



Postsurgical Parsonage-Turner Syndrome : A challenging diagnosis

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Parsonage-Turner syndrome (PTS) is a distinct clinical syndrome, characterized by acute and severe (mostly) unilateral shoulder pain, followed by paresis and atrophy of the shoulder girdle, while the pain decreases. Most authors consider it as an immune-mediated disorder. PTS is notoriously unrecognised and is usually diagnosed with delay. A PTS may also occur following a surgical procedure. Postsurgical PTS is an under-recognised and challenging clinical entity, as illustrated in the case reported here of a 59-year-old man, 4 weeks after anterior discectomy and fusion C5C7. In such cases, the differential diagnosis must be made with a complication of surgery, such as postoperative C5 palsy due for instance to a migrated bone graft. Arguments for PTS are : a certain delay between surgery and symptoms, intolerable pain followed by weakness and improvement of pain complaints, divergent distribution of weakness, sensory deficit and pain, which may be confirmed by electrodiagnosis. Early recognition of postsurgical PTS may avoid unnecessary investigations or surgical exploration. It allows to treat the patient properly, leading to greater satisfaction of both surgeon and patient ; pain management, physical therapy and reassurance are the cornerstones.

Keywords : postsurgical ; Parsonage-Turner syndrome ; neuralgic amyotrophy ; cervical decompression ; post-operative weakness ; C5 palsy.

INTRODUCTION

Parsonage-Turner syndrome (PTS), also known as neuralgic amyotrophy or brachial plexus neuritis, was first described in 1887 by Dreschfeld (7). Parsonage and Turner (23) went further into the subject in 1948, and from then on the condition was named after them. This distinct peripheral nerve disorder is characterized by acute and severe, mostly unilateral shoulder pain, which decreases after a few days, to be followed by muscle weakness and atrophy, slowly recovering after months to years, but not always completely. The syndrome may be bilateral (11). The upper trunk of the brachial plexus is mostly affected. Postsurgical PTS is diagnosed on

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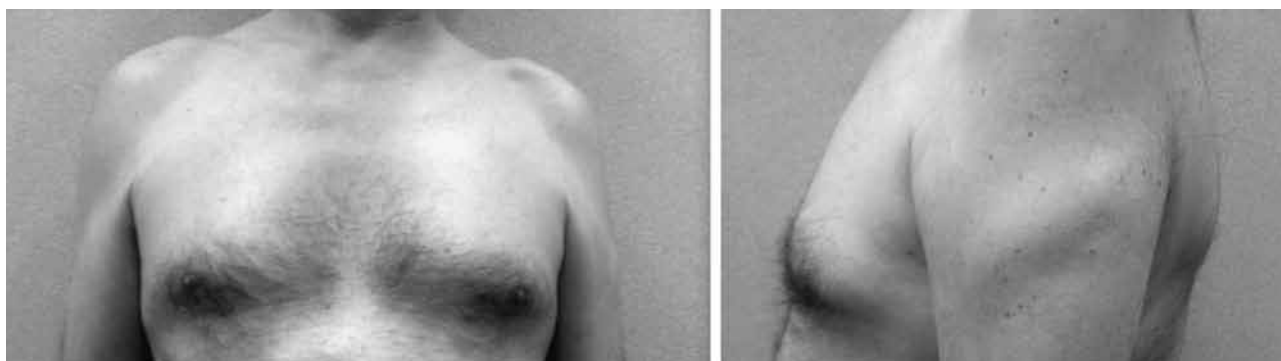


Fig. 1. — Postsurgical Parsonage-Turner syndrome. Atrophy of the deltoid and biceps muscles.

clinical symptoms and recent surgical history. It should be differentiated from other causes of post-operative upper extremity weakness.

ILLUSTRATIVE CASE

A 59-year-old man with a history of B-cell chronic lymphocytic leukemia (CLL), presented with a left cervicobrachialgia, due to foraminal stenosis C5C6 and C6C7. He underwent an uneventful anterior cervical discectomy and fusion (ACDF) C5C7 with an excellent result. Four weeks postoperatively, he experienced severe pain in the left shoulder and arm, after pruning a hedge. The pain increased in the next two days, and was followed by weakness of the left arm. There was also hypaesthesia in the left thumb. Three months after surgery he was seen with marked atrophy of the left shoulder girdle, mainly of the deltoid and biceps muscles (Fig. 1). The circumference of the biceps muscle was 29 cm on the left and 35.5 cm on the right. There was severe tenderness over the left shoulder, biceps and elbow. Active and passive range of motion of the left shoulder were limited. The left biceps and brachioradialis reflexes were absent. The left deltoid strength was graded 2/5, the right biceps strength 3/5 ; all other muscles were normal in this respect. Electromyography showed slight denervation in the triceps, and severe denervation in the biceps and deltoid muscles. Plain radiographs and sonography showed glenohumeral subluxation but an intact rotator cuff. A CT-scan of the cervical spine showed

correct instrumentation (Fig. 2). MRI of the cervical spine did not show any significant residual foraminal stenosis (Fig. 2). MRI scans of brain and brachial plexus were normal. The *Borrelia* serology was negative. Examination of the CSF pleaded against malignant infiltration of the central nervous system. The diagnosis of postsurgical Parsonage-Turner syndrome was made. After temporary oral administration of corticosteroids, conservative treatment was continued with distension arthrography of the left shoulder, shoulder muscle exercises and analgesics. At 6 months follow-up, there was significant pain relief with marked improvement of deltoid and biceps muscle force, but still persisting frozen shoulder. EMG showed increased reinnervation in the C5C7 myotomes.

DISCUSSION

Postsurgical PTS

Postsurgical PTS typically develops within a few weeks after cervical decompression surgery (3,29), exactly as in the case described here. Ninety-six percent of all PTS patients go through an acute phase of relentless pain, worse at night, and unlike anything they have ever experienced before (26). The pain decreases after a few days. It becomes chronic in more than 75% of the patients. In two thirds of the patients, the pain is situated at the insertion of the paretic or the compensating muscles, especially in the periscapular and cervico-occipital

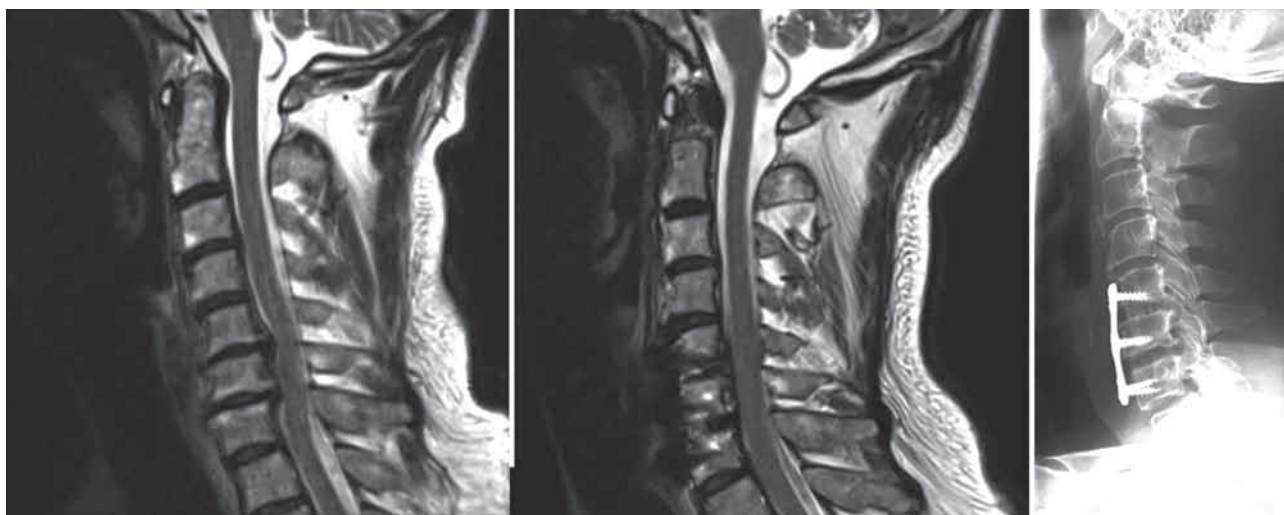


Fig. 2. — From left to right : preoperative MRI showing compression C5C6C7, postoperative MRI showing decompression, postoperative plain radiograph showing correct instrumentation C5C6C7 after decompression.

region. Such pain is most often seen in patients who have persistent scapular instability and/or glenohumeral joint pathology as a complication of altered shoulder biomechanics. Our patient developed a frozen shoulder with glenohumeral subluxation, as in 17% of the cases (27,28). Weakness appears mostly within two weeks after the onset of pain. An upper brachial plexus distribution is the most common pattern, with a patchy paresis of the periscapular and glenohumeral muscles ; it occurs in 71% of the patients (27). The infraspinatus, supraspinatus, serratus anterior, biceps, deltoid and triceps are the most commonly affected muscles (22). Involvement of the long thoracic nerve may lead to winging of the scapula. Phrenic nerve involvement may alter the position of the diaphragm, which can be detected on a chest radiograph (17). Almost 80% have sensory deficits on examination (28,29). Signs of autonomic nervous system dysfunction are present in 15% of the cases (27).

Differential diagnosis

Distinction between postoperative PTS and postoperative C5 palsy is very difficult. C5 palsy *shortly* after recent cervical decompression, for instance due to migration of a graft, is a well-documented phenomenon, occurring after about 5% of such

operations (2,12,21). Indeed, certain cases of postoperative C5 palsy are clearly caused by intraoperative root injury and have been documented via monitoring (14) ; such cases are more likely to manifest immediately on awakening from anaesthesia. However, a proportion of these C5 palsies has a tendency to develop in a delayed fashion after the cervical decompression, making differential diagnosis with PTS more complicated (12,21). However, intolerable pain, followed by weakness and improvement of pain, pleads for PTS. Another important discriminator is the fact that in case of PTS the weakness, sensory deficit, and pain usually do not correspond to the same nerve root or peripheral nerve distribution (9,22). This is in sharp contrast with C5 palsy where C5 motor symptoms predominate. Of course, the differential diagnosis should also be made with other conditions, which can manifest in a delayed fashion after surgery : shoulder joint pathology, thoracic outlet syndrome and peripheral nervous system infections such as neuroborreliosis or HIV (27).

Pathogenesis of postsurgical PT

A number of theories have been put forward to explain the aetiology of postsurgical Parsonage-Turner Syndrome.

Firstly, the stress of surgery may suppress the immune system, allowing activation of a dormant virus (3). Indeed, neuralgic amyotrophy is thought to be of autoimmune origin, and a few studies have attempted to unravel its pathophysiology (25,31). More than 50% of the patients report an antecedent such as infection, unusual physical exercise, surgery, pregnancy and puerperium, vaccination, psychological stress and trauma (29). The patient presented here had several risk factors: recent cervical decompression surgery, unusual physical exercise while pruning the hedge, and a deficient immune system (known CLL). As stated before, the association of PTS with a surgical procedures has been extensively described: the orthopaedic procedures include lumbar spinal surgery (1), cervical decompression (3), knee arthroplasty (8) and hip arthroplasty (23).

Secondly, postsurgical PTS might stem from mechanical insults, such as inappropriate patient positioning, traction or pressure injuries during operative procedures, surgical manipulation of a sensitive root, extradural tethering, predisposition caused by pre-existent myelopathic injury of the corresponding spinal cord segment, and reperfusion after relief of a chronic compression (20).

Diagnosis often missed

Despite all these, postoperative PTS is notoriously underrecognised and usually diagnosed with undue delay (15,18). Suspicion of postsurgical PTS is based on clinical symptoms and history (20,27). When the deficits present in a delayed fashion like in the current case, PTS moves up on the differential list. Notwithstanding, the surgeon should perform a thorough examination and obtain the appropriate imaging (CT/MRI) to rule out C5 palsy (e.g., inadequate decompression, bone graft migration). Electromyography can support the diagnosis of PTS. It is abnormal in 96.3% of the patients (27). Because 1 to 4 weeks are required for Wallerian degeneration to take place, a few weeks should be allowed to pass after the onset of weakness to ensure useful electrodiagnostic findings (4,13). Diffuse denervation, not compatible with a single nerve root, is generally seen in the affected limb. Complete denervation is

often the case. In the first years after an initial EMG, a review EMG may show reinnervation, making it of both diagnostic and prognostic value, like in the current case (10). MRI of the brachial plexus may be indicated: Gazioglu *et al* (12) reported bilateral thickening and hyperintensity signaling of the brachial plexus on MRI. Laboratory data do not contribute to the diagnosis of PTS and are normal in about 75% of the patients (27,29).

Treatment

The cornerstone of the treatment is good pain management, physical therapy and reassurance. A combination of a long-acting NSAID and an opiate has a good effect on the acute pain. Tricyclic antidepressants can be helpful for the second phase pain (30).

van Alfen (34) advises to administer oral prednisolone at a dose of 1 mg/kg/day for one week, tempering to 0 mg/kg during the second week to shorten the duration of symptoms and to obtain earlier recovery. If no evidence of regeneration or recovery within the nerve distribution is found by 6 to 9 months after onset, nerve transfer procedures should be considered to restore function to that nerve distribution (3). Sixty-six percent of the patients show recovery of motor strength within the first month after the onset of weakness and 75% of all patients make a complete recovery within two years (6,13). Sequelae are however more frequent than accepted in the past.

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