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Letter to the Editor

Wide resection as a solution to excruciating pain in intraneural hemangioma: Follow-up of a previously published case report

Hemangiomas are rare benign vascular tumors that typically occur as a purplish or reddish slightly elevated area of skin. They mainly occur in the central nervous system but rarely in the liver, retina, and the skin [1,2]. Intraneural hemangioma is a very rare benign mesodermal tumor. It can cause severe pain, neuropathy, and paresthesia due to sudden nerve compression. Treatment options are limited, and surgical excision seems to be the best option in the treatment of intraneural hemangiomas [3–5]. This case report was prepared in accordance with the SCARE guidelines [6].

In a previously published case report [7], we discussed the case of a 6-year-old boy with a two-year history of progressive left forearm pain with swelling and paresthesia on the ulnar-side fingers. Imaging studies and pathological findings strongly favored the presence of a hemangioma in the vicinity of the ulnar nerve. The patient underwent surgery for tumor excision which sacrificed part of the ulnar nerve. Nerve transfer was performed as described by Mackinnon et al. [8]. Two months later, the patient began to experience excruciating pain in the limb, which caused him to assume a bizarre limb posture resulting in an inferior dislocation of the shoulder.

Under general anesthesia, we performed closed reduction to correct the malposition.

In response to the onset of pain, a nerve block technique was performed for about a year to control his continuous pain. The treatment was initially successful in reducing the patient's pain. However, as time passed, the time to pain onset decreased until the block no longer had any effect, resulting in a bizarre position for him. Despite continuous nerve blocking, limb amputation was still considered the alternative option for treatment.

As the patient went on to experience excruciating pain, we decided to perform a second surgery in order to widely resect the tumor with free margins. A volar extensile approach was used to expose the ulnar nerve and the presumed location of the tumor [9]. The ulnar nerve was entirely surrounded by a discrete, dark red, sponge-like subcapsular mass which could evidently be due to tumor recurrence (Figs. 1–4). Wide resection of the tumor was performed along with the involved portion of the ulnar nerve from the elbow to the medial side of the hand (Guyon's canal). Despite sacrificing the ulnar nerve, his Mackinnon's nerve transferred was functional and prevented severe motor problems [8].

Following surgery, the patient had a one-day hospital stay. A long-arm splint was utilized for three weeks. Following this surgery, the patient showed a dramatic response in terms of pain and bizarre postures. For three weeks, the range of motion exercises gradually started (Figs. 5–8). Our follow-up period was one year and three months. The latest update on the relevant reports in the literature is provided in Table 1.

In conclusion, intraneural hemangiomas may produce intense pain at times, and the only alternative to amputation is excision of the entire nerve, which can be an effective treatment.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

This study was approved by the ethical committee of the Tehran University of medical sciences.

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Ramin Zargarbashi: Conceptualization, Data gathering, Project Administration, Performing the surgery
Fardis Vosoughi: Manuscript preparation, Review & editing
Nesa Milan: Investigation, Visualization, Manuscript preparation – Original draft.

Declaration of competing interest

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Fig. 1. Intra neural hemangioma.

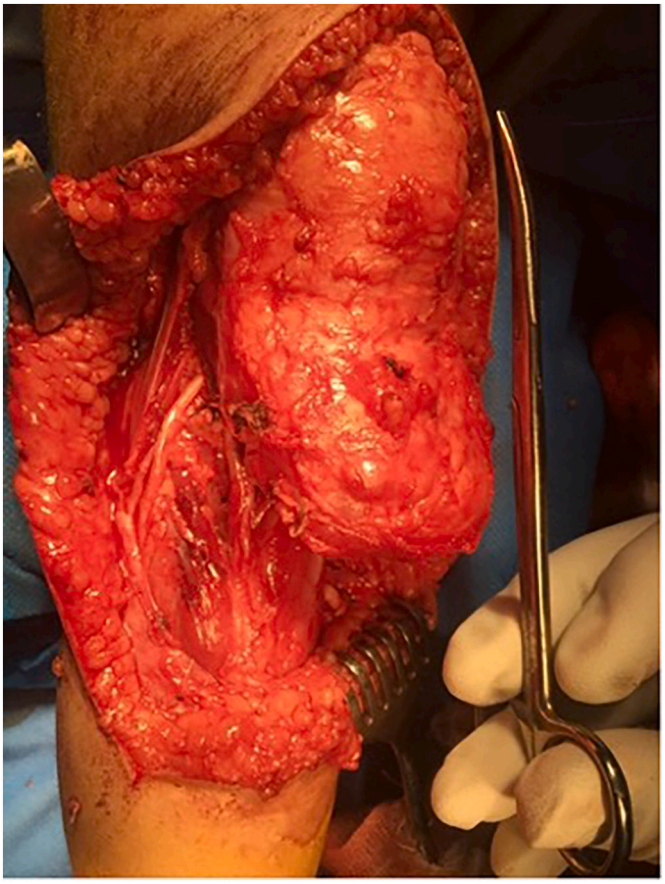

Fig. 2. Intra neural hemangioma.

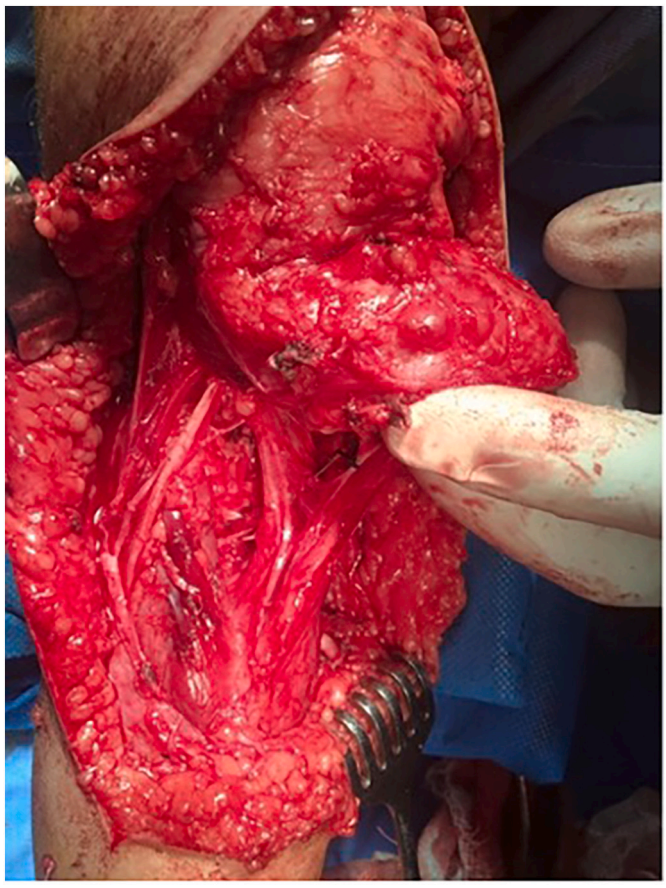

Fig. 3. Intra neural hemangioma.

Fig. 4. Intra neural hemangioma.

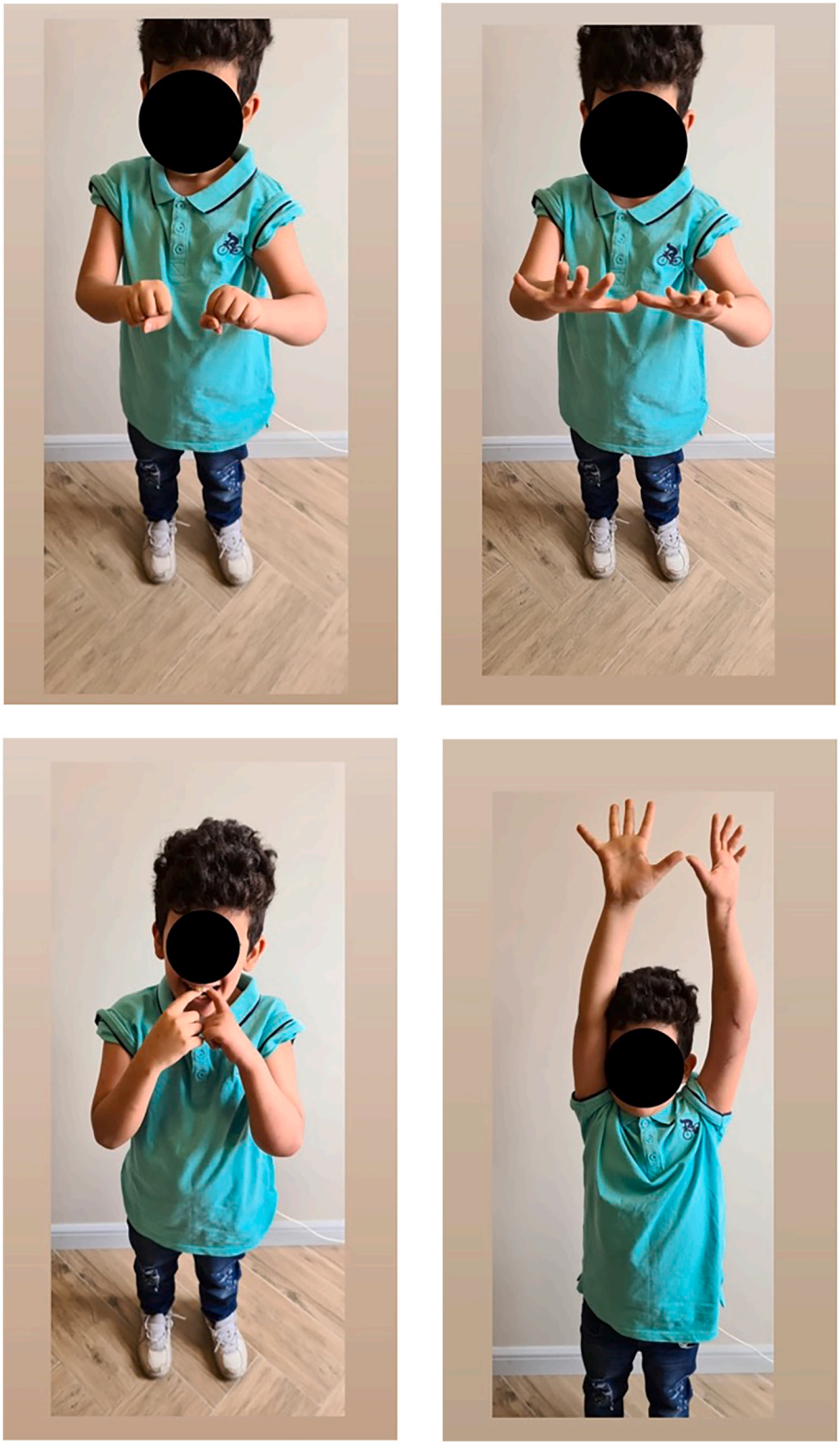

Fig. 5, 6, 7, 8. Patient's range of motion post-operation.

Table 1
Related cases of intraneural hemangioma.

Authors	Location	Treatment	Follow up
Düzgün et al. [10]	Radial digital nerve	Surgical excision	Responded completely to the treatment-No recurrence
Kim et al. [11]	Ulnar nerve	Surgical excision	Responded completely to the treatment-No recurrence
Ravanbod et al. [12]	Ulnar nerve	Surgical excision	Limited function-No recurrence
Châtillon et al. [3]	Inferior trunk-adherent to adjacent nervous and vascular structures.	50 % of tumor with Surgical excision + radiotherapy	The patient was asymptomatic and the results of the motor and sensory examinations were unremarkable.-No recurrence
Pulidori et al. [13]	Intramuscular mass close to but not directly involving the ulnar nerve	Surgical excision	Responded completely to the treatment-No recurrence
Doğramacı et al. [14]	Median nerve	Surgical excision	Responded completely to the treatment-No recurrence
Bilge et al. [15]	Peroneal nerve	Surgical excision	Responded completely to the treatment–No recurrence
Bacigaluppi et al. [16]	Lateral sensory branch of the radial nerve	Surgical excision	Responded completely to the treatment–No recurrence
Kwong et al. [17]	Tibial nerve	Surgical excision	Not mentioned
Papagelopoulos et al. [18]	Median nerve	Surgical excision	Responded completely to the treatment-No recurrence
Al-Garnawee and Najjar [19]	Median nerve	Surgical excision	Responded completely to the treatment-No recurrence

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