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Percutaneous treatment of orofacial vascular malformations

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Abstract

The aim of this study was to evaluate the efficacy of fluoroscopy-guided percutaneous injection of bleomycin as the primary treatment for low-flow vascular malformations. A total of 34 patients (mean (range) age 24 (8–51) years) with orofacial vascular lesions were treated in the Department of Interventional Radiology and Maxillofacial Surgery. There were 20 low-flow venous malformations, 11 lymphatic malformations, and three of mixed type. All patients were treated by fluoroscopy-guided percutaneous injection of a mixture of bleomycin (mean (range) 15 (5–15) mg) and a radio-opaque agent (Ultravist® (iopromide), Bayer)/session. The number of sessions ranged from one to six. The clinical response was complete in 21 patients, obvious in nine, and of clinical benefit in four. Patients were reviewed within the first week, third week, and at three-month periods until 24 months. There were no serious complications such as pulmonary fibrosis. Fluoroscopy-guided intralesional injection of bleomycin should be considered as the first-line treatment for lymphatic malformations because it is effective and reliable with few complications.
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Keywords: bleomycin; sclerosing agent; vascular malformation; percutaneous treatment; fluoroscopy

Introduction

Vascular anomalies can cause tinnitus, headaches, haemorrhage, heart failure and, depending on the site, other functional and aesthetic issues.[1] Successful treatment depends on an accurate diagnosis, which includes localisation of the anomaly, confirmation of its type, arterial supply and venous drainage, and low-flow or high-flow pattern. Recommended treatments include percutaneous sclerotherapy, transarterial embolisation, laser treatment, and complementary therapy.[2]

In 1982, Mulliken and Glowacki proposed a simplified classification system that divided lesions into two groups based on their biological features: haemangiomas and vascular malformations.[3] In 1993, Jackson et al devised a new classification system that included radiological findings and treatment,[4] and this was adopted by the International Society for the Study of Vascular Anomalies (ISSVA) in 1996. It was last updated in 2014.[5,6]

Venous malformations are usually seen during infancy or adolescence, while lymphatic malformations develop during infancy. Most peripheral vascular malformations are venous. About 40% affect the head and neck,[7–10] and symptoms occur in late childhood or early adolescence even though the lesions are present at birth. Venous malformations are compressible, bluish, non-pulsatile, and soft, with expanding skin and subcutaneous tissue.[11,12] They can infiltrate adipose tissue, muscle, fascia, tendon, and bone, and may cause pain and deformity. The diagnosis is confirmed mainly on magnetic resonance imaging (MRI), and T2W2 spin-echo (SE) images show high signal intensity. However, T2 SE images

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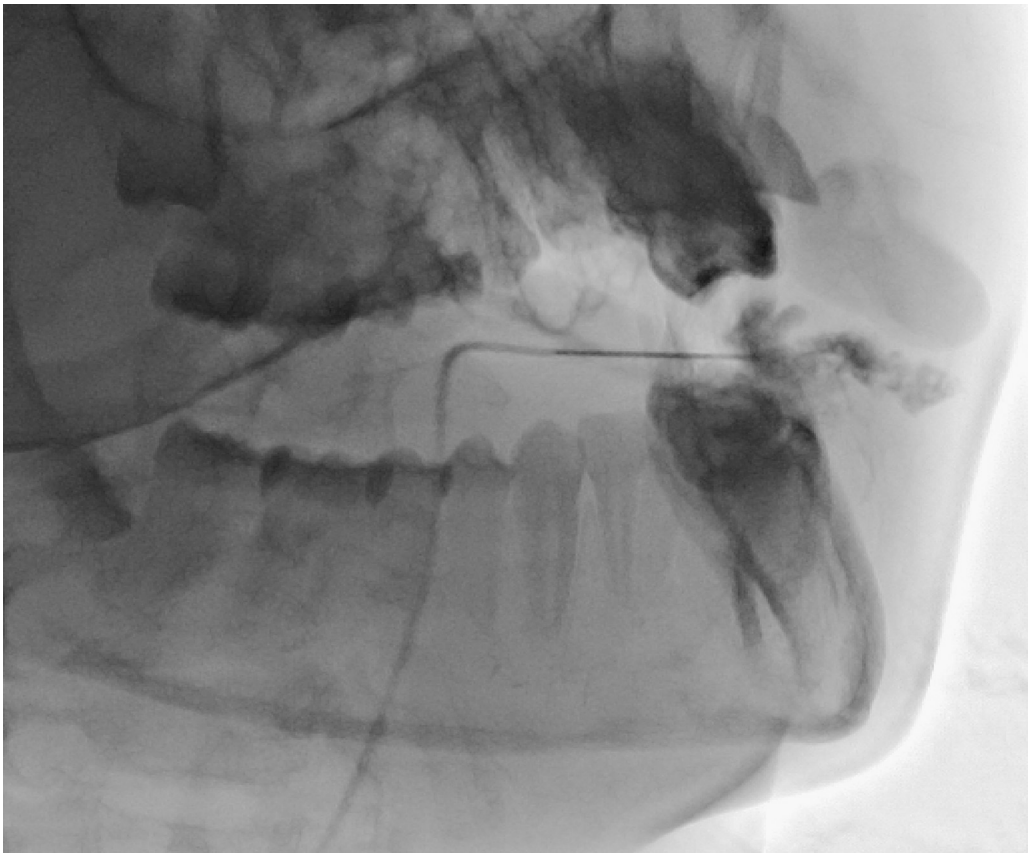

Fig. 1. Bleomycin with radio-opaque agent injected into a vascular malformation of the lip. Sclerosing agent shows stasis in the lesion.

do not show flow voids. Delayed imaging may show diffuse enhancement.[13]

Lymphatic malformations, the second most common type after venous malformations,[14] are soft, non-compressible, rubbery masses[15] that are septate and lobulated. T1W1 and T2W2 images are similar to those of venous malformations and, as with venous lesions, SE images do not show flow voids. Rim and septal enhancement may be seen but there is no appreciable enhancement in the case of microcystic lymphatic lesions.[13] Angiographic examination to detect arterial and venous routes, blood supply, and serious anastomoses can be valuable in selected cases.

Symptomatic venous and lymphatic malformations require treatment, and percutaneous sclerotherapy is considered the mainstay. Uniform or non-uniform particulate agents (ethanol, cyanoacrylate, polydacanol (Aetoxisclerol®, Kreussler), ethyl vinyl alcohol copolymer (Onyx®, ev3 Neurovascular), 2 poly-hydroxyethyl-methacrylate, Ok-432, pingyangmycin (bleomycin A5), and bleomycin) are used as sclerosing agents.[16–19] They have different embolisation characteristics and varied success. To our knowledge, few studies have reported fluoroscopy-guided techniques to reduce seepage and maximise outcome. We describe the use of fluoroscopy-guided injection of bleomycin, and report outcome.

Patients and methods

We studied 34 patients (mean (range) age 24 (8–51) years) who had percutaneous treatment of orofacial vascular malformations and follow up of two years (2013–2015). Ethics approval was obtained from Akdeniz University Ethics Board (No 123; 17.02.2016).

Lesions affected the lips (n = 11), lips and buccal area (n = 12), and tongue (n = 11). There were 20 low-flow venous malformations, 11 lymphatic malformations, and three of mixed type. Before treatment, the mean (range) volume of the lesions was 7.6 (4.0–12.7) cm^3 and the mean (range) maximal diameter 3.5 (2.5–6.5) cm, except in the case of two lesions on the tongue that could not be measured. The main symptoms included pain and difficulty in chewing. Spontaneous or contact bleeding was a frightening symptom in 17 patients, and all the patients complained about the aesthetic appearance of the lesions. The venous malformations were blue, soft, and compressible; lymphatic malformations were non-compressible and rubbery.

We used Doppler ultrasonography to measure the size of the superficial lesions and to assess blood flow. For submucosal or deep lesions we used MRI (sagittal, coronal, and axial views). The volume of the lesions was calculated automatically for both techniques (area of lesion on each slice

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