Case Report

Complete Excision of Nerve Originating Hemangioma: A Case Report

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Abstract

The hemangioma is a hamartomatous malformation and not a true tumorous of the hemangioma occur during childhood. It affects three girls for every boy. Commonest site of involvement is skin. Nerve originating hemangioma refers when it arises from the vessels supplying the nerve (vasa nervosum). It is very rare condition and presents painful, soft, compressible swelling in the course of nerve. Microscopically, hemangioma consist of poorly demarcated, non-capsulated masses and leashes of vascular channels, most of which contain blood. Although diagnosis is clinical; FNAC, Doppler Ultrasonogram study, MRI and EMG all investigations are helpful for confirmation. Surgery is the main stay of treatment but recurrence may occur. Per-operative microsurgical facility and in recurrent cases, administration of corticosteroid can prevent recurrence. We have operated one case with our own circumstances and patient has got uneventful recovery in one year follow up.

Keywords: Hemangioma, Vasa nervosum, Recurrence, Corticosteroid

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Introduction

Hemangioma is the developmental malformation of blood vessels[1]. It is one of the most common benign tumors of infancy and occurs in approximately 5 to 10% of infants[2]. The word 'hemangioma' comes from the Greek haema- (αίμα), 'blood'; angeio (αγγείο), 'vessel'; -oma (-ωμα), 'tumor'[3]. In 1982, Mulliken and Glowacki proposed a new classification system for vascular anomalies which has been widely accepted and adopted by the International Society for the Study of Vascular Anomalies (ISSVA). Hemangioma may be capillary ('Port-wine' stain, Strawberry naevus and Salmon patch), Venous (cavernous), Arterial (Plexiform) in origin[4].

Approximately 80% of hemangiomas are single tumours and 20% occur in the multiple sites[5]. Pathologically it although of vascular origin, result from cellular proliferation and differ from malformations which are embryonic, developmental abnormalities representing errors in vascular morphogenesis[6]. Some of the hemangioma are involuting, a common rule is 50% of hemangioma regress by age 5 and 70% by age 7[7]. It is usually soft, subcutaneous, red or bluish in colour, compressible, sometimes painful. Diagnosis can be confirmed by colour doppler ultrasonogram, contrast CT, MRI and FNAC for tissue diagnosis[8]. Nerve originating hemangioma is a literary or theoretical term, practically it arises from vasa nervosum. Intraneural hemangioma of the median nerve is a rare condition and only few cases have been described in the literature. Due to mechanical compression carpal tunnel syndrome (CTS) is the main presenting feature. Raynaud's phenomenon may be an associated complaint[9]. Complications of hemangioma are included as ulceration, infection, hemorrhage, recurrence & malignant transformation (angiosarcoma, which is very rare)[10]. Hemangiomas that develop lesions or sores may require treatment. Treatment options include the corticosteroid medication, laser treatment, medicated gel and surgical removal[11].

Here we presented a case of intraneural hemangioma of the median nerve of a 16-years of age male which removed surgically by combined interfasicular and epineural resection under regional anaesthesia, no recurrence observed during the one year of postoperative follow-up period.

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Case report

A 16-years of age male worker presented to our outpatient clinic with painful longitudinal swelling in the palmar surface of the right hand for three years duration; associated with tingling and numbness in the thumb, index, middle finger and radial half of the ring fingers, difficulty in gripping, boxing and opposition of thumb. There was no history of trauma and relevant medical condition.

Physical examination revealed a tender, soft elongated mass, 18×3×2 cm in dimension in the palmar aspect of the right hand. Tinel sign was positive.

Radiographic examination revealed no bony lesion. Ultrasonographic examination done to exclude any vascular lesion of the radial artery, revealed non pulsatile, cystic mass with color signal. FNAC remarked the lesion as hemangioma.

MRI and EMG examination was planned for this patient, but the party refused due to their financial constrain.

After preoperative assessment, the patient was admitted for surgical treatment under the diagnosis of hemangioma and before surgery informed written consent was taken.

The operation was done under supraclavicular block, using a pneumatic tourniquet. Exploration revealed a yellowish-brown soft tissue mass with areas of hemorrhage and dimensions of approximately distal forearm to root of index finger, originating from the palmar surface of the median nerve with intraneural extension and adhesions to the surrounding tissues (Figure 1-4).

The mass was removed completely by interfasicular and epineural surgical resection technique, without structurally damaging the nerve fibers by using 4x loop. Wound was closed with subcuticular 3/0 vicryl and in skin with interrupted 4/0 prolene and a drain was kept in situ. Patient's Hand was elevated in cock up position and splinted by the anterior cast. Prophylactic and therapeutic antibiotic was used for 10 days. Stitch was removed on 14th POD and the wound was found healthy.

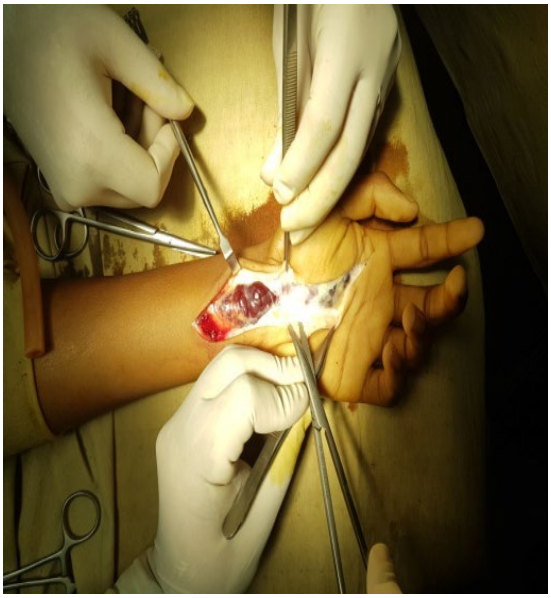

Figure-1: Per-operative view of nerve originating hemangioma case

In macroscopic view of the lesion, intraneural and fasicular involvement is obvious. Histopathologic and microscopic evaluation revealed dilated and congested vascular structures in a fibro-collagenous stroma with areas of bleedings, consistent with histopathologic findings of hemangioma (Figure-5). The symptoms were relieved in the first three weeks after the operation. On clinical and ultrasound examination, no recurrence was observed in the first one year following the operation.

Figure-2: Use of loupes to magnify the operative field of the hemangioma case

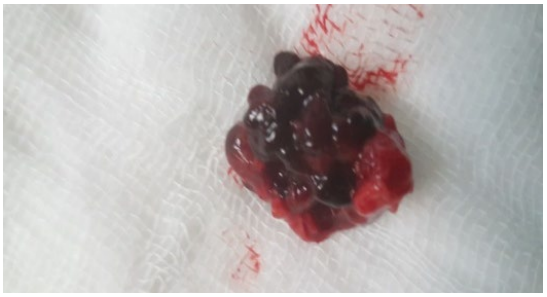

Figure-3:Piece of excised hemangioma

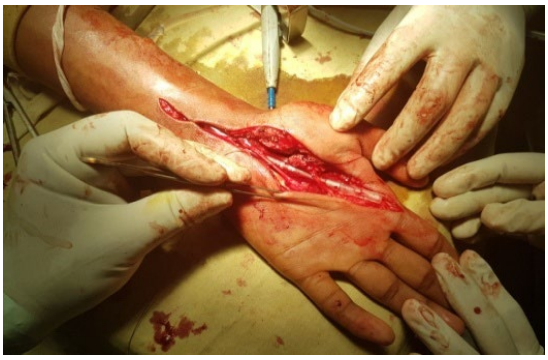

Figure-4: Wound closure with drain after the excision of hemangioma

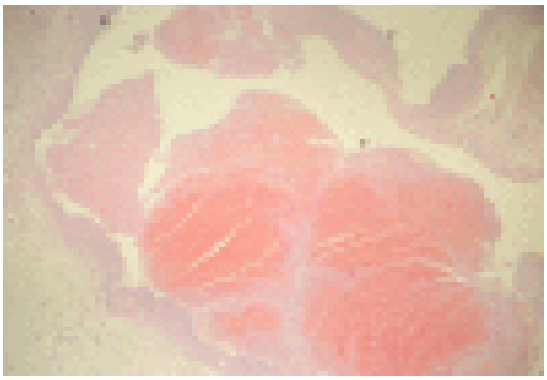

Figure-5: Histopathological slide of the nerve originating hemangioma case

Discussion

Benign intraneural hemangioma originating from peripheral nerves is rare. Most of the lesions are painful, soft mass along the distribution of a nerve with features of nerve compression and entrapment[12].

A thorough search through the literature revealed ten cases of hemangioma of the median nerve. In most of the cases, carpal tunnel syndromes are the presenting feature and in one case Raynaud's phenomenon was an associated presenting feature[1].

The tumor may not be easily recognized until it becomes symptomatic and it is rarely diagnosed before surgery. In the differential diagnosis, lipoma, lipofibroma, hamartoma and intraneuronal Schwannoma must be considered[11].

Ultrasonography may give useful information about the nerve's dynamic relation to the surrounding musculotendinous structures and nerve conduction studies may reveal nonspecific features of compressive neuropathies. For appropriate planning of surgical therapy and preoperative diagnosis, MRI is essential and gives useful information regarding tumor location, size, extent and relationship of peripheral nerve[13].

Hemangioma shows a hyperintense signal on T1 and T2 weighted images with fat suppression sequences. Flow voids are usually apparent and feeding vessels may be visualized; these lesions are also noted to enhance after Gd-addition. On angiography an early and persistent tumoral blush is demonstrated[14,15].

No certain protocol has been established to manage this difficult condition, however conservative treatment usually fails and surgery is the treatment of choice. When possible total resection of intraneural hemangiomas is curative, partial resection may relieve symptoms but recurrence may occur which may require en-bloc nerve resection and repair with nerve graft[14].

We couldn't convince the patient for MRI and EMG due to lack of cooperation and financial constraints. The longest period of follow-up without recurrence has been reported by Oztekin et al.[13]. They reported a case of CTS due to a cavernous hemangioma of the median nerve, which was successfully removed by epineural resection and no recurrence was observed over a 6-year follow-up period. Patel et al.[13] reported two cases of hemangioma of the median nerve which they treated by partial excision and resulted in recurrence in the third year, one of the recurred cases managed by resection of median nerve and nerve grafting without recurrence, four years after the surgery.

Chatillon et al.[12] reported the first case of using radiotherapy in the treatment of intraneural hemangioma. Preoperative embolization and postoperative radiotherapy combined with partial resection were beneficial in a case of intraneural hemangioma involving inferior trunk of brachial plexus and resulted in symptomatic relief and radiologic shrinkage in the size of the mass seen on serial follow-up MRI images, with a follow-up period of two years.

Microsurgical dissection and resection should be decided at the time of surgery and careful preoperative planning using MRI and if needed angiography is essential for cystic lesions of the palmar side of wrist. Excision of the affected nerve and grafting should be the last choice and should only be used in complicated cases and when there are frequent recurrences.

Some author suggests intralesional corticosteroid as triamcinolone acetonate in recurrence after dissection shows curative in long term follow up[6].

We couldn't adopt microsurgical technique due to lack of facility. In spite of that we have used magnifying loupes for better visualization during dissection.

In our case, total resection of the hemangioma was achieved by combined epineural resection and interfasicular macroscopic resection technique, no neurologic complications observed postoperatively and no recurrence observed in the one-year follow-up period of the case.

Conclusion

Nerve originating hemangioma in younger age group has a diagnostic difficulty, so before confirmation differential diagnosis should be kept in mind. Complete resection of the tumor with the help of microsurgical technique is the only hope of cure. Recurrence may be a problem, but can be managed with corticosteroid.

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