Treatment of Lymphangioma of the Face with Intralesional Bleomycin: Case Discussion and Literature Review

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Abstract Surgical treatment of lymphangioma of the face is a difficult task to achieve due to close vicinity of the lesion to the facial nerve and possibility of scar tissue formation. Inefficient surgical removals generally will give rise to high recurrence rates because of infiltrative and diffuse extension of the lesion. However, complete cure has been described by non-surgical methods. A 5-year-old girl with extensive lymphangioma of the left cervicofacial area was treated with intralesional bleomycin injection under ultrasonographic guidance. Case discussion and related literature review was presented.

Keywords Face · Lymphangioma · Bleomycin · Treatment

Introduction

Lymphangioma is a rare congenital malformation of the lymphatic system and frequently seen in the head and neck. Surgery is the first line of treatment in general. However, major concern regarding the surgery is difficulty in complete resection due to infiltrative nature of the lesion and therefore high recurrence rates. Recently, nonsurgical alternatives are proposed particularly for those which

envelope vital structures [1]. We report a 5-year-old girl with extensive lymphangioma of the left cervicofacial region extending from zygoma to the submandibular region who has been successfully treated with intralesional injection of bleomycin.

Case Report

A 5-year-old girl attended to our outpatient clinic with the complaint of a large purple mass in her left cheek since birth. She was sometimes unable to swallow due to swelling of her tongue. Magnetic resonance imaging demonstrated a 4 cm cystic lesion extending from anterior edge of the left masseter muscle to the left zygoma superiorly and to the left submandibular region inferiorly and to the contra lateral neck at the submental area. Ultrasonographic evaluation demonstrated anechoic multicystic lesion (Fig. 1). The lesion was hypo intense at T1 and hyper intense at T2-weighted images with no contrast enhancement (Fig. 2a, b). Surgical handicaps were discussed with the parents since the lesion was surrounding peripheral branches of the left facial nerve. Therefore, the patient was planned to be treated with intralesional bleomycin sclerotherapy prior to any surgical intervention. The patient was operated under general anesthesia and at least 4 ml of cystic effusion was evacuated at several sites and a 5 ml of 30 mg of bleomycin was injected to the lesion under ultrasound guidance. The procedure was uneventful. The lymphangioma showed more than 90% shrinkage after bleomycin injection at sixth months (Fig. 3a, b). One year later, magnetic resonance imaging showed 90% decrease of the lesion in size due to fibrotic regression. She has remained asymptomatic since then.

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Fig. 1 USG shows anechoic multilobulated cystic lesion

Discussion

Bleomycin is an antineoplastic drug, but it has been also used locally as a sclerosing agent particularly in cases of congenital lymphatic malformations because of the drug effect on fibrotic transformation of the vascular endothelium. Sclerotherapy has been preferred in the treatment of childhood lymphangiomas recently due to minor side effects and considerable success rates. Sanlialp et al. [2] have reviewed the results of 15 children injected with bleomycin. They reported 90% regression in 53.4% and more than 50% regression in 26.7% of those. Fever (11%), local reactions (4%), and vomiting (2%) were minor

complications following a total of 55 injections. Okada et al. [3] reported significant results in 25 of 29 patients (86%) and complete disappearance in 55% of those (No: 16) after bleomycin injection with a total dose of 3 to 32 mg. However, they have also reported less extent of surgery due to reduced size of the mass as a result of previous injection of bleomycin in 13 patients who have undergone surgery later. Orford et al. [4] have reported complete clinical resolution in 44% and more than 50% resolution in also 44% in 16 patients with cystic hygroma.

Baskin et al. [5] reported complete disappearance of the lesion in six of the nine cases after one single injection. No attempt is usually necessary as long as the mass continues to decrease in size clinically and radiologically. We prefer to use ultrasound for the follow-up. Next injection could be a few weeks later not to let overdosing. Cystic lesions usually show better response to the treatment. Okazaki et al. [6] have compared the effectiveness of the therapy in cavernous and cystic lesions and have found better results in cystic forms. They have recommended sclerotherapy only for cystic lesions. However, sclerotherapy was with OK-432. Ogita et al. [7] have reported significant shrinkage in 22 of 24 cystic and nine of 22 cavernous lymphangiomas after intralesional injection of OK-432. Sclerotherapy can also be used for vascular lesions of head and neck [8]. However, it has been reported that it is much more effective in cystic lymphangioma. In comparison of intralesional bleomycin injection treatment of hemangiomas and lymphangiomas Muir et al. [9] have found complete resolution in 49% of hemangiomas and in 80% of cystic hygromas.

Intralesional bleomycin therapy is an effective alternative to surgery where one can expect to endanger vital structures or cares for poor cosmetic results due to neural

Fig. 2 **a** Coronal and **b** axial T2 weighted image shows left facial heterogeneous hyperintense lymphangioma

Fig. 3 **a** Axial T1 and **b** T2 weighted images show complete regression of the lymphangioma 1 year after application

involvement [10]. Long-term results are also satisfactory [11]. Very few problems can be seen after injection like fever for a few days or skin inflammation. However, in literature review of 289 cases Acevedo et al. [1], have reported seven major complications, including two mortalities most probably due to pulmonary fibrosis. Therefore, keeping in mind overdosing and exact intralesional application under ultrasound guidance are the important points of the procedure. Although our results suggest that intralesional injection of bleomycin can be effective in the treatment of extensive cystic lymphangiomas of the face, larger randomized trials are warranted to assess the current technique.

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