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External Beam Radiotherapy as an Adjuvant in the Management of Recurrent Digital Heamangiomatous Tumour: A Case Report

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Authors' contributions

Both authors played active and collaborative roles in the case being reported. Author RSA managed the surgical and conservative aspects of the case while author AAA was responsible for the radiotherapy management. The draft and final manuscripts were prepared by author RSA. The submitted manuscript was read and approved by both authors RSA and AAA.

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Case Report

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ABSTRACT

Background: Heamangioma constitutes the largest subset of vascular lesions. It is predominantly congenital with predilection for the head and neck region of the body but can involve any part or region of the body. The capillary variant may be post-traumatic. After initial proliferation it may involute especially if infantile. Treatment modalities include drugs, laser and surgery. Capillary heamangioma may be refractory to drug therapy or recur after surgical excision. A case of capillary heamangioma in a middle aged male negriod (Nigerian) that developed from an ulcerated nodule over the middle phalanx of the left 4th finger with recurrence after several reexcisions is being reported. A permanent cure was achieved after a re-excision preceded by low dose external beam radiotherapy without functional compromise or altered aesthesis of the finger.

Conclusion: Post traumatic heamangioma clinically presents as hyper granulated tissue and may pose a management challenge. Arising from the excellent outcome of the low dose external beam radiation followed by excision, we hereby propose that this should be adopted as one of the treatment options of heamangiomas especially in adults.

Keywords: Recurrent capillary heamangioma; external beam radiotherapy.

1. INTRODUCTION

Vascular lesions largely manifest in the skin but could affect internal organs such as the liver and may be located in any region of the body however most cases are restricted to the head and neck region. These vascular lesions may be congenital or post-traumatic. They include capillary heamangioma (commonest), venous lake and venous malformation. Unlike infantile heamangioma that has been reported to be the commonest benign soft tissue tumour of childhood with an incidence of 5% [1,2]; the exact epidemiology of adult post-traumatic capillary heamangioma is unknown. Capillary heamangioma may involute, amenable to drug therapy or require surgical excision. Recurrence following excision may become a management issue.

We hereby report a case of recurrent digital capillary heamangioma that initially posed a management challenge but was finally cured with pre-excisional external beam radiotherapy.

2. CASE REPORT

A 45 year old male Nigerian Christian Cleric noticed a mildly aching nodular swelling over the dorsal surface of the middle phalanx of the left ring (4th) finger. There was no preceding trauma, bite or sting to the finger. It increased in size rather slowly and ulcerated about five months after onset and noticed to bleed considerably. It was consequently excised and dressed with normal saline and sofratulle. A month later, rather than healed, it hyper granulated (Fig. 1).

The recurrent growth was cauliflower in appearance, pinkish having a 1 cm by 1 cm diameter along perpendicular axes. It had an inverted edge, firm base and bled to touch.

He continued with the conservative management until five months later by that time, it had tripled the initial size with other characteristics remaining unchanged (Fig.2). The wound was subsequently excised completely leaving a very scanty base (Fig. 3). The excised hyper

granulated tissues were sent for histology. The report of which was Lobular capillary heamangioma.



Fig. 1. First recurrence- 1cm by 1 cm hypergranulated wound



Fig. 2. Just before re-excision. About five months after Fig. 1

Post re-excision, the wound was managed conservatively by dressing it with Sofratulle-N with the aim of achieving healing. Rather than achieving this aim, the wound hypergranulated and a decision to employ radiation therapy was taken. In preparation for the radiation therapy, he had the following investigations- abdominopelvic ultrasound scan, retroviral screening, serum electrolyte and urea; and full blood count; none

of which was abnormal. He had 12 Gy of external beam radiotherapy in three fractions at interval of two weeks to the lesion four months after the reexcision. The lesion subsequently became oedematous with sloughy necrotic roof (Fig. 4). As a result of this development, a decision for another re-excision was taken.



Fig. 3. Re-excision five months after fig. 1



Fig. 4. Post external beam radiotherapy

The second re-excision (ie the third excision was done two months after the external beam radiotherapy. The lesion was completely excised leaving healthy margin and base (Fig. 5).

Subsequently, the wound was managed conservatively with routine alternate day dressing and healing monitored. The progression of healing though somewhat slow but was satisfactory with the wound bed filling up, inward migration of the edges and epithelization of the surface. About 10 weeks post second reexcision, the wound had healed as evidenced by a hypopigmented flat but slightly scaphoid scar

measuring 1.8 by 1.5 cm (Fig. 6). The patient could actively flex both the proximal and distal interphalangeal joints of the affected finger maximally without pain. At 6 month post reexcision review, the pigmentation of the scar had increased, the scar had reduced in size but no contracture and full functionality of the finger retained (Fig. 7).



Fig. 5. Post radiotherapy re-excision

3. DISCUSSION

Heamangiomas are essentially benign tumours of the blood vessels characterized by increased number of normal and abnormal vessels. They occur largely in infancy and childhood and could be congenital. Several variants have been described but the pyogenic granulated variety is worthy of note. This variety arises from capillary, grows rapidly, reddish in appearance and pedunculated. It bleeds easily and often ulcerated. A reasonable number of cases occurs after trauma. Curettage and cautery is said to be curative in most cases. [3]. Heamangiomas result from rapid growth of endothelial cells [4,5]. Although there are several types of heamangiomas, the capillary variety is the commonest and is characterized by a closely packed numerous small capillaries separated by thin connective tissue. These capillaries are of normal morphology but large in number. [6]. Hemangiomas arise mostly from the skin and rarely from deep seated organs such as the liver, lung, intestine and brain [7]. They are well circumscribed, round to oval in shape and usually bright red in colour. Most heamangiomas

are solitary and localized however in very few cases, they may be diffuse. Although infantile heamangiomas are known to exhibit a life cycle of proliferation and involution, post traumatic variety does not involute as exemplified by the index case which underwent repetitive excisions. Drugs that have been used to manage infantile heamangioma include corticosteroids [8] and interferon-alpha [9]. These are currently not in use due to the side effects. Propranolol has been tried with remarkable success [10-12]. It is postulated that the drug causes involution of heamangioma by suppressing angiogenesis, capillary vasoconstriction and induction of apoptosis. Propranolol does this by targeting the stem cells via cAMP and mitogen-activated protein kinase regulation [10]. Infantile heamangiomas that failed to involute after 6-12 months of propranolol therapy have been treated with intralesional injection of pingyangmycin (bleomycin A₅)- is an antitumour glycopeptide antibiotic: [11]; intravenous vincristine. subcutaneous interferon α or topical application of timolol [12]. All these constitute advanced therapies and their use is limited by the side effects.



Fig. 6. Healed left 4th (ring) finger wound with minimal scaring and no loss of function

Although the index case presented as hypergranulated wound, one of the known complications of heamangiomas is ulceration. Ulcerated infantile heamangiomas have been successfully managed with Pulsed-dye laser therapy [13].



Fig. 7. Left 4th (ring) finger (six months post re-excision)

Note the increased pigmentation of the scar

Surgical management of vascular lesions with Laser has been in practice for more than five decades [14-16]. Such laser types include neodymium-doped yttrium, aluminum and garnet laser (Nd:YAG; 1064nm), erbium/chromiumdoped yttrium, scandium, gallium and garnet laser (Er,Cr:YSGG ;2790 nm), carbon dioxide laser (CO₂; 10,600 nm) and diode laser (980 nm). Several studies on laser surgeries have documented excellent outcomes in terms of aesthesis. preservation of function and recurrence [17,18]. However, affordability and accessibility limit the use of laser in the surgical management of vascular lesions. In the index case, the scar was aesthetically satisfactory with preserved active function of the finger and at more than one year of follow up, no recurrence of the capillary heamangioma.

In a retrospective study of 1180 patients who had surgical excision of heamangiomas, the post operative complications were very minor and considerably low [19]. Thus surgical excision is the treatment of choice for recurrent heamangiomas or those refractory to non-operative therapy. Vascular recanalization is the reason for recurrence of the lesions following excision thus this has to be taken into consideration during excision. In the case being reported, the lesion involved the dorsal surface of the middle phalanx of the left ring (4th) finger. Thus the extent of excision was limited in order

to avoid neurovascular injury that might have resulted in significant iatrogenic functional compromise of the finger. Hence we had to administer pre-excisional low dose external beam radiation therapy locally to the lesion.

To the best of our knowledge this is the first reported case of use of pre-excisional external beam radiotherapy in the management of recurrent heamangioma and in view of the excellent outcome in terms of functional preservation, aesthesis and none recurrence; it is thus recommended in the management of heamangioma that is either refractory to drug therapy or recurs after initial excision.

4. CONCLUSION

Post traumatic heamangioma clinically presents as hypergranulated tissue and may pose a management challenge as it may recur after excision. Arising from the excellent outcome of the low dose external beam radiation followed by excision, we hereby propose that this should be adopted as one of the treatment options of heamangiomas especially in adults.

CONSENT

Prior to excision, informed consent was obtained from the patient. Also the consent of the patient was sought and obtained for the purpose of this publication.

ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

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COMPETING INTERESTS

Authors have declared that no competing interests exist.

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