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Delayed motor weakness following peripheral nerve schwannoma resection: illustrative cases

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BACKGROUND Delayed facial palsy (DFP) after vestibular schwannoma resection is a well-documented, yet poorly understood condition. The exact pathophysiological mechanisms of DFP are unknown, although diminished intraoperative nerve response has been shown to be a prognostic factor. To date, no such condition has been described in regard to peripheral nerve schwannomas.

OBSERVATIONS Here the authors present the first reported cases of delayed motor weakness (DMW) after peripheral schwannoma resection of the ulnar nerve at the elbow and peroneal nerve in the popliteal fossa. Both patients presented with a mass lesion and radiating paresthesias and had normal motor function preoperatively. Immediately after surgical resection, the patients had full strength. Within 24 hours, both patients exhibited marked weakness that gradually resolved over the course of several weeks.

LESSONS DMW after peripheral schwannoma resection is a rare condition likely akin to delayed facial nerve palsy after VS resection. The mechanism of this phenomenon remains unknown, although symptoms appear to self-resolve with time. A better understanding of the processes driving this condition may allow for therapies that can expedite and improve long-term outcomes.

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KEYWORDS facial nerve; vestibular schwannoma; peroneal; ulnar; peripheral nerve

Similar to the goals of vestibular schwannoma (VS) resection, the aims of surgery in peripheral nerve schwannomas are to achieve complete tumor resection and preserve neurological function.[1,2] In VS surgery, this means hearing and facial nerve preservation whenever possible.[3] Delayed facial palsy (DFP) is seen in a small subset of patients who undergo VS surgery. This phenomenon is generally defined as a decline in facial nerve function from the immediate postoperative period and can occur any time from 1 day to several weeks postoperatively.[4] Most patients tend to self-resolve without further intervention, although the extent of recovery may vary.[5] The exact pathophysiological mechanisms of DFP are unknown, although diminished intraoperative nerve response has been shown to be a prognostic factor.[6]

To date, no such condition has been described in regard to peripheral nerve schwannomas. Herein, we detail two cases of delayed motor weakness (DMW) after peripheral schwannoma resection of the ulnar and peroneal nerves, which was documented by serial examinations in the first 24 hours by the senior author. Both cases had normal motor examinations in the recovery room and within 6 hours of surgery and displayed new weakness by the first postoperative morning.

Illustrative Cases

Case 1

A 33-year-old right-handed female presented to our clinic with a 6-month history of progressive numbness and pain in her right fourth and fifth digits. Around the same time as symptom onset, she noticed a small mass just above the ulnar groove that had grown in size over the last several months. The patient was otherwise healthy and denied any trauma to the elbow or history of neurofibromatosis. Physical examination revealed an approximately 2 cm mass just above the cubital tunnel. The mass was locally tender and caused distal paresthesias on palpation. No motor deficits were noted. Magnetic resonance imaging (MRI) revealed a 1 × 2 cm tumor of the ulnar nerve at the elbow suggestive of a benign nerve

ABBREVIATIONS DFP = delayed facial palsy; DMW = delayed motor weakness; MRI = magnetic resonance imaging; VS = vestibular schwannoma.
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sheath tumor (Fig. 1A and B). Due to the progressive nature of her symptoms and growth of the lesion, resection of the mass was deemed appropriate. Given the need to mobilize the nerve intraoperatively, a nerve transposition was planned.

An 8-cm curvilinear incision was made centered over the posteromedial elbow. The ulnar nerve was identified. Proximal and distal control of the ulnar nerve relative to the tumor was achieved. The spindle-shaped nerve sheath tumor was identified, and a fascicular sparing approach was used to resect the tumor (Fig. 1C–F). There were no myotonic discharges using electromyographic monitoring. The pathology was consistent with a schwannoma. A subcutaneous nerve transposition was then performed.

Postoperatively, the patient did well and had normal motor and sensory examinations documented serially by the senior author. Overnight, the patient developed moderate to severe weakness in the ulnar nerve distribution. There was no swelling at the incision site. By 1 month, the patient's strength had improved to near baseline. At 2-year follow-up, the patient's motor weakness had fully resolved, with only some slight paresthesias on occasion. MRI showed no residual or recurrent growth.

Case 2

A 56-year-old female presented to our clinic for a recently discovered peroneal nerve tumor. The patient noted a mass on her right popliteal fossa several years ago which was originally thought to be a Baker's cyst. Recently, the patient was experiencing worsening pain in the dorsal aspect of her foot which prompted further workup, revealing a 7-cm neurogenic tumor just distal to the sciatic bifurcation (Fig. 2A and B).

Through an 8-cm incision over the popliteal fossa, the distal sciatic nerve and its branches (common peroneal, tibial and sural nerves) were identified proximal and distal to the large tumor.

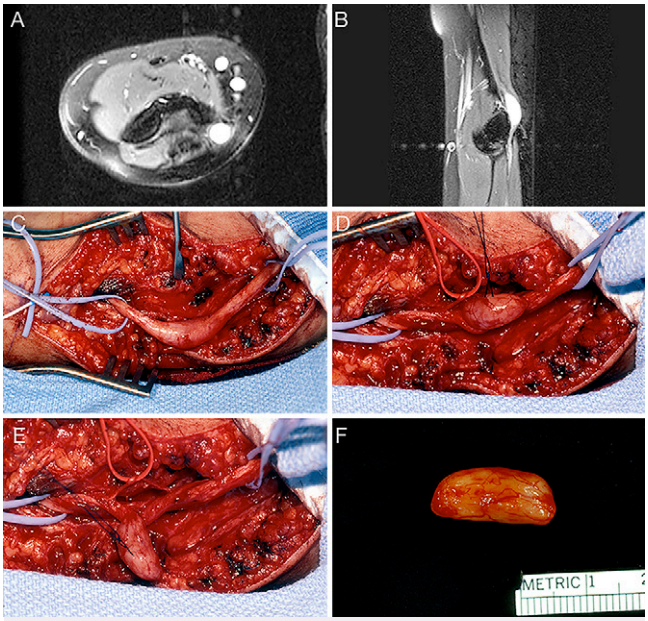

FIG. 1. Case 1. Axial (**A**) and sagittal (**B**) fast spin echo (FSE) magnetic resonance images showing the ulnar nerve schwannoma in the cubital tunnel region. Intraoperative photographs demonstrating the intraoperative dissection (**C**–**E**) and resected specimen (**F**).

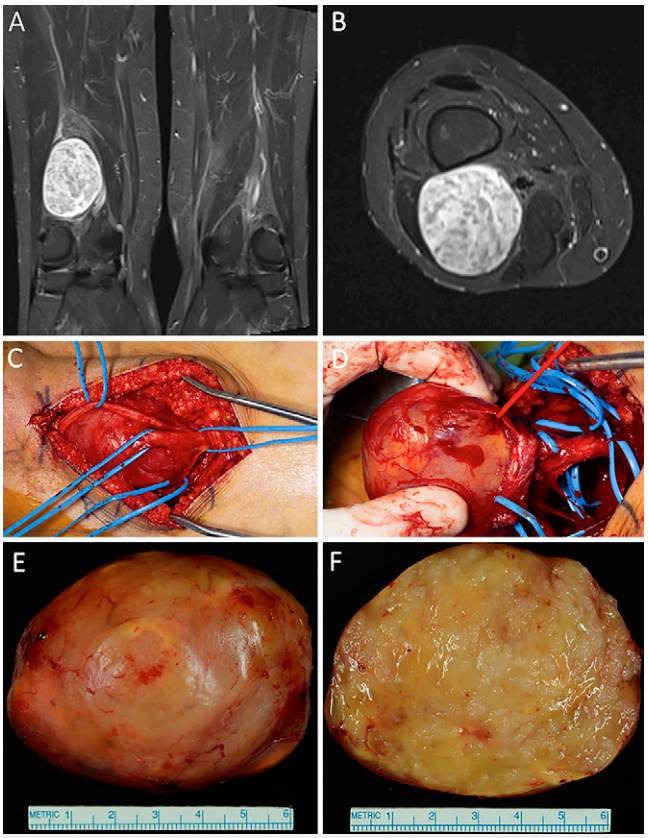

FIG. 2. Case 2. Coronal (**A**) and axial (**B**) short tau inversion recovery (STIR) magnetic resonance images showing the common peroneal nerve schwannoma in the distal thigh. Intraoperative photograph of the common peroneal nerve schwannoma prior to dissection (**C**). With mobilization of the tumor, the entering fascicle is identified (red vasoloop, **D**). Pathology specimen of gross tumor (**E**) and cut in half (**F**), measuring approximately 7 cm in diameter.

Circumferential dissection and mapping of the tumor was performed visually and with a disposable nerve stimulator, and a longitudinal epineurotomy was performed. A single entering fascicle was seen, which allowed mobilization of uninvolved en passant fascicles. The entering fascicle was divided, and the tumor was removed (Fig. 2C–F). After the tumor resection, stimulation of the common peroneal nerve proximally showed good responses in the tibialis anterior and peroneus longus muscles.

In the recovery room and several hours postoperatively, the patient had full ankle eversion and dorsiflexion but mild sensory loss along the lateral shin documented by the senior author. The next morning, the patient's ankle eversion and toe extension remained at full strength, but her ankle dorsiflexion weakened to 1+. The incision site had no signs of abnormal swelling. At 1-month follow-up the patient reported and demonstrated strong foot and ankle dorsiflexion and was able to ambulate several miles.

Discussion

Observations

To our knowledge, these are the first reported cases of DMW after peripheral schwannoma resection. Of note, these two cases occurred 21 years apart, indicating the rarity of this condition and

were culled from the senior author's surgical experience of more than 1,000 benign nerve sheath tumors. Although there are no studies on DMW after peripheral nerve schwannoma resection, data can be gleaned from its counterpart, DFP after VS surgery.

Lessons

The incidence of DFP has been estimated to be between 5% to 25% with an average time of onset of 8.1 days.[4,6–9] DFP is more likely to occur in patients who have larger VSs, lower intraoperative facial nerve stimulus responses, and lower rates of gross total resection.[9] Some studies have shown the translabyrinthine approach to have a higher incidence of DFP (15.3%) compared to the overall incidence, while the retrosigmoid approach has a lower incidence (3.7%).[9] Most patients tend to have an excellent recovery (House-Brackman of 1 or 2) though a minority of patients have permanent moderate dysfunction (House-Brackman of 3).[9] Tawfik et al.[4] analyzed 291 patients who underwent VS surgery at their institution. They found that immediate facial palsy occurred in 21% (61) of patients whereas DFP occurred in 38% (112) of patients. On univariate analysis, DFP was found to be associated with a smaller tumor size and better long-term facial nerve outcomes ($p = 0.0101$). Multivariate analysis showed that longer time to onset of palsy predicted more favorable facial nerve outcomes.

Numerous mechanisms have been proposed over the years to explain DFP including postoperative inflammation, vasospasm, and viral reactivation. Given the time course and self-resolving nature of DFP, postoperative edema is the most widely accepted theory. A study performed on canines by Sekiya et al.[10] showed that facial nerve injury likely occurred in the days after surgery as the facial nerve is progressively compressed within the facial canal. If this is the case, steroids or simple observation may be the best approach for both DFP and DMW. In opposition, Scheller et al.[11] purport that vasospasm is the underlying cause of DFP. At their institution, 78 of 264 patients (29.5%) who underwent VS surgery received vasoactive treatment for hearing preservation. After termination of a 10-day treatment course, 7 patients (9%) developed DFP within 2 to 5 days. As this is a purely observational account, it is difficult to assume causation. If future studies corroborate this mechanism, and it can similarly be replicated in DMW, an argument could be made for prophylactic nimodipine after peripheral schwannoma resection. Finally numerous studies and case reports have given credence to the viral reactivation theory.[12–14] One of the earliest reported cases of this by Gianoli and Kartush[12] presented a patient who underwent resection of a VS and went on to develop DFP with concurrent vesicles in the ear canal. Serology confirmed herpes zoster oticus. A follow-up study by Gianoli prospectively analyzed the viral titers pre- and postoperatively of patients undergoing VS surgery. Of 20 patients who underwent surgery, seven developed DFP. Immunoglobulin M titers for herpes zoster 1 and 2 as well as varicella zoster increased by 92%, 70%, and 495%, respectively. Patients who did not develop DFP showed a modest increase ($<10\%$) or decrease in these titers. These data support the use of prophylactic antiviral therapy for VS surgery.

We are unsure as to the mechanism of delayed nerve injury in our peripheral nerve tumor practice, but speculate a similar mechanism to VS. Intellectually, we believe fascicular swelling as the most likely, but have not seen this phenomenon in patients who underwent extensive intraneural dissection as part of other procedures. Postoperative swelling in a tunnel could have been implicated, but the first case was transposed; the second one was several centimeters proximal to the fibular tunnel, which was not decompressed to avoid lengthier dissection. No compressive mass was evident on clinical examination and both patients were extremely thin. Steroids were considered but were not administered. Given the prevalence of DFP in VS, we are surprised that similar deficits do not occur more commonly after tumor resection in the periphery.

DMW after peripheral schwannoma resection is a rare condition likely akin to delayed facial nerve palsy after VS resection. The mechanism of this phenomenon remains unknown, although symptoms appear to self-resolve with time. A better understanding of the processes driving this condition may allow for therapies that can expedite and improve long-term outcomes.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Spinner. Acquisition of data: Spinner. Analysis and interpretation of data: Spinner. Drafting the article: both authors. Critically revising the article: both authors. Reviewed submitted version of manuscript: both authors. Approved the final version of the manuscript on behalf of both authors: Spinner. Administrative/technical/material support: Spinner. Study supervision: Spinner.

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