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Concomitant Ipsilateral Hourglass-Like Constrictions of Suprascapular and Axillary Nerves: Report of a Rare Case

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Hourglass-like constriction (HGC) of the nerve is a rare cause of nerve palsy and has been reported for many nerves. We were unable to find previous reports of concomitant constriction of the suprascapular and axillary nerve in literature. Our patient was a young male with shoulder paralysis of 7-months duration. On exploration we found two HGCs of the suprascapular and axillary nerves. A neurolysis of the suprascapular nerve was carried out. The axillary nerve constriction was deemed to be severe and a nerve transfer using the branch innervating the medial head of the triceps motor was done. Patient recovered excellent shoulder function at 1-year post-surgery. Awareness about this rare occurrence will prevent poor outcome from addressing the constriction at only one site. Nerve surgery should be considered for patients who do not show any improvement in 6 months.

Level of Evidence: Level IV (Therapeutic)

Keywords: *Hourglass-like construction of nerve, Axillary nerve palsy, Parsonage–Turner syndrome, Neuralgic amyotrophy, Nerve transfer*

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INTRODUCTION

Hourglass-like constriction (HGC) neuropathy is a neurological condition caused by fascicular constrictions of one or more individual nerves.[1] Definitive diagnosis of this condition can be made only after surgical exploration and thus can be challenging. Ultrasound and magnetic resonance neurography (MRN) can aid in preoperative diagnosis.[2] HGC have been reported more commonly around elbow and involving radial, posterior interosseous, median and anterior interosseous nerves. They are less often encountered around shoulder and have been reported for suprascapular, long thoracic and axillary nerves.[3] Concurrent constrictions of ipsilateral suprascapular and axillary nerve have not been reported to the best of our knowledge but must be looked for (if clinically suspected) as highlighted by this case report as this may result in a suboptimal outcome if only one nerve constriction is addressed. We would like to report this unique case and its outcome.

CASE REPORT

A 33-year-old right-hand dominant hotel manager, presented with a right shoulder injury sustained in a road traffic accident 8 months earlier. Patient reported having normal function of the shoulder immediately after the accident. One month after the injury, patient developed some pain in the neck region radiating to right arm with progression to weakness of shoulder abduction over a few days. On examination at presentation, there was substantial atrophy of deltoid, supraspinatus and infraspinatus with 0/5 MRC motor grading of these muscles. He was unable to perform any active shoulder abduction or external rotation (Fig. 1). However, the motor examination for the elbow and hand muscles was normal. There was sensory loss over lateral aspect of upper arm, corresponding to the sensory innervation of the axillary nerve. EMG study showed denervation potentials in deltoid, supraspinatus and infraspinatus muscles. MRI showed no injury to the brachial plexus and no local pathologies in the shoulder. As it was already 7 months since paralysis, the patient was planned for surgical exploration. Through the conventional anterior approach, upper brachial plexus was explored. HGCs were noted in the suprascapular nerve at two sites, 1 inch apart, with twisting of the nerve at those regions (Fig. 2). Under microscopic magnification, the constriction sites were neurolysed and the nerve was untwisted. Under microscope, we could observe extreme constriction of the nerve but continuity of fascicles across

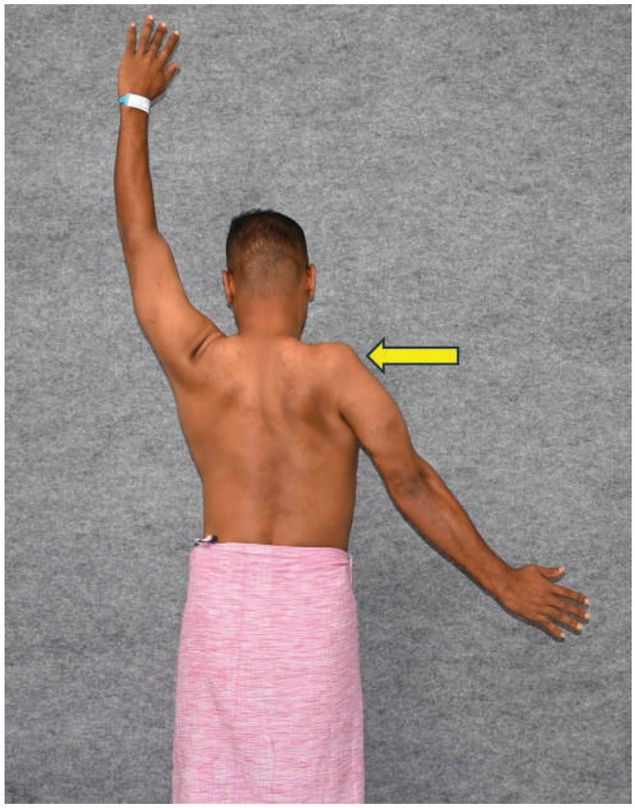

Fig. 1. Clinical picture of the patient showing absent shoulder abduction. Note the atrophy of the deltoid (arrow).

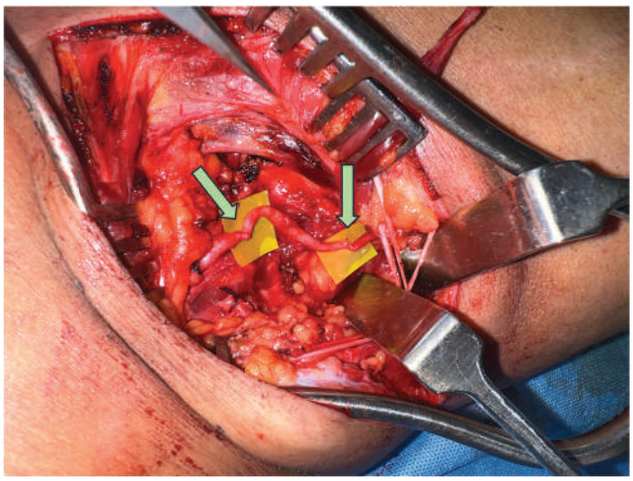

Fig. 2. Intra-operative picture of the suprascapular nerve showing two HGCs (arrows).

the constriction site and also observed some conduction across the constriction sites on intraoperative electrical stimulation with 5 mA. Hence, it was decided to leave the nerve to recover spontaneously.

Following that, through the posterior approach, axillary nerve was exposed. In axillary nerve as well, two

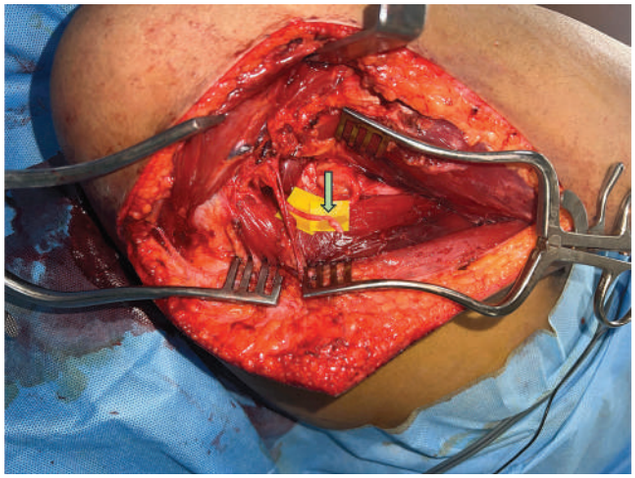

Fig. 3. Intra-operative picture of the axillary nerve showing small segment severe HGC (arrow), the second construction was in the depth of the quadrangular space.

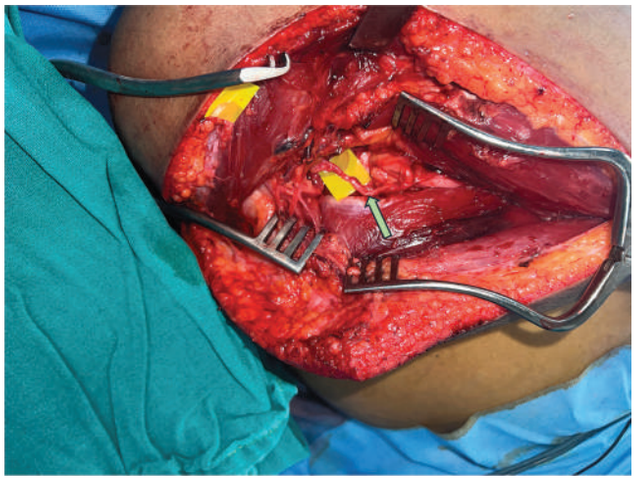

Fig. 4. The axillary nerve was divided distal to the distal constriction site and neurotised with medial head of triceps motor branch (arrow).

constriction bands were noted at the level of the quadrangular space, lying about 4 cm apart (Fig. 3). A similar procedure of microscopic neurolysis of HGC was done. However, the nerve at the distal constriction site was found to be extremely thin and axillary nerve showed no response to electrical stimulation across it. Hence, a decision to proceed with distal nerve transfer to innervate the axillary nerve was taken. The motor branch to the medial head of triceps was chosen as the donor as it showed a matching calibre and good response to electrical stimulation (Fig. 4), with sufficient length to reach the axillary nerve beyond the constriction sites. Shoulder abduction and external rotation was assessed at 3, 6 and 12 months follow up. Recovery of supraspinatus and deltoid was

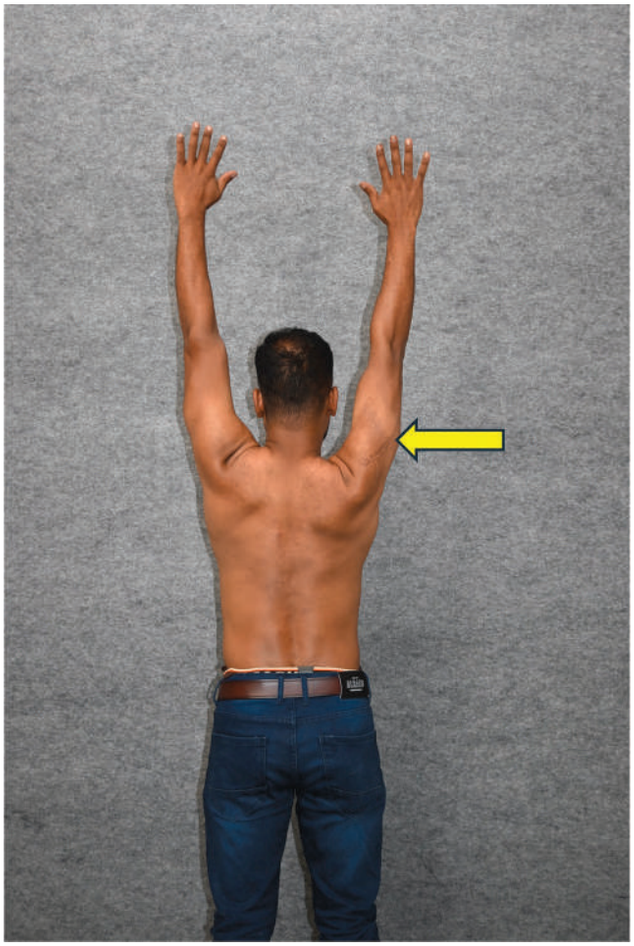

Fig. 5. One-year post-surgery outcome where the patient is able to perform full shoulder abduction. Note the 'normal like' prominence of the deltoid muscle (arrow).

noted at 6-months follow up and at 1-year follow up shoulder abduction and external rotation showed almost complete recovery (MRC 4+/5) with full range of motion at the shoulder (Fig. 5).

DISCUSSION

HGC of the peripheral nerves leading to the muscle paralysis is not related to any compressive pathology but is thought to have an unusual aetiology of its own.[4] The first intraoperative case of HGC was reported by Wilhelm in 1970.[3] He reported torsion of posterior interosseous nerve, without any visible source of external compression. The natural course of the disease involves spontaneous, acute onset of muscle palsy in the distribution of one or more upper limb nerves. There are different theories suggested for the development of HGC. Lundborg et al. emphasised that the pathogenesis starts with endoneural

oedema that persists for a long period of time.[5] He postulated that the persistent swelling of the nerve eventually leads to stiffness of the fascicles that cannot tolerate any form of stretch or kinking, leading to torsion and constrictions. On the other hand, Nagano et al. postulated an initial inflammation of the nerve leading to swelling and adhesion of fascicles renders them susceptible to mechanical trauma during limb movement.[6] Histologically, loss of both myelinated and unmyelinated fibres, fibroblasts with collagen deposition, absent regeneration, microvascular occlusion and CD8 positive T-lymphocyte infiltration on the nerve fibres has been noted.[7]

A scientific dilemma exists as to whether HGC is feature of Parsonage–Turner syndrome (neuralgic amyotrophy [NA]) or is an entity of its own. It has been postulated that, due to certain differences, the two conditions are different entities that may coexist, as elaborated by Pan et al.[7] Granata et al.,[8] in an article, distinguishes between the two entities. When the lesion involves peripheral nerves, and presentation involves both sensory and motor symptoms, the lesion is more likely to be an HGC, rather than NA. Literature describes the latter as being characterised by lesion in the roots, trunks or cords of the brachial plexus, associated with invalidating pain, and predominantly involving motor nerves. Classic NA is associated with good prognosis, while HGC is usually associated with a bad prognosis, unless neurosurgical intervention is done. In a clinical setting with severe pain followed by weakness in an upper limb, high resolution MRN of the brachial plexus and extremity may be helpful in diagnosis.[2] The presence of sensory loss in the axillary nerve territory, and absence of pathognomonic severe pain associated with NA, directed our diagnosis towards HGC, in the present case.

Timing of surgical intervention is critical but a difficult decision, because most patients with NA are known to exhibit spontaneous recovery and clinically, HGC neuropathy greatly resembles NA. It is known that 70% of patients with NA experience some degree of clinical recovery by 6 months. However, the patients who do not show any clinical and EMG evidence of recovery in first 6 months are likely to not recovery or have persistent muscle weakness. Such patients can be considered for surgical intervention even if the diagnosis of the HGC is not established. However, if the diagnosis of the HGC can be established, earlier surgical intervention should be considered. It cannot be overemphasised that the nerve recovery potential is very time bound and delaying intervention carries risk of loss of motor end plates, Schwann cell senescence and fibrosis and fatty replacement of denervated muscle, which are irreversible and drastically reduces chance of recovery. Hence, a timely surgical intervention is essential to provide a good motor recovery.

The sway towards nerve surgery in patients who are likely to have incomplete recovery is more justified in the recent times with the availability of effective distal nerve transfers. The distal nerve transfers provide for a more reliable motor recovery with no appreciable donor deficit. We prefer to offer surgical intervention to these patients if there is no recovery in the first 6 months. In view of the possibility of multiple constrictions in a nerve, it is mandatory to inspect the nerve over a segment on either side of the constriction. In our case, both the suprascapular and axillary nerves had two constrictions. The constriction site must be internally neurolysed under microscope. If fascicular continuity across the constriction can be confirmed or there is any conduction across the construction site on electrical stimulation, neurolysis alone is sufficient and is known to provide excellent results. However, if fascicular diameter is narrowed significantly, i.e. to <25% of its original expected diameter, reconstruction becomes the necessary option.[9] Type of reconstruction would depend on the length of the segment involved and the distal nerve transfer options available. Excision and end-to-end repair, distal nerve transfer or excision of the involved segment and bridging nerve graft are the options in the respective order of preference. Where timely reinnervation of the relevant muscles cannot be achieved, secondary procedures such as tendon transfers can be used to improve function, but results are inferior to nerve surgery.[10]

A high degree of suspicion for this rare cause of nerve palsy is necessary. Conservative management can be initially applied. However, nerve surgery should be considered for patients who do not show any evidence of spontaneous improvement in 6 months. Management depends on the severity of the constriction. Neurolysis is sufficient if in-continuity neural tissue is seen during microscopic release of the constriction. More severe constrictions could be considered for resection and repair/nerve grafting, or distal nerve transfer.

DECLARATIONS

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Ethical Approval: This study was exempt from institutional board review per our institutional policy on small case reports.

Informed Consent: Written informed consent was obtained from the patient before the study. He has provided his consent to share his photographs and clinical information.

Use of AI and AI-Assisted Technologies: AI and AI-assisted technologies were NOT used in writing this manuscript.

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