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Original Research Article

Parsonage turner syndrome: a common syndrome usually missed

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ABSTRACT

Background: Parsonage-turner syndrome (PTS) is a rare syndrome that can occur in normal healthy individuals with sudden, rather abrupt, unilateral shoulder pain that may begin rather insidiously but quickly amplifies in severity and intensity. The acute period of pain is subsequently replaced over a course of a few days to weeks with progressive weakness, reflex changes, and sensory abnormalities in varying presentations that typically involve the shoulder girdle musculature and proximal upper limb muscles. There are no definite etiological factors present in causation of this syndrome.
Methods: Prospective multicentric cohort study covering period from 2010 to 2020. 10 young male patients were included in the study group, all of them presented with winging of scapula and pain. Initial evaluation was done at neurology department and after failure of conservative treatment of average period of six month they were referred to plastic surgery department. All patients had ENMG findings preoperatively.
Results: 10 male patients were studied with average age of 24.8 years. Partial paralysis present for an average of 1 year. Repeated micro trauma was the most common aetiology. Intraoperatively 7 patients had anomalous vascular compression of long thoracic nerve (LTN) and three had compression due to fibrous sheath. All patients had complete recovery at end of one year.
Conclusion: Isolated LTN paralysis is a rare condition that is not well known. If conservative management fails, then neurolysis of the distal segment of LTN gives good result if performed within 6-12 months of paralysis.

Keywords: Parsonage-turner syndrome, Serratus anterior muscle, Scapula, Lateral thoracic nerve

INTRODUCTION

Different group of muscles are involved in the movement of shoulder joint, serratus anterior (SA) is one of them. SA play an important function in rotation and stabilization of scapula. Nerve supply of SA is long thoracic nerve (LTN). Because of paralysis of SA there is a very obvious deformity known as winging of scapula, which was first reported by Parsonage and Turner in 1948.[1,2]

PTS is a not a very common disorder, characterized by an acute onset of upper extremity pain. The pain is not positional in nature and usually worse at night and it may be associated with sleep awakenings. Self-limiting pain last for 1-2 weeks, rarely it may persist for longer period. After initial onset of symptoms, it takes few days to months for weakness to appear. Sensory deficits are known to occur but may vary in prevalence. Any peripheral nerve may be affected in PTS but most commonly the upper trunk of the brachial plexus is involved.[1,2] The aetiology of PTS is unknown, but it was commonly reported in various clinical situations, like post infectious, post vaccination and post traumatic conditions. International articles which are available on PTS mainly define the diagnostic technique. However,

there is very limited literature available on Indian patients which can define its aetiology and management. The aim of this study was to diagnose this entity as early as possible and find out the cause of its occurrence and early management.

METHODS

Study design, place and period

Prospective multicentric cohort study was carried out at three tertiary care super speciality hospitals, from Jan 2010 to Jan 2020.

Selection criteria

10 cases of winging of scapula were analysed. All patients presented with shoulder pain and weakness in unilateral arm. They were initially evaluated in neurology dept and managed conservatively. Steroids were given in neurology department as initial management. After 3 months of treatment, when there was no relief, patients were referred to plastic surgery department for further management. In our department patients were again evaluated to find out the cause of shoulder pain and weakness.

All ten patients were physically active and within the age group of 20-25 years. The initial clinical examination was focused to determine the patient's dominant arm, injured side, duration of the weakness, mechanism of injury, presence and location of any pain and presence of Tinel's sign over the LTN trajectory. The paralysis was labelled as either complete (entire axial edge of the scapula lifted in neutral position, visible during forward elevation of the arm, without possibility of recruitment when pushing against a wall) or partial (incomplete lifting of the scapula, especially at the tip, mainly visible during forward elevation of the arm, but relieved by pushing against a wall). An ENMG study was performed preoperative and postoperative respectively.

Inclusion criteria

Partial or complete isolated non-iatrogenic SA paralysis following a single closed direct trauma or repetitive trauma during sports or work-related activities, or an abnormal posture, which was present for at least 6 months, with no clinical or ENMG signs of recovery.

Exclusion criteria

Patients symptomatic with partial or complete SA paralysis of more than 12 months duration were excluded.

Criteria required for intervention

All the patients with mild shoulder pain, winging of scapula and mild latency of LTN, on failed conservative management of 6 months patients were observed for 6 months with no recovery or relief, patients were taken up for exploration and decompression of LTN.

Procedure

The procedure was performed with the patient in lateral position. An 8-10 cm longitudinal incision was given over the mid-axillary line at the level of the fifth rib (Figure 2). The anterior edge of the latissimus dorsi muscle was reflected backwards. The serratus anterior branch of the thoracodorsal artery was identified and followed distally. In all the cases it was seen that anomalous vascular branches of thoracodorsal artery were crossing LTN and in seven patients it caused compression, in other three the compression was due to SA fascia (Figure 3, 4). Once the LTN was identified, external neurolysis was performed by ligating the anomalous blood vessels which was causing compression on the nerve, and also the SA fascia was widely opened to eliminate any restrictions and sharp bends (Figure 5). Patients were discharged from the hospital on the day after the surgery, with no immobilization. Patients were followed up at 1 month, 6 month and 1 year with ENMG investigation.

Ethical clearance was taken from institutional ethical committee.

Figure 1: Winging of scapula.

Figure 2: Marking for exploration.

Figure 3: LTN with vessels passing over the nerve.

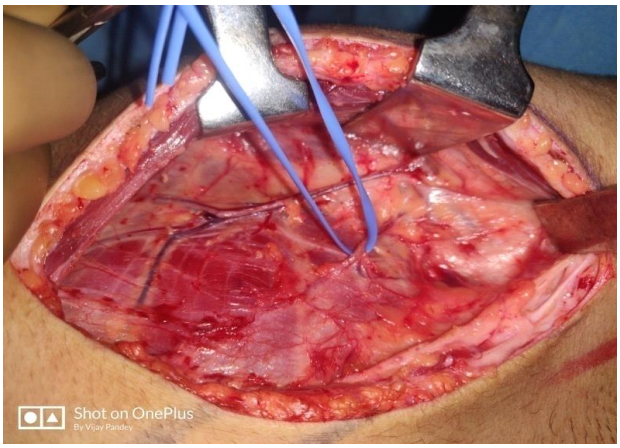

Figure 4: Compression over the nerve.

Figure 5: Compression released.

Statistical analysis

Data was coded and entered into excel sheet and double checking was done to ensure data quality. Discreet data have been presented as proportions while continuous variables (such as age) were expressed as means ± standard deviation. Statistical analysis of difference between proportions have been carried out by use of chi squared test. Statistical significance has been set at $p<0.05\%$. Appropriate graphical representation has been made.

RESULTS

The cohort consisted of 10 consecutive cases with an average patient age of 24.8 years (range 18-34). All were male patients. 09 Patients were right-handed and 01 patient was left-handed. Partial paralysis was present for an average of 1 year (median 06 months) (Table 1). One of the patients was badminton player who may had repeated micro-trauma because of sudden jerky movements. Rest cases were due to traction of the arm, such as stretching or an unusual posture. No triggering factor was identified. All the patients experienced pain. In seven cases, this pain was isolated neuropathic pain (lateral thorax and/or posterior scapula) and in three cases the pain was due to anterior rocking of the shoulder. A pseudo-Tinel sign was present in seven case on the LTN course near the fifth rib; the Tinel sign was negative in rest of the cases. The preoperative ENMG examination showed abnormal muscle innervation in all patients. The denervation was partial in all cases. All patients were having winging of scapula.

During the surgical procedure, isolated fascial fibrosis was found in three cases and in other seven cases it was associated with abnormal vascular findings. The average postoperative follow-up was 1 year. The results were excellent in all patients, none of the patients experienced worsening of symptoms. All patient experienced decrease in pain and/or improvement in serratus anterior muscle function. There were no complications related to the surgical procedure. In nine cases scapular winging was no longer present. In one case the winging was equal to the preoperative situation. In all cases, the re-innervation was fairly extensive.

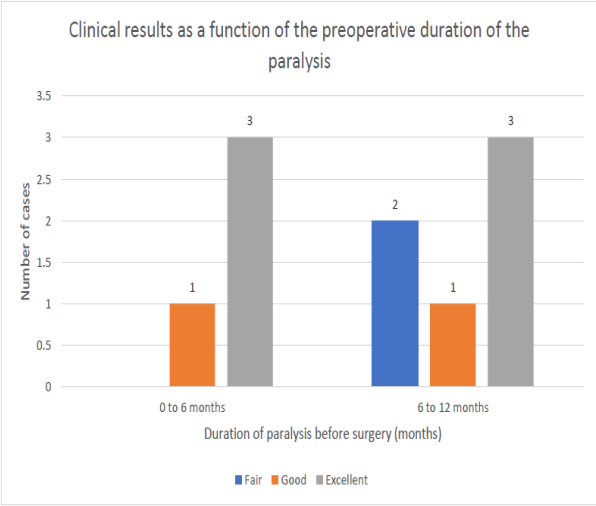

Figure 6: Clinical results as a function of the preoperative duration of the paralysis.

Table 1: Corelation between duration before surgery and neuropathic pain.

Duration (Months)	Neuropathic pain	No neuropathic pain	Total
0 to 6	3	1	4
6 to 12	4	2	6
Total	7	3	

The chi-square statistic is 0.0794. The p value is 0.77816. The result is not significant at $p<0.05$

Table 2: Corelation between recovery and neuropathic pain.

Recovery	Neuropathic pain	No neuropathic pain	Total
Partial recovery	6	3	9
Complete recovery	1	0	1
Total	7	3	

The chi-square statistic is 0.476. The p value is 0.490. The result is not significant at $p<0.05$

Table 3: Corelation between duration before final outcome and neuropathic pain.

Variables	Neuropathic pain	No neuropathic pain	Total
Fair	2	0	2
Good	2	0	2
Excellent	3	3	6
Total	7	3	

The chi-square statistic is 2.857. The p value is 0.239. The result is not significant at $p<0.05$

Table 4: Final outcome of all the cases,

Outcome indicators (n=10)	Pre-operative (%)	Post-operative (%)	% Change (Outcome in terms of improvement)
Tinel sign positive	7 (70)	0 (0)	100
Winging of scapula	10 (100)	2 (20)	80
ENMG (abnormal denervation)	10 (100)	2 (20)	80
Neuropathic pain	7 (70)	0 (0)	100

DISCUSSION

The current study presents a result of a small group of patients who underwent surgical exploration of lateral thoracic nerve. The procedure was performed on patients with serratus anterior paralysis of mechanical origin who did not recover with conservative and steroid treatment. There was a similar study conducted by Le Nail et al but they studied on isolated serratus anterior paralysis and only distal release of LTN. This injury is not very common, but has been described in several international published studies. Indian data on PTS was lacking. There are studies which included more than 10 cases, after PTS cases but iatrogenic injuries were excluded: 211 non-operated cases for Pikkarainen et al 41 operated cases for Nath et al 27 non-operated cases for Friedenberg et al and 14 cases for Gozna et al.[3-6] The aetiology in the above studies was similar in type and proportion to the one found in the current study; we also found that the condition most often occurred on the dominant side.

The long thoracic nerve is a thin nerve that averages 24 cm in length. Although a few potential compression sites exist, the exact location where the nerve is being compressed remains controversial. Several studies have suggested that it is compressed at the middle scalene muscle.[7] In cadaver studies, the LTN was found to be stretched over the 2nd rib, with a fibrous sheath causing the nerve to bend sharply during shoulder abduction. In our study, we observed that vascular and fibrous structures were mechanically restrict the LTN. Vascular branches cross over the LTN at the lower part of the serratus anterior around the 5th rib. In the current study, we found one or several anomalous branches causing compression of LTN and restricting the movement of LTN along the thoracic wall. This restriction was either proximal or distal. The immediate pain relief felt by patients following either distal or proximal neurolysis, may be explained by the removal of tension on the nerve similar results we are also found in our study.

In Nath et al study of 50 cases of proximal neurolysis (nine in the context of PTS), every patient had involvement of other shoulder muscles, evidence that the superior trunk of the brachial plexus was affected.[8] The pathology described is therefore different than the one in the current study. There are studies which suggest that if SA is not activated then also winging of scapula can occur. As a consequence, it is important to confirm the existence of an objective deficit and the neurogenic nature of the condition using clinical and ENMG examinations. Our study and very few studies were reported the type of pain, its correlation to the duration of paralysis and the prognosis. Other than neuropathic pain related to compression and/or LTN tension, compensation by other scapula-stabilizing muscles is likely to bring about pain and muscle spasms. These problems were often found in the trapezius muscle. A postural disorder can appear and significantly impact the patient's function. In the current study, neurolysis was highly effective at relieving neuropathic pain. Scapular winging and scapulothoracic dysfunction can cause subacromial impingement and acute rotator cuff injuries, which can also cause pain and increase dysfunction.[3]

Although every patient received some benefit from the surgery, the ones who were operated on within 10 months of their paralysis had the best results. Beyond this time frame, the SA has lost some of its ability to recover and compensatory muscle activation has become chronic. The ENMG analysis is not always a good indicator of injury severity and has no prognostic value. We always perform this analysis before performing surgery to confirm the neurogenic origin and isolated nature of the SA deficit. In many cases, it also reveals increased distal motor latency. If untreated, spontaneous recovery can be expected in 50% of cases at the most. Pikkarrainen et al reported that 8 of their 12 cases of partial paralysis due to trauma had recovered. If there is no spontaneous recovery, extensive palliative measures are suggested by some authors, namely pectoralis major transfer or scapulothoracic fusion, but the results are inconsistent and may not be long-lasting. Gozna et al recommended waiting 6 months before performing surgery. Pikkarainen et al recommended waiting two years before performing palliative surgery.[3] We recommended not waiting more than six months to perform surgical release because chronic pain and compensatory muscle spasms are very disabling and hard to correct, as demonstrated in this study. If there are no signs of re-innervation in this time frame, the possibility of complete spontaneous recovery later on is low.

Limitation

This study although multicentric, comprised of a small sample size involving young, otherwise healthy active male patients. The sample size could not be large in spite of duration of ten years of study period. Hence study cannot be said conclusive for general population.

CONCLUSION

Isolated LTN paralysis is a rare condition that is not well known. In cases with a mechanical mechanism of injury (trauma or micro-trauma), it can be compared to entrapment neuropathy. If spontaneous recovery does not occur, neurolysis of the distal segment of the LTN very often leads to good results because it reduces the tension on the nerve and removes any compression. However, it should be performed within 18 months and before any pain appears due to compensatory muscles spasms. This is a safe and effective procedure, especially when performed within 6-12 months of paralysis. Beyond this time frame, neurolysis can still provide useful functional improvement and spare the patient from palliative surgery.

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