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Case Report

Clinical challenges in confirming the differential diagnosis of suspected Dabska's tumour of the hand

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Abstract

There is always clinical situation where a suspected tumour by biopsy turns out to be an alternate pathological diagnosis when resected completely. In this case report, we present a case which was suspected to have Dabska's tumour of the thenar aspect of the right hand by the initial histopathological analysis. The tumour recurred and was referred to the plastic surgery department for wide local excision and soft-tissue reconstruction. Tumour was excised with adequate clearance, and the resultant soft-tissue defect was reconstructed with ipsilateral groin flap. The post-operative period was uneventful. We explain in this report, the approach made for safe resection of suspected Dabska's tumour and reconstruction of the defect with reduced morbidity as possible.

Keywords: Dabska's tumour, groin flap, verrucous tumour

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INTRODUCTION

Unusual malignant tumour often results in the biased clinical approach when it presents with recurrence. The review of the literature of an unusual tumour may mandate us to follow the oncological principle for the resection of the tumour, even though the final histopathology may vary in its presentation from the initial report. We present in this case report, the approach made for safe resection of suspected Dabska's tumour and reconstruction of the defect with reduced morbidity as possible.

CASE REPORT

A 17-year-old male presented with a history of a nodule-like growth <1 cm on right thenar eminence that appeared 1 year ago with rapid and progressive increase in size, especially after a subsequent trauma with the cricket bat. It was asymptomatic initially, later presented with minimal pain and numbness. The tumour was not associated with wasting of thenar muscles or restriction in the movements of the thumb. There was no history of weight loss, decreased appetite and no reported family history. An excision biopsy was done in the local hospital, before 1 month and histopathologist reported the tumour as Dabska's tumour. The patient was referred to the department of plastic surgery for further evaluation.

Physical examination revealed a hyperkeratotic ulcerated haemorrhagic nodule 2 cm × 2 cm at the thenar eminence of the right hand (Figure 1). Magnetic resonance angiogram

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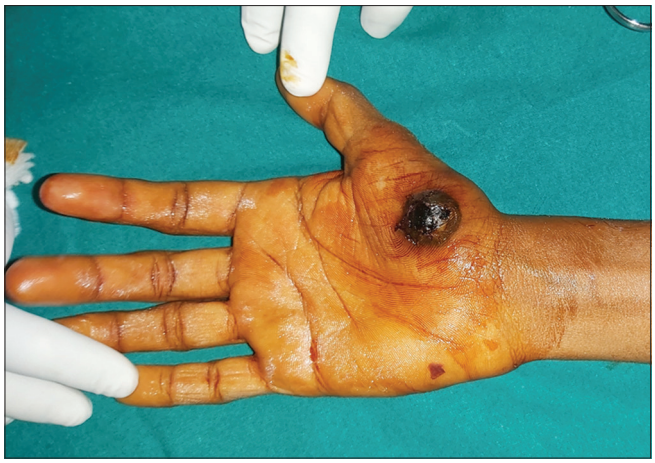

Figure 1: Clinical photograph showing hyperkeratotic ulcerated haemorrhagic nodule 2 cm × 2 cm at the thenar eminence of the right hand

was performed to rule out the vascular connection and to know the extent of the lesion. The tumour was supplied by vascular twigs from the radial artery and was above the fascia covering the thenar muscle without infiltration to the underlying thenar muscle. Wide local excision of the tumour with a clearance of 2 cm in all dimensions was done according to the oncological principles (Figure 2). Intraoperatively, tumour was infiltrating the dermis and subcutaneous tissue without involving radial neurovascular bundles of the right thumb (Figure 3). The

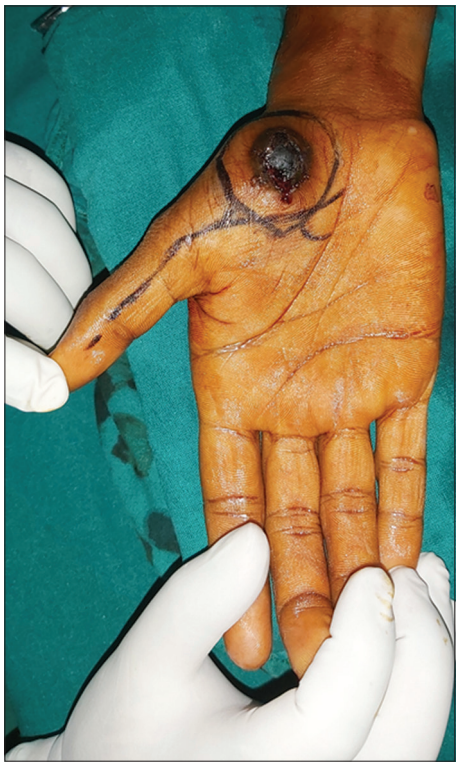

Figure 2: Clinical photograph showing wide local excision of the tumour with a clearance of 2 cm in all dimensions

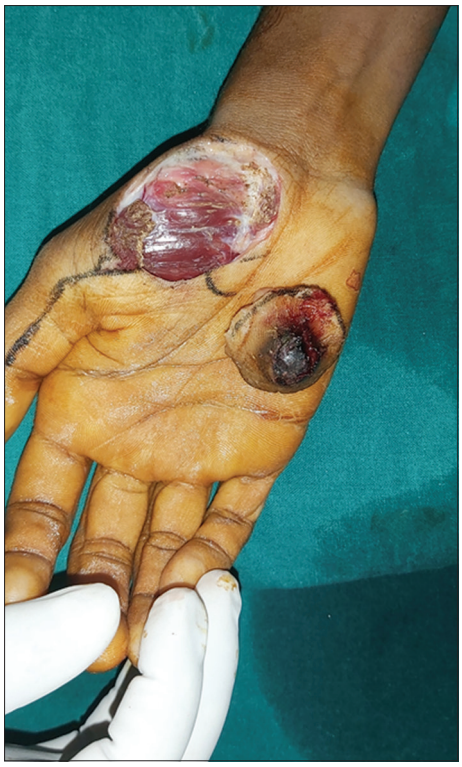

Figure 3: Clinical photograph showing tumour infiltrating the dermis and subcutaneous tissue without involving radial neurovascular bundles of the right thumb

base of the excised bed was marked with vascular clips in anticipation for future intervention with radiotherapy to the deeper tissue (Figure 4). The excised tissue was sent for the histopathological evaluation. The resultant soft-tissue defect was reconstructed with ipsilateral groin flap (Figure 5). Donor area for the flap was closed primarily (Figure 6). After 3 weeks, the flap was divided and inset completed (Figure 7). The post-operative period was uneventful.

Histological examination showed epidermis hyperkeratotic with papillomatosis with an underlying tumour invading papillary as well as reticular dermis, subcutaneous tissue composing of ectatic cavernous and capillary vessels lined by endothelial cells. All the resected margins are free from tumour, adjacent skin showing lymphoplasmacytic infiltration No characteristic hobnail lining cells (Figure 8). Combined clinical and histopathological findings determined the diagnosis as recurrent verrucous haemangioma, disproving Dabska's tumour.

DISCUSSION

Proper oncological principles are always helpful not only for proper diagnosis of tumour but also in preventing future recurrence of tumour. Recurrent tumour are clinically challenging as it requires proper planning for

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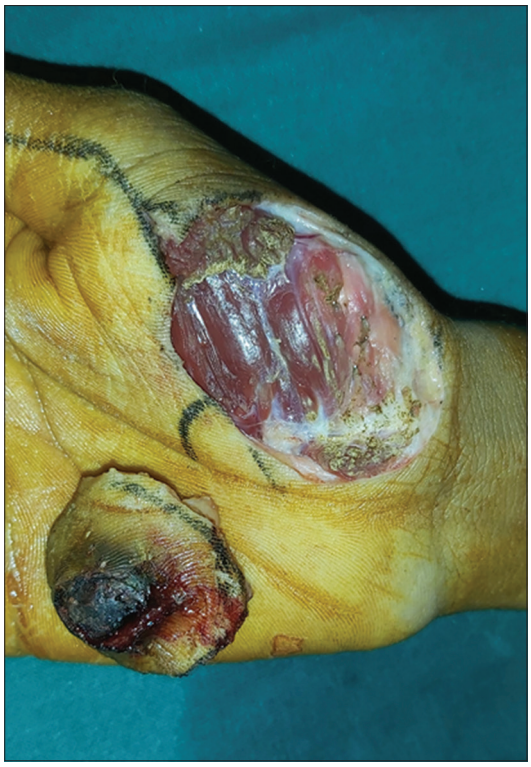

Figure 4: Clinical photograph showing base of the excised bed was marked with vascular clips in anticipation for future intervention with radiotherapy to the deeper tissue

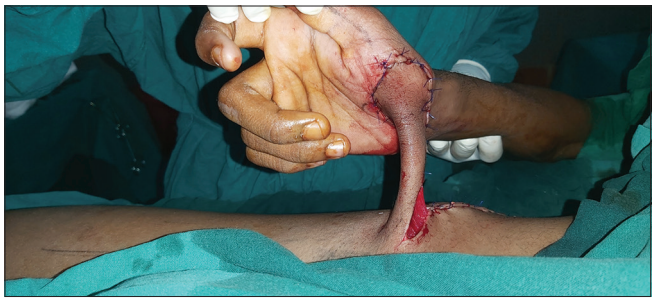

Figure 5: Clinical photograph showing soft-tissue defect reconstructed with ipsilateral groin flap

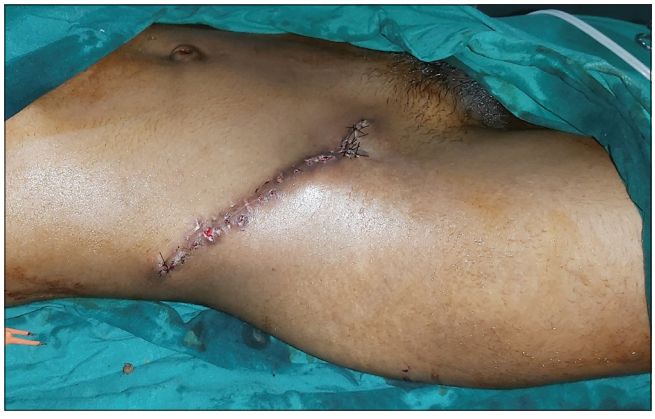

Figure 6: Clinical photograph showing donor area for the flap closed primarily

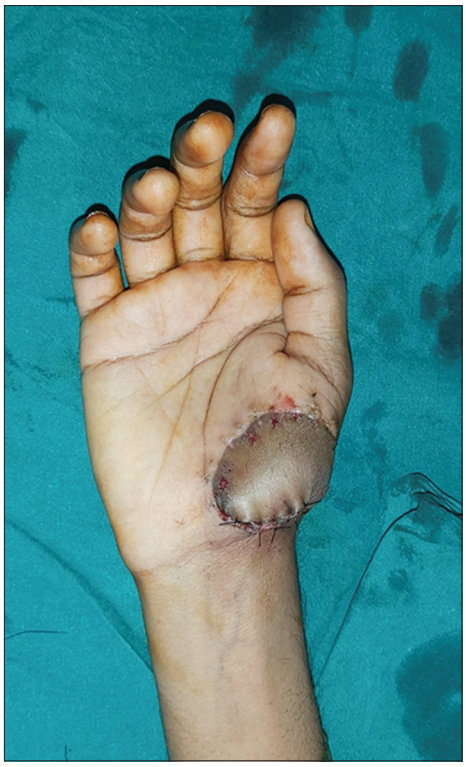

Figure 7: Clinical photograph showing flap divided and inset completed

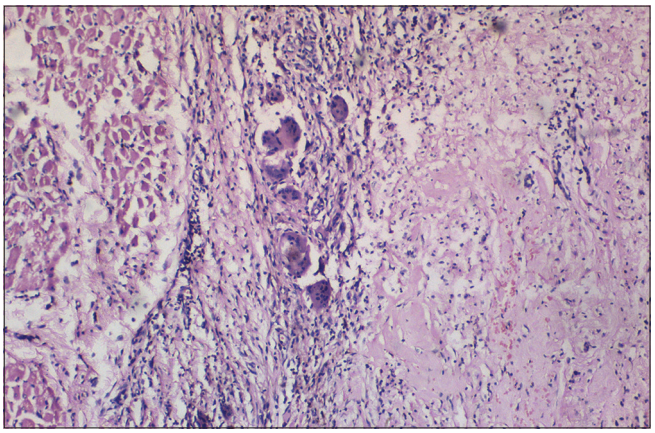

Figure 8: Histopathological findings determined the diagnosis as Verrucous haemangioma (Haematoxylin and eosin , X 400)

the resection of tumour to prevent further recurrence, adequate soft-tissue reconstruction and anticipation of adjuvant modalities. Planned operative measures should be taken in recurrent tumour as it creates immense psychological trauma for the patient subjected to recurrent operative procedure.

Dabska's tumour is an unusual malignant papillary intralymphatic angioendothelioma which often affects the skin and subcutaneous tissue in children.[1] Since its first description in 1969 by Maria Dabska,[1] only few cases have been histologically confirmed and described in literature. Verrucous haemangiomas are uncommon capillary or

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cavernous haemangiomas with growth and evolution similar to vascular malformations and immunologically similar to vascular neoplasm. Histopathology is crucial for differentiating from Dabska's tumour and Verrucous haemangiomas. Verrucous haemangioma extends deep into the dermis, reaching the subcutaneous tissue.[2] They can be present since childhood or appear later in life (during adulthood and are usually found on the lower extremities, being unilateral in approximately 95% cases). Initially, they may appear as a non-keratotic and verrucous in nature, especially after a subsequent trauma or infection.

Verrucous haemangiomas should be identified, diagnosed and treated as early as possible to limit the extent of resection. If the lesion is small, electrocautery/cryosurgery/laser treatment can be used, but resection is the definite treatment. Additional modalities may be used in adjuvant to resection for extensive lesions to further assist in reducing the risk of recurrence. The prognosis for verrucous haemangioma is good, with recurrence being low with adequate surgical margins. They are benign lesions and surveillance is mainly performed for local recurrence.

On the other hand, Dabska's tumour is a low-grade malignant endovascular papillary angioendothelioma, with <30 cases reported so far, first described in 1969. Dabska's tumour typically present as a violaceous, pink/bluish black tumour growing slowly within the dermis or subcutaneous tissue up to maximum 40 cm in diameter. Although most commonly affecting head and extremities, it reported from the palm, forearm, heel, knee, cheek and buttock. It results in the degeneration of its underlying vascular network. Histologically, the lesion resembles a cavernous lymphangioma at low power, with large dilated vascular channels and an infiltrate of small lymphocytes. The channels are typically lined by hobnail like/matchstick like appearance with apical nucleus. Unique for dabska tumour is a pattern of papillary structures, similar to renal glomeruli, with hyalinised collagenous cores. Currently, it is more often classified as a tumour of intermediate biologic malignancy. Two of dabska tumour were associated with metastasis to regional lymph nodes and one to lungs.[1-3]

Dabska's tumour has the potential to transform into angiosarcoma, hence makes it a tumour of intermediate malignancy. Hence, surgical clips have been used intraoperatively onto tumour bed after wide excision. Moreover, groin flap was considered for the repair of tumour site instead of split skin graft, anticipating radiation therapy for Dabska's tumour. The identification is essential to avoid the clinical and histological misdiagnosis and to ensure an adequate surgical excision to prevent recurrences.

Dabska's tumour is an exceedingly rare diagnosis, and it is usually confirmed by histopathology. It is important, from a prognostic and therapeutic point of view, to make a correct diagnosis between Dabska's tumour and other vascular lesions. This case is being reported to show the clinical significance of histopathological diagnosis and subsequent difference in severity grade of tumour, treatment and prognosis to have an overview on Dabska's tumour, although rare should not be forgotten.

Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understand that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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