

Hemangiomas in Children: Challenges and Outcome of Surgical Management in Benin City, Nigeria

Osarumwense David Osifo*, FWACS, FICS, and Iyekoretin Evbuomwan, FWACS, FRCS

Pediatric Surgery Unit, Department of Surgery, University of Benin Teaching Hospital, Benin City, Nigeria

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Abstract

Objective: Treatment of hemangioma/vascular tumors emphasize minimal invasion which require sophisticated facilities. This study reports the role of surgery in the management of symptomatic, and hemangioma which failed to respond to other modalities of treatment in resource-limited subregion.

Methods: A six-year (2004-2009) prospective study on the challenges and outcome of children referred for surgical management of hemangioma/vascular tumors was undertaken at the University of Benin Teaching Hospital, Nigeria.

Findings: Sixty-three children aged between a day and six years (median 5 years) comprising 38 males and 25 females (ratio 1.5:1) were managed. Upper limbs involvement, 18 (28.6%), and face/neck, 12 (19.1%), were most common and were present at birth in 27 (42.9%) babies, appeared between 2-3 weeks in 32 (50.8%), and after six months in 4 (6.3%). Cavernous hemangioma in 19 (30.2%) children, mixed cavernous/strawberry in 31 (49.2%) and strawberry in 13 (20.6), were the major types that ranged from spot-like to extensive huge lesions measuring 12×15 cm in diameter. Failure of 46 (65.1%) cases to respond to non operative treatment, ulceration in 3 (4.8%), infection in 5 (7.9%) and hemorrhage in 2 (3.2%) were indications for surgical intervention. Surgical options included complete excision and primary wound closure in 34 (54%) children, immediate skin graft after complete excision in 10 (15.9%), injection sclerotherapy in 2 (3.2%), serial ligation of feeder vessels in 2 (3.2%), and conservative treatment in 5 (7.9%). Excision and primary wound closure gave better outcome compared with others ($P<0.0001$). No mortality was recorded on 1-6 years follow-up but ugly scar, 8 (12.7%) and limb deformity, 3 (4.8%) were problems.

Conclusion: Surgical excision and primary wound closure gave good outcome which could be employed in complicated and hemangioma which failed to respond to other treatment in regions with limited resources.

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* Corresponding Author;

Address: Pediatric Surgery Unit, Department of Surgery, University of Benin Teaching Hospital, Benin City, Nigeria

E-mail: leadekso@yahoo.com

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Introduction

Vascular tumors and hemangioma are hamartomas, a group of benign tumors that can be found anywhere in the body and are collectively referred to as hemangiomas by many researchers^[1-3]. They may be syndromic or non-syndromic and are rarely present at birth but appear during the second to third weeks of life as bluish, pink or reddish subcutaneous lesions of any shape that may rapidly increase in size^[4,5]. The lesions are very common in infants and children comprising a spectrum of cavernous, strawberry, port wine stain, spider nevus, arterio-venous malformation and truly vascular tumor^[1-3,6]. Various internal and external organs involvement including hepatic, mediastinal, pulmonary, gastrointestinal, and cerebral hemangiomas, as well as simultaneous multiple organs lesions have been described^[5,7].

Spontaneous involution that is rare in vascular tumor is said to be very common in true hemangioma^[1-3]. Although the lesions are benign and composed of vascular tissues that are haphazardly arranged, life threatening complications have been reported in many series^[8-10].

Various modality of treatment which includes conservative treatment, surgical excision, injection sclerotherapy, cryotherapy, laser treatment, angiographic embolization, angioplasty/binding of feeder vessels, use of angiogenesis inhibitor drugs, and especially the use of corticosteroids which are readily available have given good results in many centers^[5,8,11,12].

In many evolving pediatric surgical centers particularly in sub-Saharan Africa, however, there are limited approaches to management owing to lack of facilities^[13]. Management of hemangioma/vascular tumors in such resource-limited centers remains a major challenge especially when the lesion is located in sensitive places and in patients presenting with life threatening complications. There are, however, scant reports on the challenges and outcome of surgical management of hemangioma and vascular tumors in this Africa subregion^[13].

The aim of reporting this six-year experience on the challenges and outcome of surgical management of hemangioma/vascular tumors is to emphasize the role of surgery in the

management of very symptomatic hemangioma that may not respond to other modalities of treatment.

Subjects and Methods

This prospective study on the challenges and outcome of surgical management of hemangiomas in children was undertaken between January 2004 and December 2009 at the University of Benin Teaching Hospital, Benin City, Nigeria. Following ethical approval by the hospital Local Ethics Committee, all the children who were referred from pediatric department of this hospital and other hospitals in neighboring cities after failure of at least four years of non operative management and/or complicated lesions were included in the study.

Those whose lesions resolved spontaneously and/or during non operative treatment follow-up were excluded. The distinction between vascular tumor and hemangioma was not applicable in this study as it did not significantly influence the choice and outcome of treatment.

In view of the lack of adequate facilities required for injection sclerotherapy, cryotherapy, laser treatment, angiographic embolization, angioplasty/binding of feeder vessels, and use of angiogenesis inhibitor drugs, the following goal oriented classification and treatment options were adopted during the period:

1. Skin/subcutaneous lesion up to 5cm in transverse diameter had excision and primary wound closure.
2. Skin/subcutaneous lesions >5cm in transverse diameter had excision and immediate skin graft.
3. Skin/subcutaneous lesions ≤5cm but complicated by infection, ulceration or hemorrhage had excision and delayed wound closure.
4. Skin/subcutaneous lesions >5cm but complicated by infection, ulceration or hemorrhage had excision and delayed skin graft.
5. Huge lesion involving all soft tissues had excision and immediate myocutaneous flap.

6. Huge lesion involving all soft tissues but complicated by infection, ulceration or hemorrhage had excision and delayed myocutaneous flap.
7. Huge lesions involving vital structures had attempted serial feeder vessels ligation, injection sclerotherapy (5% phenol in almond oil).
8. Isolated and extensive strawberry lesions had conservative treatment.

The biodata of the children, type of hemangioma, associated malformations, clinical conditions on arrival, prior treatment received, surgical options, challenges and outcome which were collated on pre-structured forms were entered into Microsoft Office Excel 2007 sheet and analyzed as counts, frequency and percentages. Continuous data were expressed as mean/standard deviation while categorical data were analyzed using Chi-square test with a P -value ≤ 0.05 regarded as statistically significant.

Findings

On the whole, 63 children who were aged between a day and six years (median 5 years) comprising of 38 males and 25 females (ratio 1.5:1) were referred for surgical management.

As shown in the bar chart (Fig. 1), upper limbs involvement was most common accounting for 18 (28.6%), followed by face/neck 12 (19.1%), while axillary and intra-oral hemangioma accounted respectively for 3 (4.8%) cases. The lesion was

present at birth in 27 (42.9%) babies while it appeared between 2-3 weeks in 32 (50.8%), and after six months in 4 (6.3%). Cavernous hemangioma seen in 19 (30.2%) children, mixed cavernous/strawberry in 31 (49.2%) and strawberry in 13 (20.6), as shown in figures 2-5, were the major types of venous and capillary hemangioma referred for surgical management. The sizes of the hemangioma ranged from spot-like lesion to extensive lesion measuring about 12×15 cm in diameter, and from a flat lesion to a huge mass (Fig. 4 and 5). The diagnosis of hemangioma was made based mainly on clinical evaluation and required no sophisticated investigations.

However, in some patients where it was indicated, radiological and laboratory investigations were employed to screen for syndromic hemangiomas. Apart from Kasabach-Merritt syndrome which was diagnosed in two children, other types of syndromic hemangiomas, internal and multiple sites involvement were not recorded.

Forty-one (65.1%) cases who were clinically stable with uncomplicated hemangioma presented within the first three months of life to pediatricians and received corticosteroid and non operative treatment for at least four years without response. On the other hand, 22 (34.9%) children presented after three months and were referred with life threatening complications which included ulceration in 3 (4.8%), infection in 5 (7.9%) as shown in Fig 3, and hemorrhage in 2 (3.2%) children who had thrombocytopenia (Kasabach-Merritt syndrome). Surgical options and the outcome are as depicted in Table 1.

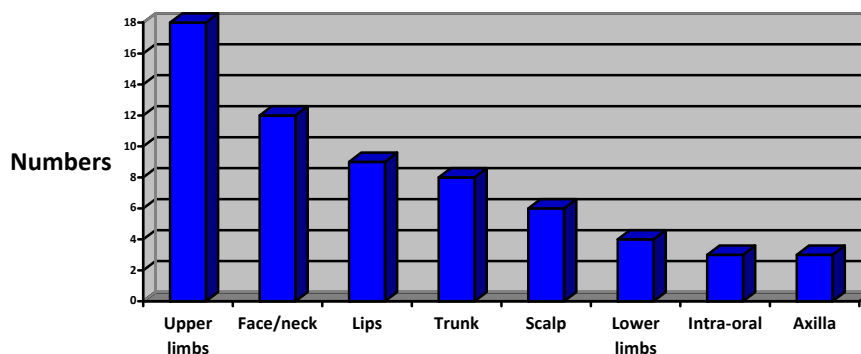


Fig. 1: Site distribution of haemangioma



Fig. 2: A 2-month-old male baby with mixed cavernous/capillary lesion involving the face and neck. He had successful excision and primary skin graft with good cosmetic result.



Fig. 3: Extensive capillary hemangioma involving the right hand in a 5-month-old girl. The lesion underwent complete spontaneous involution during three years of follow-up.

Complete excision and primary wound closure in 34 (54%) children was the most commonly employed option followed by immediate skin graft after complete excision in 10 (15.9%).

Outcome of injection sclerotherapy in 2 (3.2%) children, serial ligation of feeder vessels employed in 2 (3.2%), and conservative treatment in 5 (7.9%), were poor and required conversion to surgical excision. During another non operative follow-up for two years in four children with strawberry hemangioma complete spontaneous

involution still occurred (Fig 3). Excision and primary wound closure gave better outcome compared with other surgical options ($P < 0.0001$). Multiple excisions were required to completely eradicate the hemangioma in 12 (19.1%) children, including six of the nine children with lips lesions.

Although ugly scar and limb deformity were recorded in 8 (12.7%) and 3 (4.8%) children respectively, no postoperative life threatening morbidity or mortality were recorded on 1-6 years of follow-up.



Fig. 4: A huge cavernous hemangioma involving all the soft tissues of the left arm with recurrent episodes of bleeding and thrombocytopenia (Kasabach-Merritt syndrome) in a one-year-old girl. She received platelet and whole blood transfusion and had excision and delayed myocutaneous flap with residual deformity.



Fig. 5: A two month-old-boy with infected huge cavernous hemangioma involving all soft tissues of the left forearm. Incision and drainage, broad spectrum antibiotics coverage followed by excision and delayed myocutaneous flap were employed with resultant severe postoperative deformity.

Table 1: Surgical options and outcome of hemangiomas over 6 years

Surgical options	Frequency	Outcome
Excision/primary closure	34 (54%)	Wound infection 2, excellent cosmetic result 34
Excision/delayed closure	6 (9.5%)	Scarred 3, excellent cosmetic result 3
Excision/Immediate skin graft	7 (11.1%)	Graph failure 1, excellent cosmetic result 6
Excision/delayed skin graft	3 (4.8%)	Excellent cosmetic results in all
Excision/myocutaneous flap	4 (6.3%)	Deformity 3, scarred 1
Serial feeding vessels ligation	2 (3.2%)	Hemorrhage/infection, poor outcome
Sclerotherapy (5% phenol in oil)	2 (3.2%)	Failed, required excision
Conservation treatment	5 (7.9%)	Resolution 4, required excision 1

Discussion

Although no mortality was recorded, surgical management of hemangioma/vascular tumors posed challenges in this and other studies in similar settings [7,13]. This was especially so in children who presented with huge mass involving all soft tissues, those with lesion located in sensitive places like the face/neck and those who presented with life threatening complications [7,9,10,13-15]. Male children were more affected than females with a ratio of 1.5:1, and children older than six years were not seen with hemangioma/vascular tumors in this study which corresponded with other reports [1,3,5,14]. Surgical excision was the only available and affordable treatment option for the referred children owing to lack of modern facilities that are currently in use in other centers [4,11,12,16]. Utilization of such facilities would have lessened surgical burden and the postoperative deformity and poor cosmetic results in lesions which involved the limbs and face/neck. Nevertheless, lesions which were amenable to surgical excision and primary wound closure gave overall best cosmetic results followed by excision and primary skin graft.

Those which required delayed myocutaneous flap were mostly associated with postoperative ugly scars, deformity and impaired functions.

The use of injection sclerotherapy and selective feeder vessels ligation were associated with complications and poor results which necessitated conversion to surgical excision. This could be because 5% phenol in almond oil was the only available and affordable sclerosant in the center. Other authors [5,8] recorded successes with the use of hypertonic saline as sclerosant in their centers

which was not attempted in this study. The lack of facilities required for angiography ligation and embolization made outcome of selective ligation of feeder vessels very poor because identification of the major feeding vessels was difficult and the occlusion of the major draining vessels required to retain the sclerosant to the lesion was not possible [5,7,12,17].

Facial involvement posed cosmetic challenge but non availability of other treatment options and the failure of spontaneous involution and injection sclerotherapy influenced the choice of surgical excision as a last resort. However, excision with primary wound closure and/or primary skin graft gave the best results in lesions located in this region among the surgical options.

Life threatening hemorrhage, ulceration and infection were the main complications which necessitated surgical intervention as recorded in other studies [5,9,10,13-15]. These complications influenced the choice of delayed surgical wound closure with poor cosmetic results. Infected massive hemangioma required incision and drainage, dressing and use of broad spectrum antibiotics before excision. This increased the duration of hospitalization and cost of treatment compared to children with similar uninfected lesions. The two children with thrombocytopenia (Kasabach-Merritt) who presented with massive tumor and recurrent bleeding posed a major challenge in this study unlike reports from more equipped settings [5,10,18,19]. Multiple transfusions of whole blood and platelet concentrate followed by excision and myocutaneous flap were able to save the lives of affected children though some were with residual deformity.

Hemangioma/vascular tumors involving the

upper limbs were more common, followed by face/neck involvements unlike other studies in which face/neck lesions predominated [5,12]. This could be because many face/neck lesions were not referred for surgical consultation as they were successfully managed by paediatricians, involuted and required no surgical intervention [20]. Some hemangiomas referred for surgical management still underwent spontaneous involution in this study.

All hemangiomas were said to involute during childhood in other studies [5,6,20]. Many hemangioma that involuted spontaneously did not seek surgical consultation in this setting. Lips involvement, though less commonly encountered, posed a unique challenge as complete single stage excision of the majority of hemangioma in this location was not possible.

The rapid recurrence and incomplete excision recorded required multiple secessions to completely eradicate the tumor as also reported by others [21]. The limitation of this study, however, was the difficulty in categorizing hemangioma and vascular tumors into separate entities as have been emphasized by other authors [1,2,5,6,19,22] who stressed this as the major factor determining the choice of management. In centers with sophisticated facilities, surgical excision would have been avoided in many cases which make findings from this study applicable mainly to centers with limited resources.

Conclusion

Surgical management of hemangioma/vascular tumors was challenging owing to involvement of sensitive places, huge lesions involving all soft tissues and presentation with life threatening complications. Excision with primary wound closure and primary skin grafts gave overall best outcome in this setting. No mortality was recorded but residual deformity and poor cosmetic results were problem in some cases.

Complicated hemangioma and those which failed to respond to non operative treatment should be offered surgical excision and primary wound closure when possible in regions with limited resources.

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Conflict of Interest: None

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