Successful Treatment of a Painful Neuroma Using Fascicular Shifting in the Ulnar Nerve: A Case Report

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Abstract

Objective   We report the case of a 40-year-old man with an inveterate ulnar nerve neuroma following a laceration injury of his left wrist twenty-three years ago. The patient presented with a typical ulnar claw-hand deformity and debilitating neuropathic pain in his hand (VAS 8.4). Pre-operative imaging revealed a neuroma of the ulnar nerve at the Guyon's canal. Moreover, a complete atrophy of all intrinsic hand muscles innervated by the ulnar nerve was present.

Methods   A Zancolli lasso procedure was performed to reduce the clawing effect. The neuroma was resected producing a nerve defect of five centimeters. Since the injury dated back more than two decades and any motor recovery was deemed impossible at that point, the motor fascicle of the ulnar nerve, i.e. the deep branch, was selectively neurolysed and harvested as an autologous nerve graft. Then the graft was shifted into the defect to be coapted with the superficial branch fascicle in an end-to-end fashion.

Results   The presented fascicular shift procedure resulted in satisfying and sustained pain reduction. At the six-month follow-up, the VAS decreased to 1.2, and two years post-operatively, the patient reported 2.5 on the VAS.

Conclusion   The fascicular shift procedure offers an alternative approach to conventional nerve grafts. If nerve grafting is required, using a locally harvested graft avoids additional donor site morbidity. Assuming the clinical scenario allows for fascicular grafting, we strongly suggest considering the fascicular shift procedure as a cost-effective alternative to expensive conduits and processed nerve allografts in sensory nerve reconstruction.

Keywords
► neuropathic pain
► neuroma treatment
► nerve reconstruction

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When a peripheral nerve is subject to traumatic injury such as transection, the formation of a bulbous swelling at the proximal end of the nerve, referred to as a neuroma, is almost invariably observed.[1] In an effort to restore neural continuity, axons of a severed nerve regenerate without fascicular organization.[2,3] Additionally, a mass of connective tissue proliferates around the proximal nerve stump.[4,5] In some cases, neuromas induce severe neuropathic pain, for which several neuropathological processes are thought responsible.[6–8] Even light touch or pressure on the neuroma may elicit tremendous pain in the associated innervation area due to increased axonal excitability.[9] Pain may also occur at rest as well as during night time due to spontaneous ectopic neuronal discharges.[10,11]

To prevent the unorganized axonal sprouting in neuroma formation, a great number of surgical techniques have been proposed.[2,3,12–14] Most of them include resection of the neuroma and translocation of the proximal nerve stump to an extra-anatomical target such as muscle,[15–17] bone,[18] or vein.[19,20] After surgical neuroma resection, the use of autologous nerve grafting,[21] nerve transfers,[22,23] nerve conduits,[24] and processed nerve allografts[5,25,26] has also been described. Studies of the various surgical approaches are limited by small sample sizes and nonrandomized case series study designs.[27] Furthermore, none of the techniques described so far provided predictable and durable pain relief.[28] Finally, the best treatment of any neural division is providing appropriate end organs to allow regeneration of the nerve and restoration of function.

In the context of peripheral nerve reconstruction, a technique called "fascicular shift procedure" (FSP) has recently been described.[29] This approach involves harvesting a fascicle of the target nerve and shifting it into the defect to overcome the peripheral nerve injury. It is coapted in an end-to-end fashion to both proximal and distal nerve ends, thereby bridging the defect. Hader et al[29] demonstrated that the FSP facilitated successful nerve regeneration in an experimental rat model. The local harvesting of the transplant avoids affecting another healthy nerve and helps to prevent extending the lesion. Hence, the risk of additional neuroma formation is reduced.

This report describes the clinical application of the FSP in the treatment of an inveterate ulnar nerve neuroma at the Guyon's canal. To the best of our knowledge, no similar case of neuroma treatment has been reported.

Case Report

Patient History and Clinical Examination

In 2017, a 40-year-old male patient, who had suffered a laceration injury of his left wrist 23 years ago in Poland, presented to our clinic. Primary care back then included wound treatment and skin closure without revision of neurovascular structures. The patient's main purpose of consultation was severe and disabling pain in his left hand, which impaired hand function in daily life and at work. At the initial consultation, the pain in the hand averaged visual analog scale (VAS) 8.4. Regular pain medication included gabapentin 300 mg five times daily. The patient's night's rest was sufficiently compromised by the persistent neuropathic pain.

Moreover, typical clinical signs of long-standing ulnar denervation were evident. There was a typical ulnar claw-hand deformity of the fourth and fifth finger. Abduction of the fifth finger and adduction of the fingers two to five was rated M1 (according to the British Medical Research Council scale) with a positive Froment's sign. Complete atrophy of the hypothenar eminence and intrinsic hand musculature was present. Sensation was completely absent in the fifth finger as well as the ulnar half of the fourth finger and also mildly reduced in the hypothenar region. However, the patient's subjective hand function was predominantly limited due to the persistent and tremendous pain.

Initial Imaging

Ultrasound of the patient's left wrist revealed an ulnar nerve's neuroma at the Guyon's canal, which measured 2.4 cm (►Fig. 1). Additionally, complete atrophy with fatty degeneration of all interossei, the hypothenar and 4th lumbrical muscles were evident. No further structures were found altered.

Magnetic resonance imaging was performed to evaluate the flexor tendons of the fourth and fifth fingers as well as their annular pulleys A2 and A4. Since these structures were found intact, a Zancolli lasso procedure was planned.
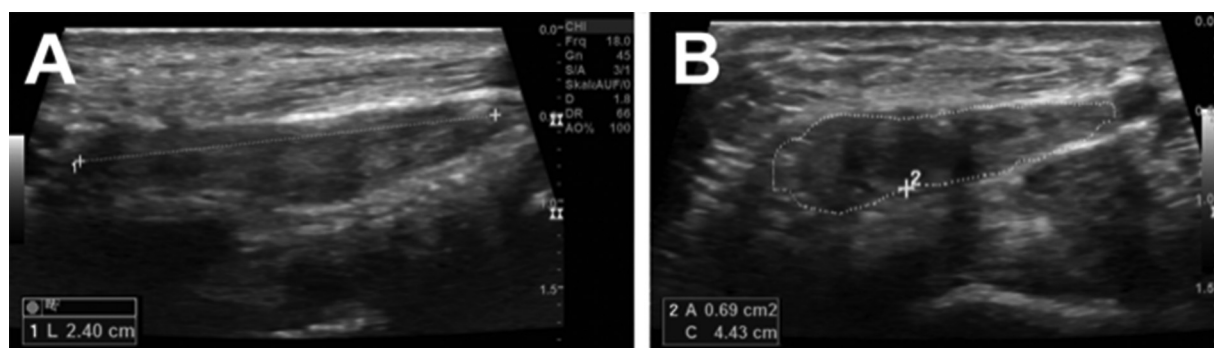

Fig. 1 High-resolution sonography of the ulnar nerve proximal to Guyon's Canal. (**A**) Here, a 2.4 cm long neuroma-in-continuity was identified. (**B**) Especially note the complete disruption of normal fascicular architecture in this transverse orientated sonography.
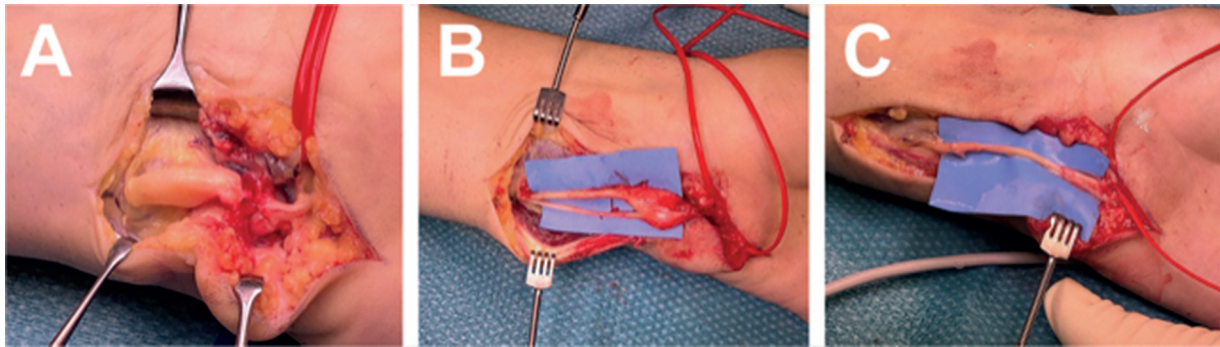
Fig. 2 Surgical neuroma resection and fascicular shift procedure. (**A**) Dissection of the neuroma. (**B**) Separation of motor and sensory fascicles extending proximally from the neuroma. (**C**) Shift of the proximally harvested motor fascicle to overcome the large nerve defect after neuroma resection.

Surgical Treatment

Within 3 months preoperatively, the patient was urged to regularly visit an occupational therapist to improve passive motion of the fourth and fifth finger. Consecutively, it was feasible to perform the planned Zancolli lasso procedure in a standard manner (for a detailed description of the surgical technique, see Goldfarb et al[30]).

To treat the painful neuroma, a novel surgical technique was applied.[29] First, the neuroma was fully exposed using a 10-cm skin incision over the volar, ulnar-sided wrist. From the Guyon's canal, the enlarged, neuromatous swelling extended proximally for 5 cm (►**Fig. 2**), which exceeded the reported length of the preoperative ultrasound images. Upon careful dissection of the neuroma, two main fascicles were identified. Due to the location within the Guyon's canal, they were recognized as the ulnar nerve's deep and superficial branch. Following neuroma resection, the two fascicles could be easily separated proximal to the injury over a length of 5 cm, which corresponded to the created nerve defect. The motor fascicle was harvested, and its ends were coapted proximally to the sensory portion of the ulnar nerve and distally to the superficial branch.

Postoperative histological analysis of the resected bulbous swelling revealed typical disorganized fascicular proliferation of small fascicles including edematous nerve sheaths corresponding to a posttraumatic neuroma.

Postoperative Results

The patient was examined 2 weeks, 3 months, 6 months, and 2 years after surgery. Neuroma pain in the patient's hand, as assessed with the VAS, decreased from 8.4 preoperatively to 1.2 at 6-month follow-up, and to 2.5 2 years postoperatively (see ►**Fig. 3**). The two-point discrimination for both right and left hands is schematically shown in ►**Fig. 4** at the 2-year follow-up. While sensation in the fifth finger did not recover, the patient developed protective sensation with a two-point discrimination of 12 mm on the ulnar side of the fourth finger.

Overall, hand function was satisfactorily restored by the Zancolli lasso procedure. The subjective disability evaluated by the Disabilities of the Arm, Shoulder, and Hand (DASH) questionnaire improved from 48.3 preoperatively to 28.3 at 6 months postoperatively.
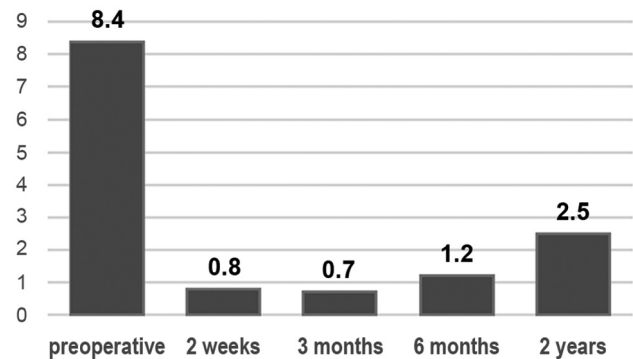

Fig. 3 Visual analog scale score measuring neuropathic pain in the hand over the course of 2 years. The initial pain reduction might be attributed to the neuroma resection per se. However, the patient also reported no substantial increase in pain at the 2-year follow-up.

Discussion

Today, it is well known that the proximal nerve end will try to reconnect with its distal stump or target organ after traumatic transection.[28] In 1890, Ramon y Cajal was the first to describe that regenerating axons from the proximal nerve stump form a growth cone.[31] If the outgrowing axons fail to find their distal endoneurial tubes, they will grow into surrounding scar tissue and form a neuroma.[32]

To date, the exact mechanism of neuroma-associated pain is not yet fully understood, and the question why some patients develop painful neuromas and others do not remains unresolved.[14,33] It is, however, widely accepted that the prevention of neuroma formation is paramount to avoid subsequent neuropathic pain.[14] Therefore, various surgical techniques have been proposed during the past decades. They include translocation of the proximal nerve stump into muscle tissue,[15–17] into vein,[19,20] and even into bone.[18] Further, various authors have presented the technique of capping the nerve's end with different synthetic materials, such as adhesive tape,[34] and biomaterial resembling tubes or conduits.[35–37] Nerve capping is assumed to allow for epineurial healing of severed fascicles, lessening improperly and irregularly regenerating nerve fibers.[14] Surgical fascicle ligation has also been described in the treatment of neuroma prevention.[2,32] Nerve defects exceeding several centimeters, which are created upon
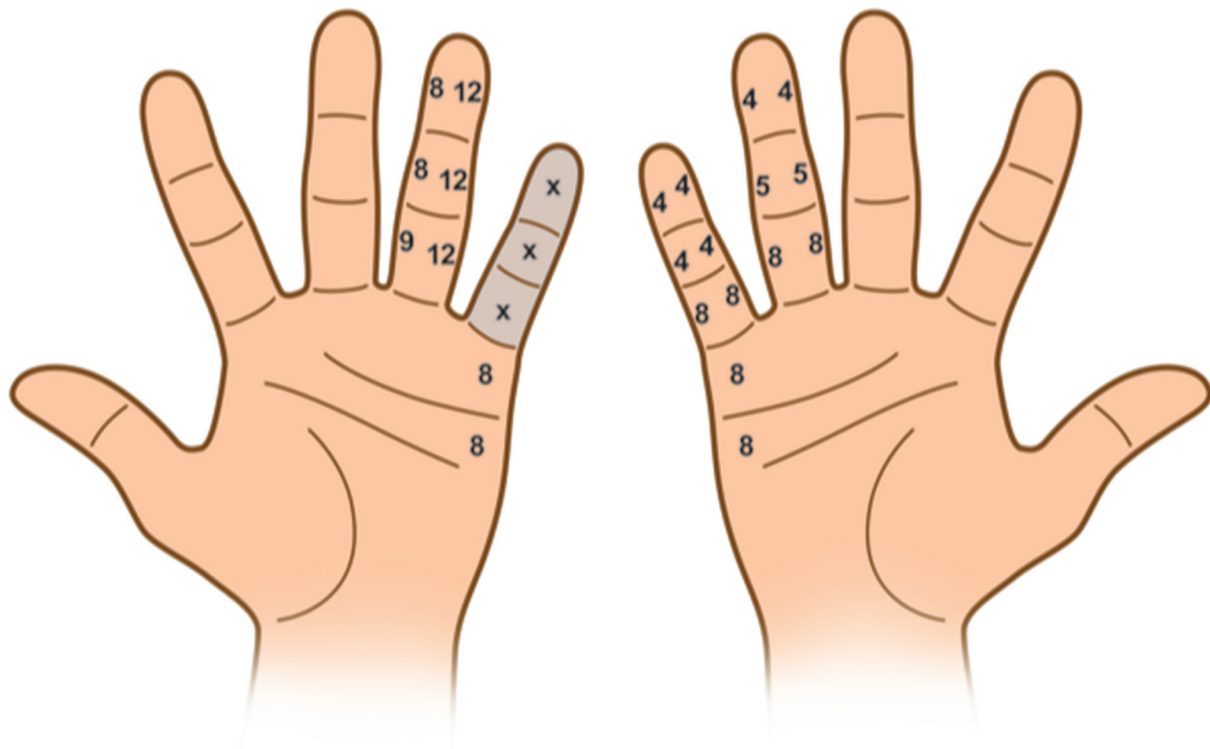
Fig. 4 Two-point discrimination in millimeter for both hands at the 2-year follow-up. The left image shows the operated left hand; the right image shows the patient's right hand. While sensibility in the fifth finger was not improved, protective sensation for the fourth finger could be restored, following nerve reconstruction with the fascicular shift procedure.

resection of large neuromas, may be reconstructed using conventional autologous nerve grafts,[21] nerve transfers,[22] nerve conduits,[23,24] and processed nerve allografts.[5,25,26] The latter two, in particular, have recently been increasingly used in sensory nerve defects.[24–26] However, due to the lack of adequate epitopes, nerve conduits are mainly limited to defects of 30 mm,[24] and processed nerve allografts have hardly been applied clinically in nerve defects extending 50 mm.[25,26] As neuroma treatment does not generally aim for functional restoration, the cost aspect of allografts and conduits also should be considered. Overall, insufficient pain relief and neuroma recurrence are still common findings,[28] keep challenging experts in the field, and novel alternatives are needed.

In the presented case, an inveterate, painful neuroma of the ulnar nerve was resected producing a nerve defect of 5 cm proximal to the Guyon's canal. Since the initial laceration injury dated back more than 23 years and had resulted in complete atrophy of all intrinsic hand muscles as well as an ulnar claw-hand deformity, no motor recovery was expected at the time of revision surgery. Therefore, an autologous donor nerve, which would have produced additional donor-site morbidity, was not considered for nerve reconstruction. Burying the proximal nerve stump into adjacent muscle or capping it with a vein could have resulted in the recurrence of a painful neuroma as reported in the literature.[15,19] Furthermore, this would have annihilated any chance of sensory recovery. On the other hand, two fascicles could readily be separated proximal to the resected neuroma. Hence, the senior author decided to reconstruct the nerve defect by locally harvesting the vital, though "functionless"

motor fascicle in the proximal segment of the ulnar nerve as a fresh autograft without causing any additional donor-site morbidity (see ►**Fig. 5** for schematic illustration).

The initial pain reduction from VAS 8.4 to 0.8 at 2-week follow-up probably originated from neuroma resection per se. However, at the final inspection 2 years postoperatively, the patient reported that he remained mainly pain-free (VAS 2–3) and merely experiences individual pain episodes following intense manual labor. Hence, we concluded that outgrowing axons were guided through the shifted fascicle toward their natural target. The recurrence of a neuroma was therefore prevented by the FSP. The partial defect elongation produced by the proximal fascicle harvest did not affect distal motor function since complete denervation had been present for over two decades. Although the DASH score aims to assess musculoskeletal disorders, it was used in this patient to illustrate the impact of this painful neuroma on hand function. The patient's DASH score slightly improved from 48.3 preoperatively to 28.3 at 6-month follow-up. This may be explained by the pain reduction, as it is commonly accepted that the painful neuroma itself can be more disabling to a patient than an anesthetic surface area or the loss of motor function it has caused.[3] Protective sensation for the fourth finger was restored. Overall, the patient was satisfied with the procedure, his final pain situation, and the partial regained sensibility.

Sir Sydney Sunderland, a pioneer in experimental nerve research, first described that the undamaged, intact perineurium may be seen as an impenetrable barrier for regenerating axons.[38] Thereby, the restoration of neural integrity and
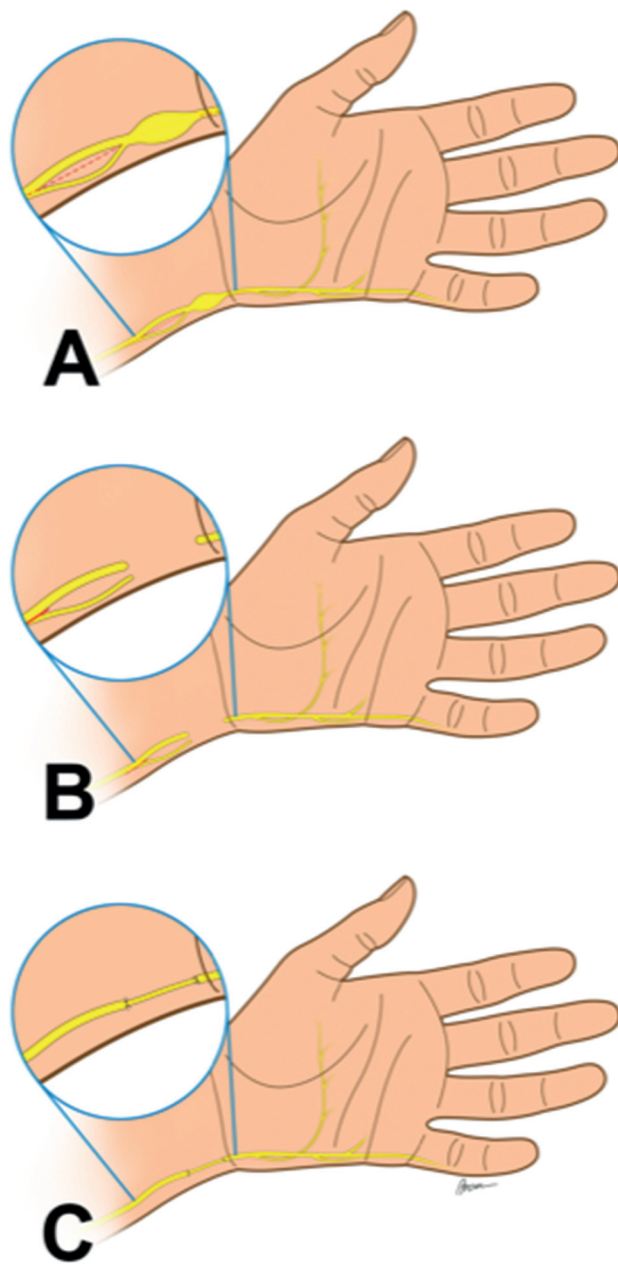
been mentioned in the literature and has hardly been applied.[41] Recently, Hader et al[29] reinvestigated a similar technique and could show that harvesting a fascicle distal to the injury site and shifting it into the defect led to successful nerve regeneration in a rat sciatic nerve injury model. At the time of initial description, the continuous blood supply to a large nerve graft was considered the main advantage of the discussed technique because it might affect nerve regeneration. Although we do not agree with this concept, we also see the true potential of fascicular shifting in large, proximal nerve defects, as adequate epitopes and humoral support, provided by the fascicular graft, are hypothesized to promote satisfying nerve regeneration.[29,39] Moreover, the fascicular structure can easily be identified in proximal nerve architecture and hence a fascicular graft may be conveniently harvested. In ongoing anatomical and clinical explorations, we are currently evaluating the applicability of the FSP in brachial plexus reconstructions and sciatic nerve lesions. Further indications include, like the present case, delayed repairs in which the donor-site morbidity of an autologous nerve graft is not justified, but some nerve regeneration is still possible. Koshima et al[41] also reported on successfully repairing one facial nerve and three digital nerve defects by using a fascicular shift.

From an anatomical perspective, the presented case raises the question of how far proximal the two branches of the distal ulnar nerve may be separated within their mutual epineurial sheath, that is, the deep motor branch and the superficial branch, which course apart in the Guyon's canal. Ongoing dissection studies in human cadavers and immunofluorescence analyses may soon deliver new insights on the intraneural topography of the distal ulnar nerve and might elucidate further fields of application.[42]

In conclusion, the FSP presents an alternative to conventional nerve grafts. Whenever nerve reconstruction is strived, local harvest of a graft avoids affecting another nerve and the appropriate nerve fiber composition is maintained. Presuming that the clinical scenario allows for fascicular grafting, the authors highly recommend considering the FSP in peripheral sensory nerve reconstruction as a cost-effective alternative to pricy conduits and processed nerve allografts.

Fig. 5 Schematic illustration of the performed nerve reconstruction using the fascicular shift procedure. (**A**) Extending distally from the neuroma within the Guyon's canal, the superficial and deep motor branches were identified as dividable fascicles. (**B**) The two fascicles were dissected within the neuroma and followed proximally. (**C**) The motor fascicle was shifted thereby bridging the nerve defect after neuroma resection.

continuity acts as a protecting sheath and counteracts the disorganized outgrowth of regenerating axons into scar tissue.[8,38] In the presented case, the shifted fascicle acted as a consistent structural matrix and signal source, shepherding axonal regeneration toward its natural target.[29] In contrast to nerve conduits and processed nerve grafts, the fascicular graft provides adequate epitopes and humoral support, which are responsible for axonal survival among others (see Bolívar et al[39] for a detailed review). The concept of proximal fascicular shifts has first been described by MacCarty[40] in 1951. Since then, this technique has rarely

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