From disabling shoulder pain to full functional gain: A hectic approach for higher yield

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ABSTRACT

Idiopathic brachial neuritis also known as Parsonage-Turner syndrome is a well-defined but relatively uncommon clinical entity affecting young adults. It presents with acute-onset severe shoulder pain persisting from days to weeks along with associated weakness and subsequent atrophy of muscles of the shoulder girdle, leading to marked impairment of activities of daily living. Often, the diagnosis is late and sometimes missed in the vast ocean of its mimics. We report a case of idiopathic brachial plexus neuritis in a child, who was aggressively investigated and promptly treated very early in the course of the disease, and subsequently, muscle atrophy was prevented. This case highlights the importance of picking up the disease amid its diagnostic dilemmas and managing proactively before it evolves along its natural course which may take months to years for complete, sometimes partial recovery.

Key words: *Amyotrophic brachial neuralgia, Brachial plexus neuritis, Idiopathic brachial plexitis, Parsonage Turner, Pediatric shoulder pain*

Parsonage-Turner syndrome (PTS), also known as acute idiopathic brachial neuritis, paralytic neuritis of the brachial plexus, cryptogenic brachial neuropathy, and scapular belt syndrome, was first described in 1948, from a series of 136 cases reported by Parsonage and Turner [1,2]. In that cohort, 35% of patients were Indians; however, reporting of this entity from India is very infrequent, possibly reflecting a lack of awareness regarding the disease entity [2]. The disease had a prevalence of 1.64 cases per 100,000 people among the population of Minnesota, United States [3] in the early 80s. PTS is characterized by sudden onset of severe pain, with no preceding history of trauma followed by flaccid paralysis [1]. Gradually, the pain subsides and weakness comes at the forefront [4].

The diagnosis is based on ruling out differentials, important ones being cervical radiculopathy, adhesive capsulitis of shoulder join, shoulder impingement syndromes, and peripheral nerve lesions [5]. A cervical magnetic resonance imaging (MRI) scan helps in ruling out underlying structural abnormality, possibly leading to cervical radiculopathy. Electromyography typically shows acute denervation pattern although variations have been reported [5]. PTS tends to be a self-limiting entity and it takes about months to years for a complete or partial return of power and resolution of pain [6,7].

CASE REPORT

A 15-year-old boy, developmentally normal, born to non-consanguineous parents with good scholastic performance, was apparently asymptomatic till 5 days back. One evening, he started having sudden onset sharp, burning pain of severe intensity, initially in the left neck and shoulder, followed by involvement of the entire left upper limb over the next few minutes. There was no preceding trauma. The pain was more while abducting the arm, lifting arms over the head, and on turning the neck to the opposite side. He was maintaining left upper limb in a position of mild flexion at left elbow joint and adduction at left shoulder joint to minimize pain.

Over the next 2 to 3 h, he was unable to make a fist, flex his elbow, or abduct his arm completely. His opposite upper limb was completely asymptomatic. He reported shock-like sensations over the left arm and forearm on being touched. He was advised over-the-counter nonsteroidal anti-inflammatory drugs following which he had symptomatic relief. However, the next day, the pain of greater severity, weakness, and more pronounced than before were relapsed. He was able to move his arms completely when lying down but had marked restriction of passive range of motion. The child had no history of trauma, unaccustomed heavy weight lifting, and exacerbation of pain with cough or sneeze, local skin changes, recent massage over the neck, recent immunization, preceding viral infection, recent antibiotic use, surgery, or similar illness in the past.

On examination, the child was grimacing with pain (Visual Analog Scale: 8/10). The pulse of 84 beats/minute, regular and all peripheral pulses were equally felt with no inter pulse delay.

Muscle bulk and tone was normal in all four limbs; power was decreased in the left upper limb. As per the Medical Research Council (MRC) grading, power of his shoulder abductors was 3/5 (Figure 1), adductors 4/5, flexors 3/5, extensors 4/5, and small muscles of left hand 3/5 along with drooping of the left shoulder (Figure 2). Sensory examination showed left upper limb hyperesthesia as well as dysesthesia. Deep tendon reflexes were normal. Plantar reflexes showed bilateral flexor response. Wright's, Roos', Adson's, Neer's, Hawkin's, Spurling, and provocative tests were negative [8]. Spine examination was normal.

To begin with, a shoulder X-ray was done which ruled out shoulder dislocation (Figure 3 and 4). Routine blood investigations were normal. MRI brachial plexus was normal (Figure 5) [9,10]. Nerve conduction studies showed axonopathy of the left medial cutaneous nerve of the arm and ulnar nerve. Muscle USG done of shoulder was normal.

The patient was given a short course of tramadol and paracetamol along with intravenous methylprednisolone at a dose of 30 mg/Kg/day for 5 days followed by oral prednisolone (1 mg/Kg) in tapering doses over 6 weeks. He was also initiated on physiotherapy and advised aeroplane splint for the next 2 weeks. Following the treatment, his power improved to MRC grading of 5/5 in all joints of the left upper limb (Figures 6 and 7), pain reduced to a visual analog scale of 2/10, and dysesthesia subsided completely [11,12].

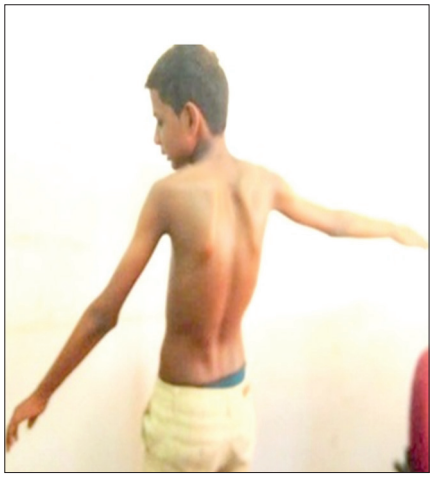

Figure 1: Restriction of abduction of affected shoulder

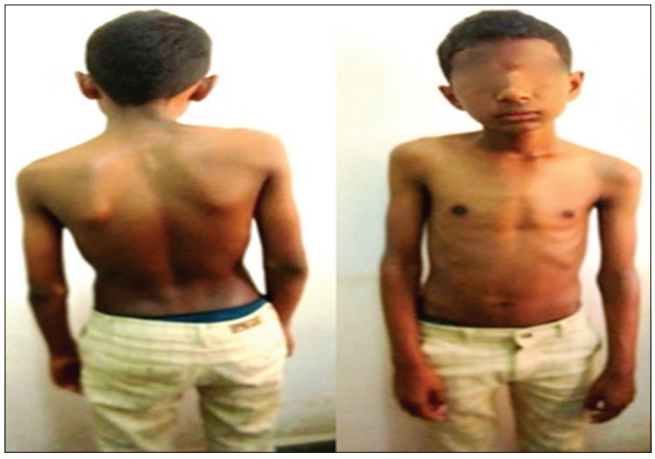

Figure 2: Drooping of affected shoulder

DISCUSSION

Neuralgic amyotrophy commonly manifests as a severe relentless neuropathic pain. It may have an infrequent onset of action in some patients. This pain can occur in two phases: one being severe neuropathic shooting type and the other being the more localized musculoskeletal type. Other sensory symptoms include hyperesthesia and paresthesia which usually accompanies the attack. The onset of weakness usually varies, the majority manifesting within 24 h of the onset of pain, whereas in other cases, there can be a delay of up to >2 weeks [13]. Signs of distal vasomotor autonomic dysfunction can be present in a few cases which are usually visible in the form of local temperature dysregulation, edema at the onset of pain, and trophic skin changes.

Different case studies have found variable patterns of recovery in patients with neuralgic amyotrophy. Almost one-third of patients can continue to have chronic pain and functional deficits even after >6 years [13]. Recent studies have shown less optimistic recovery in the form persistence of pain, weakness, and impaired activities of daily living on long-term follow-up [13]. Corticosteroid-treated patients had favorable outcomes as shown by Van Alfen *et al*. in their 2005 study [12]. A short course of

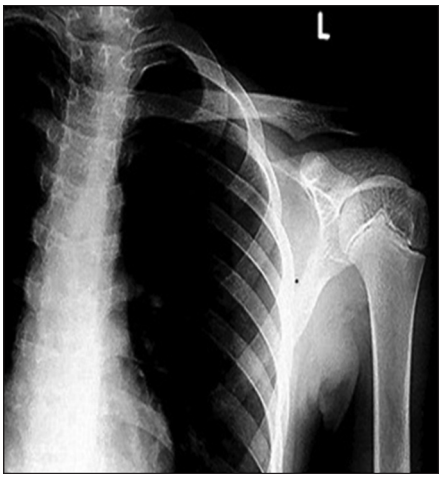

Figure 3: Normal shoulder on X-ray

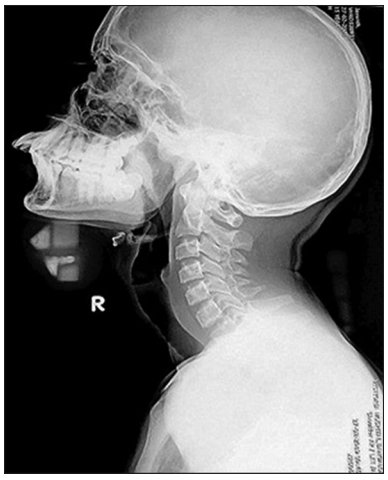

Figure 4: Normal Cervical Spine on X-ray

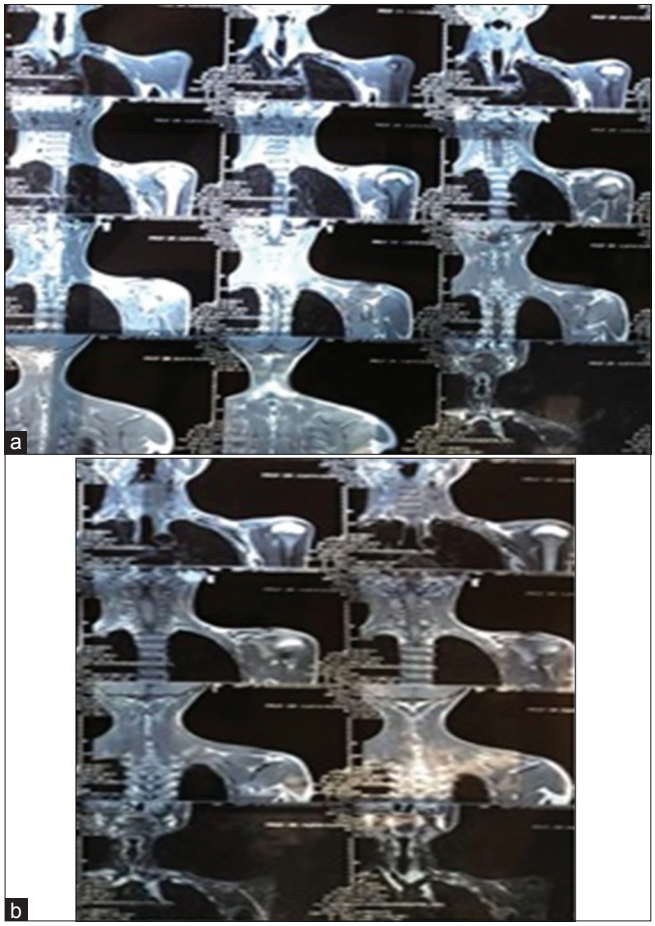

Figure 5: (a and b) Magnetic resonance imaging showing normal brachial plexus with normal shoulder girdle muscles

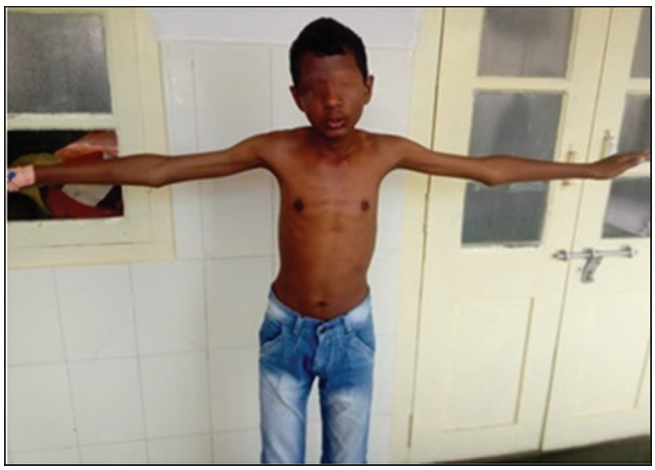

Figure 6: Full range of abduction of shoulder after treatment

oral steroid for 2 weeks, beginning with 60 mg in the 1st week followed by tapering doses over the next week, was used in the above study [12].

Cochrane review shows the dearth of randomized controlled trials regarding the use of steroids in brachial plexopathy [12]. However, retrospective studies do infer that the use of steroids has resulted in reduced duration of pain and relatively faster recovery [11]. The use of pulse intravenous corticosteroids and intravenous immunoglobulin has been reported to have a favorable outcome in a Japanese study of 2013 [11]. To summarize the present case, an onset of acute, non-traumatic and severe pain was observed in a 15-year-old boy. Along with a reduced range of motion, the upper left limb constantly felt dysesthesia and hyperesthesia.

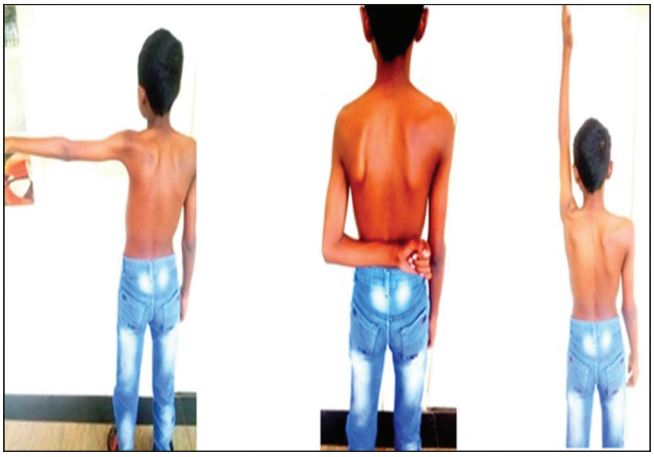

Figure 7: Full range of shoulder movements: After treatment

Bedside tests for shoulder dislocation, adhesive capsulitis, shoulder impingement, cervical radiculopathy, thoracic outlet syndrome, and other regional pain syndromes were all negative. Hence, a provisional diagnosis of neuralgic amyotrophy of left brachial plexus was considered. Other differential diagnoses were ruled out with targeted investigations. Nerve conduction studies showed axonopathy of the left medial cutaneous nerve of an arm and ulnar nerve, which suggested two different nerve involvements arising from the left brachial plexus. This confirmed our provisional diagnosis, and prompt treatment was initiated.

CONCLUSION

Parsonage-Turner syndrome is a diagnosis of exclusion. This case highlights the importance of early diagnosis and prompt management which can potentially halt the natural course of the disease. Awareness among pediatricians and general care physicians is needed for early recognition, appropriate treatment, and prevention of disability.

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