

# Outcomes of surgery for haemangiomas in the upper limb

Andrzej Żyluk ✉

Pomeranian Medical University in Szczecin, Department of General and Hand Surgery, Unii Lubelskiej 1, 71-252 Szczecin, Poland

<sup>A</sup> ORCID: 0000-0002-8299-4525

✉ azyluk@hotmail.com

## ABSTRACT

Haemangiomas are fairly common tumours encountered in the upper extremity. They are composed of non-homogenous groups of lesions of arteriosus, venous or lymphatic origin. The results of surgical treatment of haemangiomas in the upper extremities of 19 patients, 11 women and 8 men, at a mean age of 46 years are presented. Eleven patients (58%) had lesions localized in the digits, 6 (32%) in the wrist or metacarpus and 2 (10%) in the forearm or arm. The duration of the disease until operation was 6 years on average. The most common motivation to undergo

surgery was cosmetic concern. The follow-up assessment was performed in a form of a phone interview for all patients, at a mean of 4 years after the operation. No serious postoperative complications were observed. Four patients were not satisfied with the appearance of the postoperative scar, 1 reported sensory disturbances in the operated finger and another had reduced grip strength of the operated hand. One case (5%) of a recurrence of the lesion was noted, at 1 year after the operation.

**Keywords:** haemangioma; upper extremity; operative treatment; treatment outcomes.

## INTRODUCTION

Haemangiomas are fairly common tumours encountered in the upper extremity. They are composed of non-homogenous groups of lesions of vascular (arteriosus, venous or lymphatic) origin [1, 2]. Haemangiomas occur more frequently in children than in adults and, although histologically are similar, they differ to each other in many respects. Infantile haemangiomas can be congenital, however they usually appear in the first months after birth. They present as flat, reddish spots in the skin or soft, shining through, red-cyanotic masses located subcutaneously. Infantile haemangioma is considered an immature form of the lesion, having a tendency towards spontaneous involution. Adult haemangioma is a mature form of the lesion, and, unlike the infantile form, after reaching a static phase of growth, it does not tend to disappear [1, 3].

The appearance of haemangiomas varies at a macroscopic level. Some variances include telangiectasia, soft subcutaneous lesions, until exophytic tumours resembling dark grape clusters. Histologic evaluation of adult haemangiomas reveals 2 distinct types. Capillary type lesions are nonvascular, with a spongy appearance. The cavernous type has large vessels, with thin walls lined by a flat endothelium. The distinction, however, is difficult because there is a spectrum of disease [1]. Capillary tumours are usually smaller than cavernous, not disturbing and are relatively easy to treat (Fig. 1). The venous type of cavernous lesions can reach a big size and may cause several problems, including ulceration, bleeding, finger deformation, bone atrophy and functional impairment (Fig. 2, 3, 4).

Another form of the lesion is an intramuscular haemangioma. This tumour is uncommon and is estimated at less than 1% of all haemangiomas. Intramuscular lesions are more prevalent in the lower extremity than in the upper extremity. These

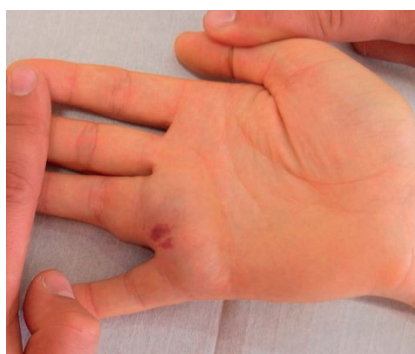


Please note cyanotic shining-through of the lesion under the skin.

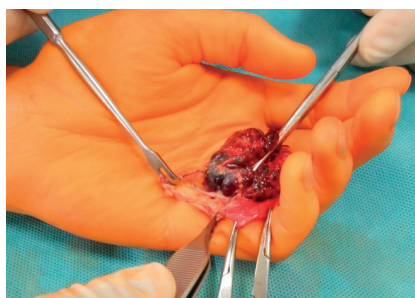
**FIGURE 1.** Haemangioma in the ring finger

tumours typically grow between the flexor muscles in the forearm or in the calf, and, unlike classical haemangiomas, they are not visible through the skin. Intramuscular lesions present as a painful swelling, particularly during periods of strenuous exercise. Magnetic resonance imaging (MRI) is particularly useful in the diagnosis of these tumours, especially if surgical intervention is planned. Histologic evaluation of lesions is important because they can be confused with malignant tumours such as sarcomas or neuroblastomas [1, 3].

Vascular malformations are considered another type of vascular tumour, different from haemangiomas. They are caused by errors of vascular tree morphogenesis; they do not have involution and grow in proportion to the individual, although their growth might accelerate during puberty and pregnancy. Male and female patients have equal incidence. Vascular



**FIGURE 2.** Haemangioma in the palmar side of the metacarpus



**FIGURE 3.** Excision of the tumour from the palm



**FIGURE 4.** A hole after excision of the tumour from the palm

malformations primarily fall into 2 categories: low-flow (more common) and high-flow (less common). Low-flow vascular malformations are divided into capillary, venous, lymphatic, and combined types. This distinction is made based on the predominant type of vessel comprising the mass [1, 3].

### Diagnostics

Imaging modalities such as USG, Doppler-USG, angio-CT and MRI are used for the diagnosis of large, intramuscular haemangiomas or when the diagnosis is questionable [3]. Lesions less than 3 cm in diameter and superficially located are excised without additional examination. In some cases, an incisional biopsy may be necessary.

The treatment of haemangiomas is primarily observational. Most haemangiomas will spontaneously involute. However, some symptomatic or persistent lesions require additional management. Treatment options include intralesional or systemic steroids, pulsed dye lasers, intralesional sclerosing agents and bleomycin. Surgical treatment is indicated when the haemangioma is refractory to first-line treatment or is symptomatic.

Typical indications to surgery include: pain, symptoms of nerve compression, ulceration, bleeding, functional impairment or cosmetic concern, where the tumour looks inaeesthetic. The range of operations varies and is related to the size of the tumour, its location (deep or superficial) and presence of nerves in the vicinity. Small lesions are resected *in toto*, whereas large tumours, occupying the whole digit or a part of the hand, may require staged operations, as resection of such a mass in 1 session can result in ischaemia and subsequent necrosis. Surgery can be difficult owing to the abnormal anatomy, as well as the potential for substantial bleeding [1]. In some tumours, an axial (central) artery supplying the lesion can be identified; its ligation facilitates then excision of the tumour, reducing bleeding significantly. Embolization may be considered too, however it is associated with some risk of necrosis of the digit. Recurrence rate after surgical treatment of vascular tumours is estimated at 1–5% [4].

### Associated syndromes

There are several clinical syndromes in which haemangiomas are one of the components [1, 3]:

- Klippel–Trenauney syndrome consists of a mixed venous and lymphatic lesion, cutaneous nevus flammeus, varicosities, and hypertrophy of the bone;
- Kasabach–Merritt syndrome is a bleeding diathesis due to a consumptive coagulopathy with thrombocytopenia and purpura secondary to a vascular lesion;
- Maffucci's syndrome is a rare, congenital enchondromatosis and hemangiomatosis that appears at birth. It results in deformities of the hands and can have a malignant transformation;
- Parkes–Weber syndrome includes arteriovenous fistulas, vein varicosities, and limb hypertrophy from lipomatosis and lymphatic hyperplasia.

Only 1 article presenting outcomes of the surgery for haemangiomas in the upper limb in adults was found in Polish literature [5]. The objective of this study was an assessment of the results of surgical treatment of haemangiomas in the upper limb in adults.

### MATERIALS AND METHODS

Between 2015–2018, 19 patients, 11 women and 8 men, at a mean of 46 years of age (range 23–79) with soft tissue tumours in the upper extremity which after histologic examination appeared to be haemangiomas were operated on in the author's institution. These constituted 5.5% (19/346) of all tumours operated on in this period. Three cases of intramuscular cavernous haemangiomas were not included in the study group due to different treatment modalities and a poorer prognosis of this type of lesion. Eleven patients (58%) had a tumour localised in the digits, 6 (32%) in the wrist or metacarpus, and 2 (10%) in the forearm and arm. Ten lesions were present in the left limb, whereas there were 9 in the right one. Three patients had



more than 1 tumour in one extremity (Fig. 5). The duration of the disease until the operation was 6 years on average (range 1–20). The most common motivation to undergo surgery was cosmetic concern, but 6 patients also complained of a mild, throbbing pain. Four patients reported some problems with grasping objects, when lesions were localized in the palm or fingers. In 13 cases, diagnosis was made on a clinical basis, in 5 it was completed by ultrasonography and in 1 by MRI.



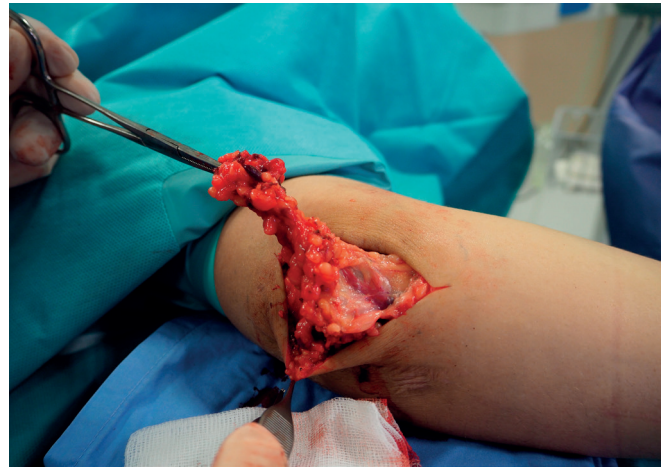
**FIGURE 5.** Haemangiolipomas at the elbow and in the forearm

All patients were operated on; small tumours in the digits were excised under local anaesthesia and with a rubber tourniquet on the digit. Bigger lesions and those localised in the wrist, metacarpus, forearm or arm were resected under brachial plexus block anaesthesia and with the tourniquet on the arm. All resected tumours underwent histologic examination which confirmed clinical diagnosis of the specific form of haemangioma. The follow-up assessment was performed through a phone interview with all patients, at a mean of 4 years (range 3–5) after the operation.

## RESULTS

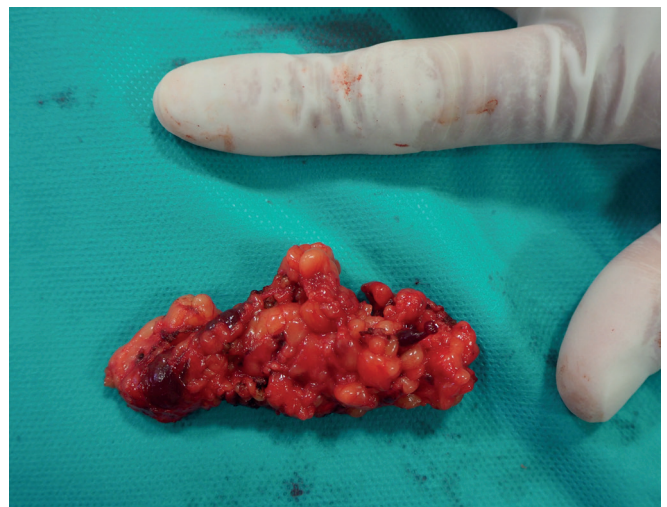
The results of the treatment were evaluated in all 19 patients. Intraoperatively, none of the tumours had a well-defined capsule, however 1 was distinctly separated from surrounding tissue (Fig. 3, 4).

All tumours were located superficially under the skin or in the subcutaneous tissue. No particular problems with bleeding control during the operations were observed, however obtaining haemostasis was usually time consuming. No cases of ischaemia of the digit after resection of the lesion were observed. The healing of wounds was uneventful in most patients except in 3 cases (16%) where there was superficial infection and 2 (10%) of marginal skin necrosis. Histological examination revealed the following variants of haemangiomas: 7 of adult type, 6 of venous type, 4 of capillary type, 1 spindle-cell type and 1 haemangiolipoma (Fig. 5, 6, 7). All patients were satisfied with the outcome of the treatment. However,



Note mixed structure of the tumour

**FIGURE 6.** Resection of the tumour from the elbow



**FIGURE 7.** Resected tumour

4 patients (16%) complained of an inaeesthetic postoperative scar, 1 of sensory disturbances in the finger operated on and 1 of a weaker grip in the operated hand. All employed patients returned to their previous jobs. One case (5%) of recurrence occurred, 1 year after resection of the haemangiolipoma from the forearm. This patient was re-operated on with a good final outcome.

## DISCUSSION

The results of this study show that haemangiomas constitute a relatively small subgroup among all benign, soft tissue tumours encountered in the upper extremities in adults. In most cases, they were asymptomatic, and the principal cause to undergo operation was due to cosmetic (aesthetic) concerns. For most patients, the result of the operation was satisfactory and no serious complications were noted. The most common complaint at the follow-up assessment was the same as pre-operatively: an inaeesthetic appearance of the hand. No case of serious deterioration of function of the hand was observed. It should be mentioned that the study group comprised only

superficially located haemangiomas, and intramuscular cavernous lesions were excluded as treatment is more demanding and associated with poorer outcomes. Surgery of haemangiomas may be demanding for the following reasons:

- it rarely has a well-defined capsule and usually needs resection from adjacent tissue;
- despite a tourniquet on the arm, the operative field is usually unclear because of residual blood filling the tumour vessels which outflows during preparation. It makes identification of fine anatomical structures more difficult;
- large lesions may require resection with the overlying skin and the defect may need coverage with a skin graft.

There was 1 case of haemangiolipoma in the study group (Fig. 3). This lesion represents rare tumours of mixed nature and vague aetiology, characterised by a simultaneous outgrowth of endothelial cells and adipocytes. They typically present as soft, shining through, red-cyanotic masses located subcutaneously. In most cases, they are indistinguishable from typical haemangiomas preoperatively and the correct diagnosis is made after histologic examination.

## LITERATURE REVIEW

There is scarce information in the literature about the results of surgical treatment of haemangiomas within the upper limb of adults. In contrast, management of these lesions in children is much better documented. Only 1 paper was published in Polish literature, by Jabłecki et al. [5], regarding adult haemangiomas. These authors presented the results of the operative treatment of 11 patients, 8 women and 3 men, aged 24–39 years, suffering from haemangiomas and vascular malformations in the upper limb. The lesions were localized in the fingers of 4 patients, in the fingers and metacarpus of 2, solely in the metacarpus of 2 and in the forearm or arm of 4. Some of the patients had symptoms of nerve compression caused by the tumour. No details concerning operations were provided, but the pictures attached to the paper had shown a relatively excessive range of resection of the tumours localized in the palmar metacarpus and in the forearm. At least one of these cases was a cavernous type of haemangioma. Histological examination revealed haemangiomas in 6 patients and vascular malformations in the other 5. The authors reported 4 cases of recurrence, but the follow-up period was not provided. All relapses occurred following resection of vascular malformations involving fingers. The authors conclude that

surgical treatment of vascular tumours in the upper extremities is demanding, requires knowledge of microsurgical techniques and is associated with a relatively high recurrence rate. Vascular malformations are often located in the digits which may increase the risk of postoperative ischemia [5].

Tang et al., in a retrospective review of 89 patients treated over 25 years, evaluated a variety of vascular tumours, including haemangiomas. This study showed a marked decrease in recurrence following removal with clean surgical margins. Intralesional or marginal resection resulted in a 19% recurrence rate. A larger tumor was the primary determinant for recurrence. Overall, 83% of the patients reported excellent function with no impairment and only 2% reported poor function [6].

In a relatively old study, Palmieri reported results of surgery for subcutaneous haemangiomas of the hand. A review of 160 patients, 99 women (62%) and 61 men (28%) at a median age of 32 years (range 1–68) with the histologic diagnosis of subcutaneous haemangiomas is presented. None had a history of trauma. The most common location of lesions was the palmar side of the hand. The most common symptoms reported by the patients were a progressive enlargement of the lesion and throbbing pain. Most tumours presented as readily compressible, poorly defined, bluish, subcutaneous masses that distended when the venous return was obstructed and contracted at elevation of the limb. Diagnosis was made on a clinical basis, with the addition of X-ray examination in some cases. All of these haemangiomas were surgically excised, with identification and ligation of the axial vessels as far distant from the tumor as possible. In a 4-year follow-up, 9 cases (6%) of recurrence were noted [2].

## REFERENCES

1. Jacobs BJ, Anzarut A, Imbriglia JE. Vascular anomalies of the upper extremity. *J Hand Surg Am* 2010;35(10):1703-9.
2. Palmieri TJ. Subcutaneous hemangiomas of the hand. *J Hand Surg Am* 1983;8(2):201-4.
3. Ernemann U, Kramer U, Miller S, Bisdas S, Rebmann H, Breuninger H, et al. Current concepts in the classification, diagnosis and treatment of vascular anomalies. *Eur J Radiol* 2010;75(1):2-11. doi: 10.1016/j.ejrad.2010.04.009.
4. Freshwater MF. Treatment of vascular anomalies of the upper extremity. *J Hand Surg Am* 2011;36(2):370.
5. Jabłecki J, Elsaftawy A, Kaczmarzyk J, Kaczmarzyk L. Surgical treatment of hemangiomas and arteriovenous malformations in upper extremity. *Pol Przegl Chir* 2013;85(3):107-13.
6. Tang P, Hornicek FJ, Gebhardt MC, Cates J, Mankin HJ. Surgical treatment of hemangiomas of soft tissue. *Clin Orthop Relat Res* 2002;399:205-10.