Case Report

Schwannoma of the digital nerve and reconstruction with reverse-flow dorsal metacarpal artery flap: A case report

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1. Introduction

Schwannomas, also known as neurilemmomas, are tumors of Schwann cells on peripheral nerves. They are quite rare in adult population.¹ Schwannomas are commonly known as solitary masses. But multiple schwannomas may be encountered connected to some certain diseases like schwannomatosis or neurofibromatosis type 2.

Reverse flow dorsal metacarpal artery (DMCA) flap is a fasciocutaneous flap, planned on the perforator arteries located at the second web area of dorsal aspect of the hand.

A case of 65 year-old male patient with a digital nerve schwannoma was presented.

2. Case report

A 65 year-old male patient presented with a large, outgrowing mass on the palmar aspect of third metacarpophalangeal joint (Fig. 1). The mass was first detected on the surface of the palm two years ago and began to grow gradually for the past two years. The

patient does not remember a trauma or an accidental damage to skin continuity.

On the physical examination, 67 × 42 × 37 mm (length, width, height) sized, firm and minimally tender and a heterogeneously appearing mass was revealed. Tinel's sign was noted positive. Ultrasound revealed a hypoechoic encapsulated solid mass but could not detect a connection with any important adjacent neural, bony, tendinous or vascular anatomic structure. Ganglion, neurofibroma, vascular origin sarcomas were among preoperative differential diagnoses and a specimen for tru-cut biopsy was sent. But the diagnosis cannot be provided by tru-cut biopsy.

Despite the fact that superficial location and outgrowing structure of the mass, an magnetic resonance imaging (MRI) was also performed and the mass appeared as hypointense on T1-weighted MRI sequences and hyperintense on T2-weighted MRI sequences (Fig. 2). The patient was decided to be operated after MRI.

Patient was operated under axillary anesthesia. During operation, the mass was well-circumscribed, encapsulated, soft nodule and measured as 65 × 40 × 35 mm. The cut surface was brown-yellow in color with large cystic areas. Nodule was attached to ulnar digital nerve of third digit just distal to bifurcation and sent for pathologic examination. Microscopically, the tumor was composed of biphasic spindle-shaped cells with compact hypercellular Antoni A areas and myxoid hypocellular Antoni B areas. There were foamy macrophages and stromal hemosiderin. Areas of necrosis were not observed. Mitotic figures were very rare. Immunohistochemically, tumor was diffusely positive with S100 and vimentin. Further, desmin, SMA, CD117 and CD34 were negative. But, there were few CD34 positive cells in the pericapsular region. Ki-67 proliferation index was below 3%. With these findings, the case was diagnosed as Schwannoma (Fig. 5).

Preserving the skin envelope superficial to the mass was not possible perioperatively due to fragility of skin covering the mass, possibly related to relatively fast expansion of the skin connected to growth speed of the mass. Thus, the mass was excised en-block with the skin overlying and leaving a 35 × 40 mm skin defect at the palmar aspect of third metacarpophalangeal (MP) joint (Fig. 3). To cover the joint and digital neurovascular bundle with a stable

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Fig. 1. Preoperative photos of patient. A: Oblique, B: Top, C: Side.

Fig. 2. MRI views of the mass. A: T-1 weighted sequence, B: T-2 weighted sequence, C: T-2 weighted sequence.

tissue, choice of skin grafts was withdrawn keeping in mind that the defect was located on the dominant hand of the patient. Thus, a reverse flow DMCA fasciocutaneous flap on third metacarpal area was planned and the presence of perforator arteries were confirmed with a sterile-probed hand Doppler intraoperatively.

The flap was planned 1.5 cm larger than the defect size, elevated at a plane above the paratenon of extensor tendons, on the dorsal metacarpal artery. The pedicle was ligated distal to the flap tissue and dorsal metacarpal artery was not skeletonized during flap harvesting to avoid damage. After flap dissection from

proximal to distal direction, perforator arteries proximal to MP were preserved. To avoid venous congestion at postoperative period, more than two superficial veins were included underneath the flap tissue.

After rotation of flap on the pedicle around the pivot point, flap tissue reached the defect site easily and was sutured with 5/0 non-absorbable nylon sutures. The donor site of flap was closed primarily due to increased laxity of dorsal skin, probably related to advanced age.

The operation was finished uneventfully. The mass was sent to pathology department to histologic examination.

No paresthesia/hypoesthesia was encountered during early postoperative period based on two-point discrimination test, which was three mm. No complications were encountered in 6-months postoperative follow-up period (Fig. 4).

3. Discussion

Schwannomas, also known as neurilemmomas, are tumors of Schwann cells on peripheral nerves. Usually being encapsulated and benign in nature,¹ they rarely show malign transformation.^{2,3} A case with congenital aggressive schwannoma of a newborn, causing death due to diffuse metastasis at 5 months old was reported in literature in 1964.⁴ Capsule of schwannoma consists of perineurium and deepest layers of epineurium⁵ which permits surgeons to resect tumor without damaging the nerve fibers. Not commonly seen, plexiform subtype of schwannomas may invade neural bundles, making excision more difficult than expected.⁶

Although neurilemmomas account for the most common tumor of peripheral nerves, they are responsible for 5% incidence in adult

Fig. 3. Perioperative photo of patient.

Fig. 4. Six months postoperative photo. A: Dorsal metacarpal artery flap tissue, B: Flap donor site.

population,¹ generally at ages between 20 and 50 years and most commonly in the head and neck region followed by flexor surfaces of upper and lower limbs.⁷ Patients in pediatric age population,^{1,7} and patients with atypical presentation regions such as stomach,⁸ pleura,⁹ pudental nerve in the pelvis,¹⁰ even at a bronchogenic cyst's wall¹¹ were also reported.

Schwannomas are commonly known as solitary masses, multiple schwannomas can also be possible to be diagnosed in some circumstances like schwannomatosis, neurofibromatosis type 1 and type 2.⁵ Responsible genes are SMARCB1, NF-1 and NF-2 for schwannomatosis, neurofibromatosis type 1 and neurofibromatosis type 2, respectively.^{12,13} After the diagnosis of schwannoma in our patient, a whole body scan with the probability of presence of other schwannomas was not needed but the patient was informed with the consent that if any other schwannoma is diagnosed in future, he should be checked about schwannomatosis or neurofibromatosis despite the advanced age of patient.

Despite many physical examination findings can be faced with schwannoma, a positive Tinel's sign is an important finding due to nerve compression and they appear as a hypoechoic encapsulated mass on ultrasound.¹ Continuity with a nerve on ultrasound is a positive finding for a schwannoma helps radiologists to distinguish from a neurofibroma.¹⁴ Ultrasound was not helpful to show connection with the digital nerve tissue in our patient. So, we suggest that absence of ultrasound features showing connection to nerve tissues should not exclude schwannoma from differential diagnosis in a suspected case.

A classic image for a schwannoma on T1-weighted MRI sequences is an isointense or mildly hypointense solid mass, whereas they are seen as marked hyperintense masses on T2-weighted MRI sequences.¹⁵

Diffuse and strong immunostaining with surface markers S-100 and Vimentin (Fig. 5) is an important diagnostic tool for schwannomas.¹⁶ The most characteristic microscopic finding for those tumors are Antoni A zone, with interstitial substance around spindle cells in a palisade formation and Antoni B zone, composing of irregular cells and myxoid component.¹⁷ These two zones can both be seen in the same tumor.¹⁸

Immunostaining against nuclear antigen Ki67 can be useful tool to detect a schwannoma with a malignant transformation potential where Ki67 index is found 5–65% by Kindblom et al with potential to malignant transformation in neural sheath tumors⁵ and Ki-67 index if found below 5% without a malignant transformation.¹⁹

Neurofibromas are among differential diagnosis of schwannomas. Unlike schwannomas, neurofibromas originate from various phenotypes of cells, may infiltrate nerve fascicles and excision may result in nerve deficits.¹ Strong and diffuse immunostaining of schwannomas with s-100 antigen is useful in differential diagnosis with neurofibromas and Antoni B zones may express CD34 antigen.²⁰ Immunohistochemically, the tumor of our case was diffusely positive with S100 and vimentin. Further, desmin, SMA, CD117 and CD34 were negative. But, there were few CD34 positive cells in the pericapsular region.

Skin of intermetacarpal areas at dorsal aspect of hand have been used as flaps since 1990.^{21–23} They have a versatile use, reaching palmar aspect of the hand. Pivot point of reverse DMCA flap is at 1 cm from the margin of relevant web space.²⁴ Two perforators are located proximal and distal to MP joint.²⁴

After flap elevation and full mobilization, the tourniquet should be deflated and flap should be returned back to its donor site for a time of 1–3 min to let small-caliber choke vessels to open.²⁴ The

Fig. 5. Histologically the lesion was composed of spindle spindle-shaped cells (a. Hematoxylin & Eosin staining, original magnifications $\times 100$). Immunohistochemical examination revealed that the tumor cells were diffusely positive with vimentin (b) and S-100 (c). Desmin (d) and CD34 (e) immunostaining were negative. Ki-67 proliferation index was below 3% (f) (original magnifications $\times 100$).

returning of flap tissue back to its donor bed just after the elevation was also performed by us, we did not face with arterial perfusion problems but a distal-third venous congestion occurred 4–5 h after finishing the operation. We used two medical leeches without losing time and flap recovered with just a minor epithelial loss at portion and re-epithelized spontaneously without skin grafting.

No major complications encountered on flap tissue and donor site at 6-month postoperative period (Fig. 4).

In conclusion, a 65-year old man with a giant schwannoma on the palmar aspect of the hand and successful reconstruction with a reverse DMCA flap was presented.

Conflicts of interest

All authors have none to declare.

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