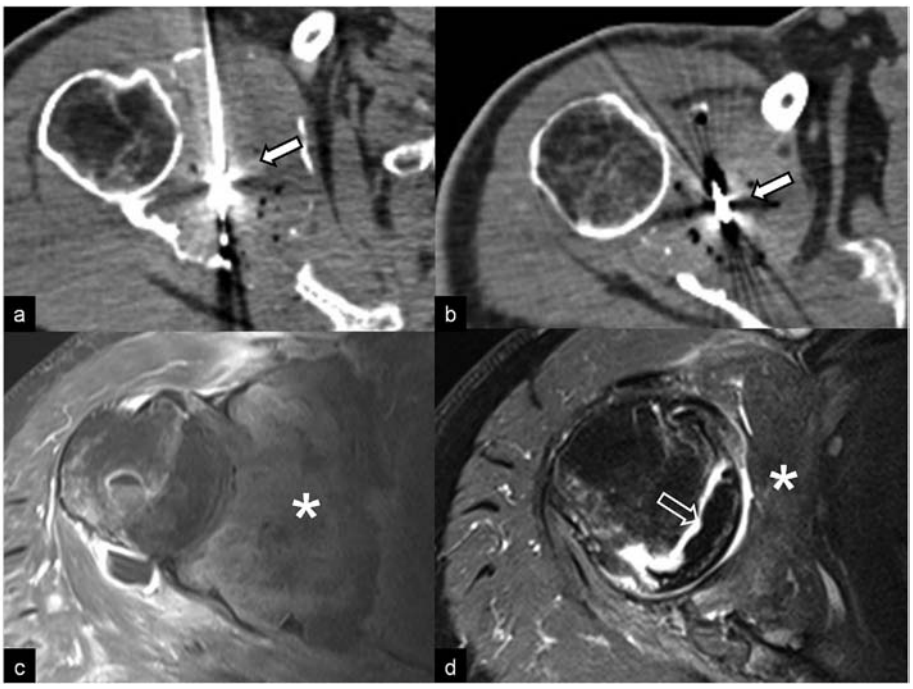

Fig. 5. (a-b) The axial multidetector computed tomography images show the needle (arrows) during ablation. (c) The axial T1W fat saturated contrast enhanced MR image (study 24 h after treatment) shows complete necrosis of the tumour (asterisk). (d) The axial T2W fat saturated magnetic resonance image (study one year after treatment) shows necrosis and shrinkage of the tumour (asterisk). Please note osteonecrosis of the humeral head (open arrow).

Table 1
Outcome measurements. VAS = Visual Analogue Scale; SPADI = Shoulder Pain and Disability Index; ROM = Range Of Motion; IR = Internal Rotation; ER = External Rotation.

Outcome measurement		Pre-treatment	After first intervention	After second intervention	At one year
VAS at rest (mm)		8.3	2.5	0	0
SPADI		117	56	30	28
Active ROM	Flexion, deg	10	45	80	100
	Extension, deg	0	20	50	50
	Abduction, deg	10	45	80	90
	IR, deg	10	40	90	90
	ER, deg	0	10	30	30

rash appeared early after the first intervention and was successfully treated with local corticosteroids ointments. On the other hand, the AVN was observed after the second cycle of treatment and was attributed to local ischemia, since the affected portion of the humeral head was included in the ablative zone (Fig. 5c and d). It is noted that the symptoms of AVN were completely covered by the tumour symptomatology, management, and reduced use of the patient's shoulder.

Discussion

Our study is the first reported case of scapular EH that was treated by means of minimally invasive treatment (i.e., a combination of chemoembolization and microwave ablation). Current literature suggests that intraosseous EHs most commonly occur in long tubular bones.[5–8] Nielsen et al in their study showed that bone epithelioid hemangiomas arose 40% in long tubular bones, 18% in short tubular bones of the distal lower extremity, 16% in vertebrae, and 8% in small bones of the hands.[7] In their study, nine patients had involvement of more than one bone. To our knowledge, the rarity of the tumour location in addition with the novel treatment applied and the complications encountered has not been previously reported in the literature for the management of EH.

Traditionally, surgical resection, image-guided percutaneous sclerotherapy and transarterial embolization either alone or in combination have been the used as the standard treatments for symptomatic peripheral soft-tissue vascular tumours depending on flow characteristics.[9–11] Recently, in a study by SM Thompson et al, ablation techniques have also been incorporated in the armamentarium of percutaneous vascular tumour management.[11] Lately, Bianchi et al have also presented the results of electrochemotherapy in the treatment of bone metastases that has similarities to our technique. They have achieved control of pain and disease progression in the majority of the patients with consequent improvement of quality of life.[12] Similarly, in our case a combined therapeutic approach with intra-arterial chemoembolization and microwave ablation was performed with successful outcomes.

The purpose of including microwave ablation to the treatment protocol of this tumour was to enhance the therapeutic efficacy of chemoembolization. Bleomycin is an effective chemotherapeutic sclerosant and has been widely used to treat vascular malformations.[13,14] The underlying mechanism of bleomycin-induced sclerotherapy was attributed to the obliteration of enlarged channels induced by endothelium damage as a result of acute or chronic inflammation and fibrosis.[13,14] However, recent evidence shows that treatment with bleomycin affects also the adhesion molecules

of the endothelium and destroys intercellular interactions.[13,14] In our case super-selective embolization was performed delivering the sclerosant trans-arterially, using micro-catheter technique. This allowed maximal dose administration to the tumour with minimal dilution and maximal protection against non-target embolization.

The combined therapeutic approach with chemoembolization and microwave ablation was also preferred in our case to maximise the treatment outcome specifically in a bone-infiltrating tumour. Many endovascular occlusive agents (embolic agents) are currently in use to treat the aforementioned vascular tumours. Although their efficacy is well documented, re-canalisation does occur.[9–14] In addition, curative ablation has been extensively applied for the treatment of specific benign or in selected cases of malignant localized bone tumours with excellent results.[9–15] We postulated that combining these two methods, we could achieve a better outcome for our patient. Indeed, pain palliation therapy and local control of the tumour was achieved with the combined use of these safe, fast, effective, and tolerable percutaneous methods.

It is also noted that the functional impairment, which would have been induced after an extensive surgical excision to the patient's predominant shoulder was avoided. Instead, by choosing minimally invasive techniques, significant clinical improvement was recorded in terms of all case-study outcomes at one-year follow-up, and no evidence of recurrence was detected. At present, the patient is substantially satisfied from the treatment he received.

Regarding adverse effects, a local skin rash presented after the first intervention that was successfully treated with local corticosteroids ointments. It is well-documented in the literature that embolization and chemoembolization of peripheral vascular tumours may result in skin complications that are managed conservatively.[9,11–14] The AVN of the humeral head was observed after the second cycle of treatment and was a rather expected complication. It was attributed to local ischemia from embolization and ablation, since normal vessels were sacrificed, even with a minimally invasive method, in order to achieve local control of an extensive tumour.[16] Taking into account the reduction in patient's daily activities and the extent of the tumour into neighbouring structures, the AVN of the humeral head was completely asymptomatic.

Summarizing, a rare case of EH occupying most of the patient's scapula is presented. Taking into account the location and size of the tumour, an extensive, debilitating surgery was needed to achieve complete surgical excision. Instead, the patient received a minimally invasive treatment with the combination of chemoembolization and microwave ablation. The tumour had shrinkage of about 70% of its size at one-year follow-up and the patient had a remarkable clinical improvement and a second course of this treatment is planned. Even if surgery is needed finally, it will be less extensive and safer. However, a larger series of patients is needed to confirm this application and establish a feasible and reproducible therapeutic protocol for the treatment of bone epithelioid hemangiomas.

References

1. Wenger DE, Wold LE. Benign vascular lesions of bone: radiologic and pathologic features. *Skelet Radiol*. 2000;29:63–74.
2. Sung MS, Kim YS, Resnick D. Epithelioid hemangioma of bone. *Skelet Radiol*. 2000;29:530–534.
3. Floris G, Deraedt K, Samson I, Brys P, Sciot R. Epithelioid hemangioma of bone: a potentially metastasizing tumor? *Int J Surg Pathol*. 2006;14:9–15.
4. Wenger DE, Wold LE. Malignant vascular lesions of bone: radiologic and pathologic features. *Skelet Radiol*. 2000;29:619–631.
5. O'Connell JX, Nielsen GP, Rosenberg AE. Epithelioid vascular tumors of bone: a review and proposal of a classification scheme. *Adv Anat Pathol*. 2001;8:74–82.
6. George A, Mani V, Noufal A. Update on the classification of hemangioma. *J Oral Maxillofac Pathol*. 2014;18(suppl 1):S117–S120. http://dx.doi.org/10.4103/0973-029X.141321.
7. Nielsen GP, Srivastava A, Kattapuram S, et al. Epithelioid hemangioma of bone revisited: a study of 50 cases. *Am J Surg Pathol*. 2009;33:270–277. http://dx.doi.org/10.1097/PAS.0b013e31817f6d51.
8. Kadlub N, Dainese L, Coulomb-L'Hermine A, et al. Intraosseous haemangioma: semantic and medical confusion. *Int J Oral Maxillofac Surg*. 2015;44:718–724. http://dx.doi.org/10.1016/j.ijom.2015.01.025. Epub 2015 Feb 19.
9. Cahill AM, Nijs EL. Pediatric vascular malformations: pathophysiology, diagnosis, and the role of interventional radiology. *Cardiovasc Interv Radiol*. 2011;34:691–704. http://dx.doi.org/10.1007/s00270-011-0123-0. Epub 2011 Mar 16.
10. Gangi A, Tsoumakidou G, Buy X, Quoix E. Quality improvement guidelines for bone tumour management. *Cardiovasc Interv Radiol*. 2010;33:706–713. http://dx.doi.org/10.1007/s00270-009-9738-9. Epub 2010 Feb 12.
11. Thompson SM, Callstrom MR, McKusick MA, Woodrum DA. Initial results of image-guided percutaneous ablation as second-line treatment for symptomatic vascular anomalies. *Cardiovasc Interv Radiol*. 2015;38:1171–1178. http://dx.doi.org/10.1007/s00270-015-1079-2. Epub 2015 Apr 1.
12. Bianchi G, Campanacci L, Ronchetti M, Donati D. Electrochemotherapy in the treatment of bone metastases: a phase II Trial. *World J Surg*. 2016. http://dx.doi.org/10.1007/s00268-016-3627-6. Epub 2016 Jul 21.
13. Zhang W, Chen G, Ren JG, Zhao YF. Bleomycin induces endothelial mesenchymal transition through activation of mTOR pathway: a possible mechanism contributing to the sclerotherapy of venous malformations. *Br J Pharmacol*. 2013;170:1210–1220. http://dx.doi.org/10.1111/bph.12355.
14. Meng J, Zhuang QW, Gu QP, Zhang J, Li ZP, Si YM. Digital Subtraction angiography (DSA) guided sequential sclerotherapy for maxillofacial vein malformation. *Eur Rev Med Pharmacol Sci*. 2014;18:1709–1712.
15. Rybak LD. Fire and ice: thermal ablation of musculoskeletal tumors. *Radiol Clin North Am*. 2009;47:455–469. http://dx.doi.org/10.1016/j.rcl.2008.12.006.
16. Friedman MV, Hillen TJ, Wessell DE, Hildebolt CF, Jennings JW. Hip chondrolysis and femoral head osteonecrosis: a complication of periacetabular cryoablation. *J Vasc Interv Radiol*. 2014;25:1580–1588. http://dx.doi.org/10.1016/j.jvir.2014.06.016. Epub 2014 Jul 23.