Typical Brachial Neuritis (Parsonage-Turner Syndrome) With Hourglass-Like Constrictions in the Affected Nerves

Yong-wei Pan, MD, PhD, Shufeng Wang, MD, PhD, Guanglei Tian, MD, Chun Li, MD, Wen Tian, MD, Mengmeng Tian, MD, PhD

Purpose To report on 5 patients who had acute brachial neuritis (Parsonage-Turner syndrome) with hourglass-like constriction in the affected nerves.

Methods We retrospectively reviewed 5 patients who were treated in our department from December 2003 to December 2008. Acute, intense pain around the shoulder girdle and upper arm was the first symptom and was followed by muscle weakness and atrophy. Clinical and EMG examinations showed involvement of 2 or more nerves in the affected extremity. Those severely affected nerves that had no response to conservative treatment were explored, and an hourglass-like constriction was identified. Neurolysis was performed at the sites of constrictions in 2 radial nerves and 1 median nerve. The constricted portion was resected, and direct coaptation was performed in 1 radial nerve and 1 musculocutaneous nerve. The constricted portion was resected, and nerve graft was performed in 2 radial nerves and 1 median nerve.

Results All patients were followed up for 24 to 84 months after surgery. Of 3 nerves treated with external neurolysis, all attained full recovery. Of 2 nerves treated with resection and neurorrhaphy, 1 attained full recovery, and the other had an incomplete recovery. Of 3 nerves treated with resection and nerve graft, 1 (4-cm nerve graft) attained full recovery, and 2 (4-cm and 13-cm nerve graft, respectively) had incomplete recovery.

Conclusions The site of nerve lesion of brachial neuritis was not necessarily within the brachial plexus. Our finding of hourglass-like constrictions in individual peripheral nerves suggest that multifocal involvement of terminal branch lesions may underlie the complex patterns of paralysis often encountered clinically. (J Hand Surg 2011;36A:1197–1203. Copyright © 2011 by the American Society for Surgery of the Hand. All rights reserved.)

Type of study/level of evidence Prognostic IV.

Key words Brachial neuritis, hourglass-like constriction, neuralgic amyotrophy, Parsonage-Turner syndrome.

Acute brachial neuritis, also known as neuralgic amyotrophy and Parsonage-Turner syndrome, is an uncommon clinical problem. This abnormality typically has a characteristic presentation: acute onset of severe shoulder and/or arm pain, followed shortly thereafter by weakness and atrophy of muscles in the shoulder girdle and arm.1,2 Although the syndrome is now well recognized, the etiology remains
unknown, and the site of lesions is still debated. We have recently encountered 5 patients who had acute brachial neuritis. The patients were treated conservatively. Some involved nerves recovered gradually. However, palsy persisted in some severely affected nerves, signs of clinical recovery were absent, and EMG examination showed complete denervation of the muscles innervated by these nerves. To define the etiology, the nerves were explored, and an hourglass-like constriction without any external compression in the affected nerves was discovered. Our findings suggest that hourglass-like constrictive lesion in the nerve might be a pathologic basis of brachial neuritis.

### TABLE 1. Preoperative Status

<table>
<thead>
<tr>
<th>Case</th>
<th>Age</th>
<th>Gender</th>
<th>Side</th>
<th>Pain at Onset</th>
<th>Cause of Pain</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>21</td>
<td>F</td>
<td>L</td>
<td>Left shoulder and upper arm</td>
<td>None</td>
</tr>
<tr>
<td>2</td>
<td>26</td>
<td>M</td>
<td>L</td>
<td>Whole upper limb</td>
<td>None</td>
</tr>
<tr>
<td>3</td>
<td>45</td>
<td>F</td>
<td>R</td>
<td>Around the right shoulder girdle</td>
<td>None</td>
</tr>
<tr>
<td>4</td>
<td>29</td>
<td>M</td>
<td>L</td>
<td>Both shoulders</td>
<td>Flu-like symptoms</td>
</tr>
<tr>
<td>5</td>
<td>38</td>
<td>M</td>
<td>L</td>
<td>Left shoulder pain with radiation into lateral upper arm and radial aspect of forearm</td>
<td>Strenuous work</td>
</tr>
</tbody>
</table>

RN, radial nerve; MN, median nerve; MCN, musculocutaneous nerve; UN, ulnar nerve; AN, axillary nerve; SN, suprascapular nerve.

### TABLE 2. Finding at Surgery and Final Results

<table>
<thead>
<tr>
<th>Case</th>
<th>Periods from Onset to Surgery (mo)</th>
<th>Nerves Explored</th>
<th>Appearance of Nerve</th>
<th>Number of Constricted Lesions</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>4</td>
<td>Left MN (left RN was explored in another hospital)</td>
<td>Edematous and hardened</td>
<td>Many, along the nerve fascicles</td>
</tr>
<tr>
<td>2</td>
<td>4</td>
<td>Left RN, Left MN</td>
<td>Edematous and hardened</td>
<td>RN: 4, MN: 1</td>
</tr>
<tr>
<td>3</td>
<td>8</td>
<td>Right MCN, Right RN (RN had been previously explored in another hospital)</td>
<td>MCN: edematous and hardened, RN: severely scarred</td>
<td>MCN: 1</td>
</tr>
<tr>
<td>4</td>
<td>2</td>
<td>Left RN</td>
<td>Edematous and hardened</td>
<td>2</td>
</tr>
<tr>
<td>5</td>
<td>10</td>
<td>Left RN</td>
<td>Edematous and hardened</td>
<td>1</td>
</tr>
</tbody>
</table>

RN, radial nerve; MN, median nerve; MCN, musculocutaneous nerve.
MATERIALS AND METHODS
All patients were evaluated at our center between December 2003 and December 2008. The criteria for inclusion for each patient were a sudden onset of severe pain in or about the shoulder girdle, without a systemic disorder that might affect the musculoskeletal system, and an onset of weakness in the involved limb within a few weeks after the onset of the pain. Physical and/or EMG examination demonstrated findings that were consistent with brachial neuritis.

Laboratory investigations included white blood cell count with differential, blood urea nitrogen, liver function tests, and antinuclear antibody titer. All tests were normal unless specifically stated.
Needle EMG was performed in muscles representing all the suspected nerves that were involved. The distribution of neuropathic features was recorded.

A diagnosis of brachial neuritis was made in all patients. They were treated conservatively with vitamin B12, physiotherapy, and rehabilitative exercises. Some mildly involved nerves recovered gradually. However, in some severely affected nerves, palsy persisted, and there was no clinical or EMG sign of recovery after conservative treatment for 2 to 11 months. Surgical exploration was then performed.

On the basis of careful analysis of clinical findings and EMG examination, we found that neurologic deficits were more compatible with focal lesions within individual peripheral nerves rather than within the brachial plexus. Therefore, we decided to explore individual peripheral nerves. The types of treatment for hourglass-like constrictive lesion in the nerve were chosen on the basis of the degrees of constriction found at surgical exploration, the results of intraoperative nerve stimulation, and personal experience of the surgeon.

RESULTS

Clinical signs and symptoms of the 5 patients are shown in Table 1. All 5 patients had a typical presentation of sudden onset of shoulder pain, which subsided in 4 days to 4 months, followed by a flaccid paralysis of muscles in the shoulder girdle and arm.

Physical and EMG examination at the time of presentation revealed multiple nerve involvement. There were marked differences in grade of weakness and denervation between muscles innervated by different nerves. We found complete paralysis and denervation in some muscles and moderate to mild paralysis and denervation in muscles supplied by other nerves. Subclinical mild denervation was also detected in some muscles.

After conservative treatment for 2 to 11 months, 8 severely affected nerves (5 radial nerves, 2 median nerves, and 1 musculocutaneous nerve) had no clinical or EMG sign of recovery. We explored these nerves with the findings shown in Table 2. There were no constriction bands or muscles compressing the nerves. An hourglass-like constriction in the involved nerve was present (Figs. 1, 2A, 2B, 3A, 4A, 5A). The areas of stricture were not consistent with overlying muscle groups or sites of known compression. The nerves were found to be grossly edematous and thickened for a length of 3 cm to 5 cm beyond the constricitions. Slight adhesions were found around the constricitions.

Neurolyses were performed at the sites of constricitions in 2 radial nerves and 1 median nerve. The constricted portion was resected and the ends directly coapted in 1 radial nerve and 1 musculocutaneous nerve. The constricted portion was resected and nerve graft performed in 2 radial nerves and 1 median nerve. The resected portions were examined histologically; the results are shown in Table 2 and Figures 3B, 4B, and 5B.

All patients were followed up for 24 to 84 months after surgery; the final results are shown in Table 2. Of 3 nerves treated with external neurolysis, all obtained recovery of at least grade 4 muscle power in affected muscle groups using the Medical Research Council grading system. Of 5 nerves treated with resection and neurorrhaphy or nerve graft, 2 obtained full recovery, and 3 had incomplete recovery (grade 0–3). The shoulders of all patients had full movement, with M4–5 muscle strength of the shoulder girdle muscles. The recovery of sensation is shown in Table 2. The initial pain of all patients disappeared completely, except occasional, uncomfortable feeling in the periscapular area or upper arm.

DISCUSSION

Recently, intraoperative cases of hourglass-like nerve constriction lesions without any visible source of external compression have been reported. The involved nerves have included the main trunk of the radial nerve and posterior interosseous nerve, the main trunk of the median nerve and anterior interosseous nerve, the axillary nerve, and the suprascapular nerve. Most of these reported cases presented as an involve-
ment of an isolated peripheral nerve, and to the best of our knowledge, only 3 cases published in the English literature had multiple nerve involvement.\textsuperscript{9,14,15} It is well accepted that this phenomenon is not caused by external compression. However, the etiology of this phenomenon is still unclear.\textsuperscript{9,11}

In our series, all 5 patients satisfied the clinical and EMG criteria of brachial neuritis\textsuperscript{1,2} and were thought to be typical cases of brachial neuritis. Surgical exploration of severely involved nerves in all 5 cases showed hourglass-like constriction in the fascicular or main trunk of the nerve, similar to lesions that have been previously described. Therefore, we consider that this remarkable and unexpected nerve pathology could be the basis of lesions in brachial neuritis.

Brachial neuritis is a misleading name because the brachial plexus might not be the site of pathology. Detailed clinical and EMG examinations of patients often indicate lesions of individual nerves or nerve branches, many of which are not compatible with a brachial plexus localization.\textsuperscript{3,16–18} England et al\textsuperscript{3} believed that patients with neuralgic amyotrophy had a pattern of neurologic deficits that was most compatible with focal lesions within nerve fascicles or individual peripheral nerves, rather than within the brachial plexus. They were dependent on clinical and electrophysiologic analysis to locate the site of nerve lesion without surgical observations. The clinical presentation and course of our 5 cases were similar to those of England’s cases. Surgical exploration of severely affected nerves showed them to be swollen, and hourglass-like constrictions were evident. In case 3, we explored the brachial plexus, which was macroscopically normal, whereas we discovered a severe constric-
tion in the musculocutaneous nerve. Our finding suggested that the basic abnormality of brachial neuritis was this hourglass-like nerve constriction, and it provided evidence suggesting that multifocal involvement of terminal branch lesions might underlie the complex patterns of paralysis that are often encountered clinically.

The etiology of this condition remains unclear, and reports of pathological study are scant. Suarez et al\textsuperscript{19} reported brachial plexus biopsy findings from 4 patients with brachial plexus neuropathy. There were prominent collections of inflammatory cells within the brachial plexus. The authors therefore suggested that brachial neuropathies have an inflammatory–immune pathogenesis. However, our pathologic findings revealed that, though present, lymphocyte infiltration was limited. Suarez et al performed a brachial plexus biopsy from patients with brachial plexus neuropathy. However, many authors regard brachial neuritis as a form of mononeuropathy multiplex, involving individual nerves or nerve branches, rather than the brachial plexus.\textsuperscript{3,16}

Many cases might have been inappropriately diagnosed because biopsies might have been taken from an unaffected portion of the nerve.

Many authors believe that brachial neuritis is a self-limited disease, and nonsurgical treatment is the accepted treatment.\textsuperscript{1,2} However, recovery from this disorder can be quite protracted, and complete restoration of strength is not always achieved.\textsuperscript{1,20} van Alfen et al\textsuperscript{1} reported that the majority of patients in their 246 cases exhibited persisting functional deficits after an average follow-up of more than 6 years. In Misamore’s\textsuperscript{20} series, 3 of 7 patients had some residual deficit at long-term evaluation. The reasons for absent recovery remain

**FIGURE 4:** Finding in case 4.\textsuperscript{A} The radial nerve around the spiral groove appeared slightly edematous and hard. Epineurectomy revealed 2 severe hourglass-like constrictions 8 cm and 9 cm proximal to the lateral epicondyle.\textsuperscript{B} Pathologic examination of the specimen showed a slightly thickened perineurium, mild perivascular lymphoid cell infiltration, and central edema.

**FIGURE 5:** Finding in case 5.\textsuperscript{A} The radial nerve around the spiral groove appeared edematous and hard. A severe hourglass-like constriction was discovered 5 cm proximal to the lateral epicondyle.\textsuperscript{B} Histologic examination revealed fibrosis, edema, and scattered lymphocyte infiltration.
unknown. England\textsuperscript{16} speculated that patients with severe nerve injuries or involvement of nerves with long regenerative distance might have poor recovery. Our findings at surgical exploration confirmed the speculation of England. An hourglass-like constriction was discovered in the fasciculus or main trunk of the nerve, and some constricted lesions were so severe that the fascicles appeared to be completely ruptured. It is questionable whether nerve regeneration can be expected in such situations. We speculate that the same severely constricted lesion might have existed in van Alfen’s\textsuperscript{1} and Misamore’s\textsuperscript{20} patients who had poor recovery.

On the basis of poor clinical recovery in some cases reported in the literature and our own surgical findings, we consider that the strategy for treating brachial neuritis should be reconsidered. Surgical intervention should be carried out in cases that do not respond to conservative treatment after several months. Nagano\textsuperscript{11} recommended that exploration of the nerve be offered to patients who did not show any signs of recovery by 3 months after onset. He recommended only interfascicular neurolysis and believed that nerve grafting was unnecessary. He believed that more fibers would regenerate after neurolysis. In our present study, 3 of 8 nerves had incomplete recovery, and all the resection cases were the ones that did not regain function. Of them, 1 patient was treated with neurorrhaphy 11 months after the onset of the symptoms, 1 was treated with nerve graft 8 months after the onset of symptoms, and 1 was treated with a 13-cm nerve graft. We believe that these patients received treatment that was too late or too aggressive. Case 1 was a comparison of varying treatment methods at surgery. It seemed that the severity of constrictive lesions in both the left radial nerve and the median nerve was similar, so the radial nerve was treated with neurolysis, whereas the median nerve was treated with resection and a 13-cm nerve graft. The radial nerve had a full recovery, whereas the median nerve had a poor recovery. Therefore, we agreed with Nagano’s opinion that the period of “wait and see” should not exceed 3 months, and neurorrhaphy or nerve grafting should be avoided. However, we are reluctant to draw any firm conclusions on the basis of such a small number of cases studied retrospectively.

REFERENCES