Uncommon Compressive Neuropathies of Upper Limb: A Case Series

M. Alam Parwaz[1] B. Chakravarti[1] S. Mehrotra[1] Vikas Singh[1] Arjun Handa[1]

[1]Department of Plastic Surgery, Army Hospital Research and Referral, New Delhi, Delhi, India

Address for correspondence M. Alam Parwaz, MS, Mch, Department of Plastic Surgery, Army Hospital Research and Referral, New Delhi, Delhi 110010, India (e-mail: alamghausia@gmail.com).

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Abstract

Known by different names like peripheral compressive neuropathies, tunnel syndromes, entrapment neuropathies, and so forth, over 30 clinical syndromes have been identified afflicting the peripheral nerves of upper limb. Due to the inherent dynamic nature of the compressive process the nerve pathophysiology remains in a state of flux which varies with limb position, exertion, or duration of compression. This leads to a plethora of clinical presentations, often noncharacteristic results of electrodiagnostic studies and no other standard diagnostic investigation. As a result the diagnosis and management of all compressive neuropathies except carpal tunnel syndrome remains difficult. This article presents a series of uncommon compressive neuropathies. In a tertiary care referral center, 14 cases of compression neuropathy excluding the carpal tunnel syndrome and thoracic outlet syndrome diagnosed over the past 11 years have been described. A representative case each of quadrangular space syndrome, anterior scalene syndrome, long thoracic nerve syndrome, suprascapular nerve syndrome, cubital tunnel syndrome, Guyon's canal syndrome, posterior interosseous nerve syndrome, radial tunnel syndrome, and syndrome of superficial branch of radial nerve will be discussed including their presentation, work-up, conservative management, surgical steps, and follow-up. High index of clinical suspicion following exhaustive clinical examination was the bedrock of diagnosis. Further evaluation with nerve conduction studies (NCS), electromyography (EMG), magnetic resonance imaging (MRI), and digital subtraction angiography (DSA)/CT angiography was mostly inconclusive. We also came across anomalies of anatomy of radial nerve during surgical exploration. The most important aid in diagnosis of rare compressive neuropathies is to be aware of the same and entertain this possibility. A thorough knowledge of anatomy and clinical examination clinches the diagnosis. Investigations may be of some assistance. Outcome after surgery is almost always gratifying.

Keywords
- compressive neuropathies
- uncommon
- upper limb

Introduction

Compressive neuropathies are a result of chronic compression over a peripheral nerve affecting sensory or motor fibers. It affects anatomical regions where it passes through bony, fibrous, osseofibrous, or osseomuscular tunnels. The syndrome may be caused by tumor compression (intraneural or extraneural), trauma (blunt, sharp, or secondary to repetitive action), infection, metabolic abnormalities, toxins, iatrogenic, vascular (ischemic, aneurysms, tumor), muscular compression, anatomical variations, or idiopathic causes. Dynamic changes of a tunnel during movements can create traction or compression of a nerve. Tunnel syndrome could produce a neuropraxis and eventually an axonotmesis. Patients present with symptoms that can range from vague complaints of diffuse pain or numbness to specific complaints of muscle weakness and altered sensations over localized skin areas. Sensory symptoms and signs present before

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motor signs. Treatment must address the etiology causing nerve compression. Conservative measures may relieve compression caused by repetitive actions. Conservative measures consist of immobilization, rest, exercise, ultrasound, heat, massage, anti-inflammatory medications, and corticosteroid injections. Surgical decompression is done if conservative therapy fails. Surgery allows direct visualization of the tunnel and the surrounding tissue. A success rate of 97% has been recorded if decompression was performed within 6 months of onset of clinical symptoms.[1] Some tunnel syndromes are treated by transposing the nerve, neurolysis, tenosynovectomy, arthrodesis, or osteotomy.

Tunnel Syndromes in Upper Limb: Case Description and Discussion

In upper limbs, the long course of the nerves and great arc of movements places them at risk in diverse locations and from various actions. Also, the possibility of double crush of nerve has to be kept in mind to obtain optimal results. While carpal tunnel syndrome is very common entity and thoracic outlet syndrome is also easily recognizable due to the accompanying vascular symptoms, there are a host of other rarer compressive neuropathies including spinal accessory nerve syndrome, scapulocostal syndrome, suprascapular nerve syndrome, quadrilateral space syndrome, intercostobrachial nerve syndrome, syndrome of the musculocutaneous nerve in the shoulder region, dorsal scapular nerve syndrome, long thoracic nerve compression, radial nerve compression in the upper arm, supracondylar process syndrome, syndrome of the musculocutaneous nerve at the elbow, medial antebrachial cutaneous nerve syndrome, sulcus ulnaris syndrome, pronator teres muscle syndrome, supinator syndrome, anterior interosseous syndrome, flexor carpi ulnaris muscle syndrome, ulnar tunnel syndrome, syndrome of the deep branch of the ulnar nerve, syndrome of the tendinous arch of the adductor pollicis muscle, syndrome of the superficial branch of the radial nerve, distal posterior interosseous nerve syndrome, digital collateral nerve syndrome, and so forth. In a tertiary care center we came across several rare compressive neuropathies which are being discussed in this article.

Quadrangular Space Syndrome

Presentation

Right hand dominant adult male, pilot by profession and a racquet player, presented with 5 months history of discomfort around right shoulder. After 2 months of onset of discomfort he developed weakness of the shoulder and a visible contour asymmetry of the ball of shoulder. Despite abstaining from sports he remained symptomatic. There was no history of any definitive injury. Clinically he had wasting of deltoid with associated paresthesia along the distribution of upper lateral cutaneous nerve of arm. Electromyography (EMG) was suggestive of deltoid denervation (►**Fig. 1a–d**). X-ray of the shoulder and humerus were normal. Digital subtraction angiography (DSA) with arm in abduction did not show any evidence of posterior circumflex humeral artery compression despite reports of arteriography and magnetic resonance imaging (MRI) of some help in the diagnosis.[2,3]

Anatomy

Quadrangular foramen is limited proximally by the lower margin of the teres minor muscle, distally by the upper margin of the teres major muscle, laterally by the humerus, and medially by the long head of the triceps muscle. The axillary nerve and the posterior circumflex humeral artery

Fig. 1 (**a–d**) Patient with contour asymmetry in shoulders with wasting of muscles in right shoulder region.

pass through the opening. The opening decrease in size as the teres major and minor muscles approach each other in abducted position of arm. Hypertrophy of teres muscles as in throwing athletes or abnormal position of arm while sleeping or while under anesthesia can cause axillary nerve compressive neuropathy. Spontaneous entrapment of the nerve by fibrous band or muscle in the quadrilateral space can also occur.

Management

As the patient had definitive features of denervation and a history of 5 months he was taken up for exploration through posterior upper arm approach. No definitive pathology was found in that region. Only teres muscle insertion was released and postoperatively there was remarkable improvement in pain within a few weeks of surgery (►**Fig. 2a**, **b**).

Anterior Scalene Syndrome

Presentation

A young adult man presented with features of numbness and vague pain along medial border of one hand and forearm with associated intermittent bluish discoloration of the hand after exertion or working overhead which developed over the last 3 to 4 months. Clinical history revealed that he was habituated to carrying heavy knapsack on long patrols. Clinical examination including Adson's test were unremarkable. Investigations including X-ray chest, color Doppler, MRI brachial plexus, and electrodiagnostic studies were inconclusive.

Anatomy

Anterior and middle scalene muscles originate from transverse processes of cervical vertebrae and insert on the tubercles of first rib. Bounded by the anterior scalene muscle, the first rib, and the medial scalene muscle, the posterior scalene foramen admits the brachial plexus and the subclavian artery to the costoclavicular space. Many anatomical variations and dynamic changes in the anatomy can cause narrowing of the foramen. The etiologies include closer insertion of two scalene on first rib, fibrous bands between the two muscles, hypertrophy of anterior scalene, strong contraction of scalene as in carrying heavy knapsack, lifting heavy weights by hand, gigantomastia, presence of accessory scalenus minimus muscle, and cervical rib.

Management

After a trial of conservative therapy for a month, the patient was managed by exploration and scalenotomy of the visibly hypertrophied anterior scalene muscle along with removal of excessive fibrous tissues at its lower end leading to improvement in all symptoms by 2 months.

Long Thoracic Nerve Syndrome

Presentation

A young adult man presented with weakness of right shoulder with associated mild discomfort around the joint especially while doing pushups in routine morning exercises for last few months. He gave no history of any definitive injury to shoulder or arm. On specific questioning he gave history of frequently carrying heavy bag with strap slung over the right shoulder. Examination revealed classical winging of scapula on doing pushups or leaning against the wall with stretched hands (►**Fig. 3a–c**). There was no tenderness or Tinel's sign anywhere along the course of the nerve. There was no evidence of involvement of any other component of brachial plexus. X-ray and MRI did not reveal any abnormality.

Anatomy

After exiting through the scalenus medius muscle, the contribution from cervical roots 5, 6 and/or 7 unite to form long thoracic nerve which lies deep to the subclavian artery and then over the chest wall abutting the serratus anterior muscle. It can potentially be compressed between clavicle and first two ribs or against the coracoids process or as it passes through hypertrophied scalenus medius muscle. Athletic activities or carrying heavy loads on one side can precipitate chronic compression.

Management

He is being managed conservatively with physical therapy including avoiding lifting weights and range of motion exercises for shoulder. Neurolysis has also been described at a point where branch of thoracodorsal artery traverses the nerve.[4,5]

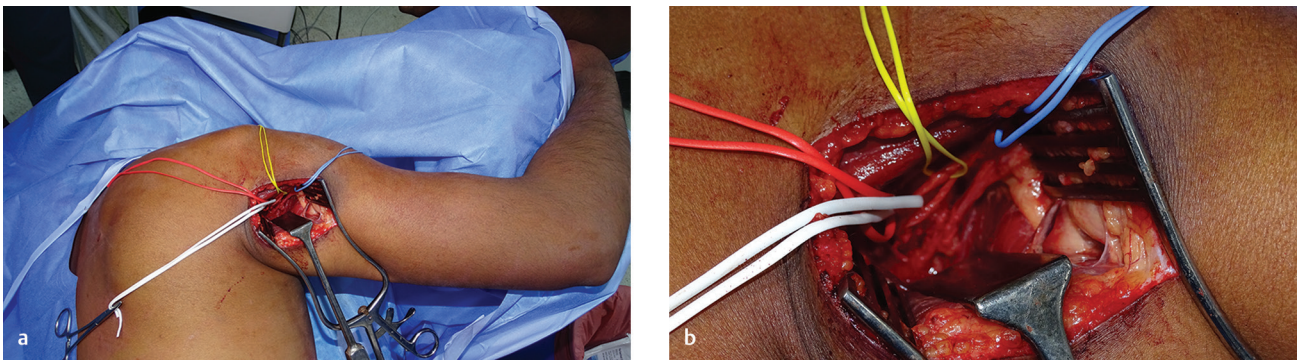

Fig. 2 (**a, b**) Exploration of quadrangular space with identification of axillary nerve and its cutaneous and motor branches dissected out with no evidence of pathology.

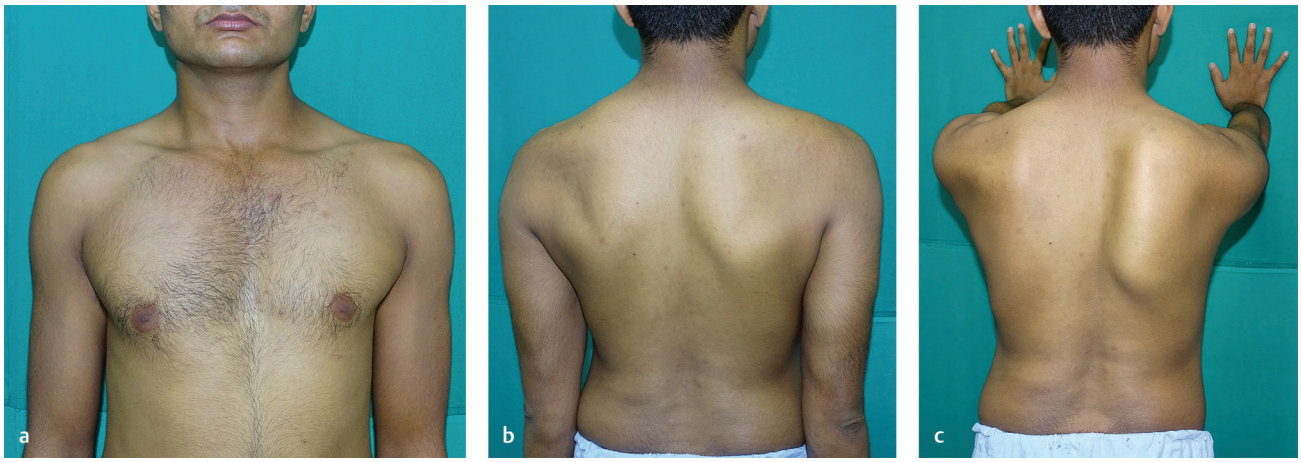

Fig. 3 (**a–c**) Patient with long thoracic nerve syndrome with winging of scapula on pushing against the wall.

Fig. 4 (**a**) Evidence of hollowing of supraspinous fossa on clinical examination. (**b, c**) Exploration of suprascapular nerve revealed entrapment of nerve for which external neurolysis was done.

Suprascapular Nerve Syndrome

Presentation

A young healthy adult man presented with severe diffuse pain around left shoulder and posterior aspect of arm of 8 months duration preceded by a fall on the shoulder while playing football. He was referred as a suspected case of upper brachial plexus injury with associated weakness and wasting around the shoulder. Clinically he had prominent spine of scapula with wasting of supraspinatus and infraspinatus muscles, tenderness along superior border of scapula, and weakness of shoulder abduction and external rotation (►**Fig. 4a**). MRI revealed Parsonage–Turner syndrome. EMG had denervation of supraspinatus.

Anatomy

Suprascapular nerve is the first posterior branch originating from Erbs point which passes posteriorly in the supraclavicular fossa. It passes through the suprascapular notch/foramen on the superior border of scapula. Here it passes through a tight fibro-osseous or osseous tunnel under suprascapular ligament. As the scapula moves extensively over thorax, the nerve is liable to compression at this tethering point. Further it supplies infraspinatus muscle by passing through the supraspinatus muscle and curving around the base of spine of scapula under spinoglenoid ligament which forms the second potential constricting tunnel. Repetitive abduction and external rotation of arm while doing overhead work cause mechanical irritation to the nerve. Rotator cuff injury or any decompensation of the cuff muscles alters the dynamics around shoulder and can precipitate suprascapular nerve compressive neuropathy.

Management

Surgical release of suprascapular tunnel was performed along with external neurolysis resulting in early relief in pain and subsequent partial recovery of muscle wasting (►**Fig. 4b**, **c**).

Cubital Tunnel Syndrome

Presentation

A software programmer, young adult woman, presented with pain and numbness on medial border of right hand and forearm of 2 months duration, which worsened after prolonged typing and at night. On clinical examination she had mild wasting of first web, positive card test, no clawing, hyperesthesia over ulnar distribution, and Tinel's sign positive over cubital tunnel. Ultrasonography confirmed edema of nerve in cubital tunnel and nerve conduction studies localized increased latency in the same region of the ulnar nerve by inching technique. She improved with conservative management including ergonomic positioning of chair in work place, splinting of elbow at night, and avoiding repeated flexion of elbow during the day. Another patient was an elderly male with similar features for 7 months along with early clawing of ulnar two digits. Additional investigation for rheumatoid arthritis for this patient was negative.

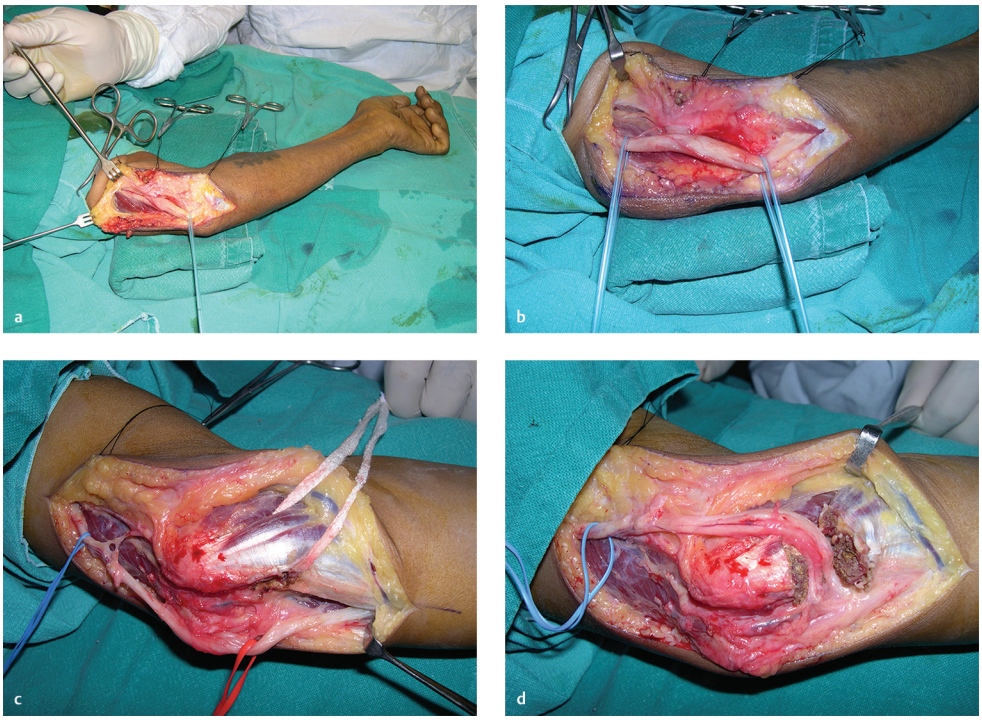

Fig. 5 (**a–d**) Cubital tunnel decompressed with ulnar nerve translocated anteriorly in submuscular plane.

Anatomy

Behind the medial epicondyle the ulnar nerve passes through a fibro-osseous tunnel surrounded by joint capsule, medial collateral ligament, and cubital tunnel retinaculum traversing between medial epicondyle and olecranon process. This 0.5 cm tunnel becomes more constricted on elbow flexion. Other constricting elements could be Struthers ligament or anomalous anconeus epitrochlearis muscle in distal medial part of humerus.

Management

The second patient was managed by surgical exploration through a posteromedial incision where release of cubital tunnel retinaculum along with the fibrous tissue encasing the nerve on medial aspect of distal humerus was done with anterior transposition of the nerve. Medial epicondylectomy is also described as possible treatment (►**Fig. 5a–d**).

Guyon's Canal Syndrome

Presentation

An adult male presented with 2 months history of severe pain in ulnar three digits of right hand which was worse at night. He was not able to work with the hand due to pain. There was no history of any specific trauma. On examination he had involvement of lower ulnar nerve with sparing of sensations on ulnar aspect of dorsum of hand. X-ray skyline view of carpal tunnel and ultrasound of wrist were normal.

Anatomy

Ulnar neurovascular bundle passes through fibro-osseous tunnel on ulnar side of wrist lying volar to carpal tunnel retinaculum between pisiform and hook of hamate which is covered by flexor carpi ulnaris tendon. Compression in this tunnel may affect the superficial sensory branch or deep motor branch or both the branches. Compression can occur due to bony lesion of surrounding two bones or degenerative joints in vicinity or any space occupying lesion like lipoma, ulnar artery aneurysm, giant cell tumor, and so forth.

Management

In presence of severe pain and muscle atrophy, the patient was taken up for exploration of the nerve where a ganglion was seen arising from pisotriquetral joint. Excision of the ganglion and external neurolysis of the nerve was done (►**Fig. 6a**,b). Associated release of carpal tunnel retinaculum is also described.

Posterior Interrosseous Nerve Syndrome

Presentation

A young adult man, clerk by trade, presented with 5 months history of weakness of left wrist leading to difficulty in typing and poor grip (►**Fig. 7a**,b). He also had difficulty in picking objects using fingers. He noticed radial deviation of hand on wrist extension. The onset was associated with pain in forearm lasting for 3 weeks. There was no history of any trauma. On examination there was wrist drop with radial deviation on attempted extension. There was associated drop-finger deformity. Weakness of extensor compartment muscles was seen with some sparing of extensor carpi radialis longus (ECRL) and extensor carpi radialis brevis (ECRB) (supinator 4+/5, brachioradialis 5/5, ECRL, ECRB 4/5, extensor carpi ulnaris (ECU) 3/5, extensor digitorum communis (EDC) 2/5, extensor pollusis longus (EPL) 3/5). No sensory deficit was noticed. Electrodiagnostic studies revealed compound muscle action potentials (CMAPs) from left radial not recordable and sensory nerve action potentials (SNAPs) to be slightly reduced in amplitude. EMG was normal in triceps but had features of

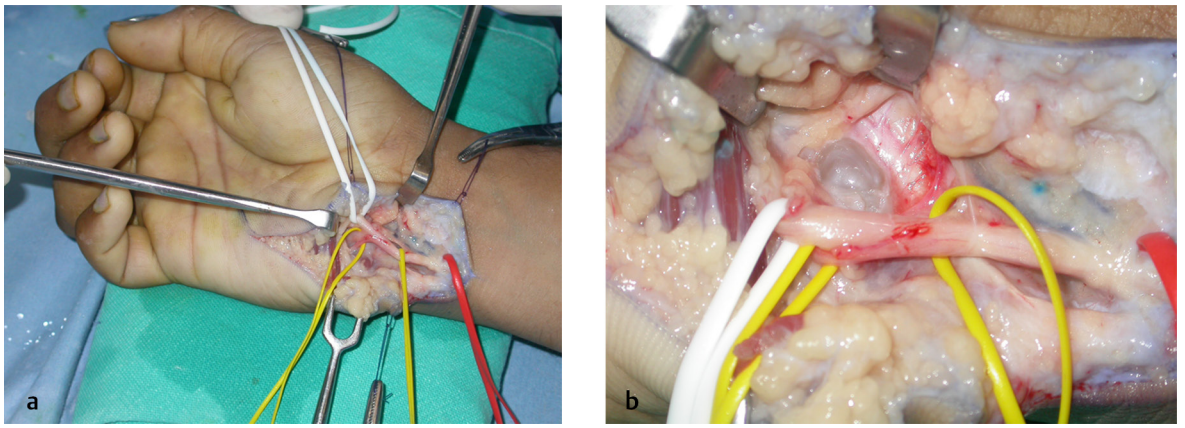

Fig. 6 (**a**) Guyons canal explored and branches of ulnar nerve dissected and identified. (**b**) Sensory branch encircled by white vascular loop and deep motor branch encircled by smaller yellow loop deep to it with ganglion seen posteriorly.

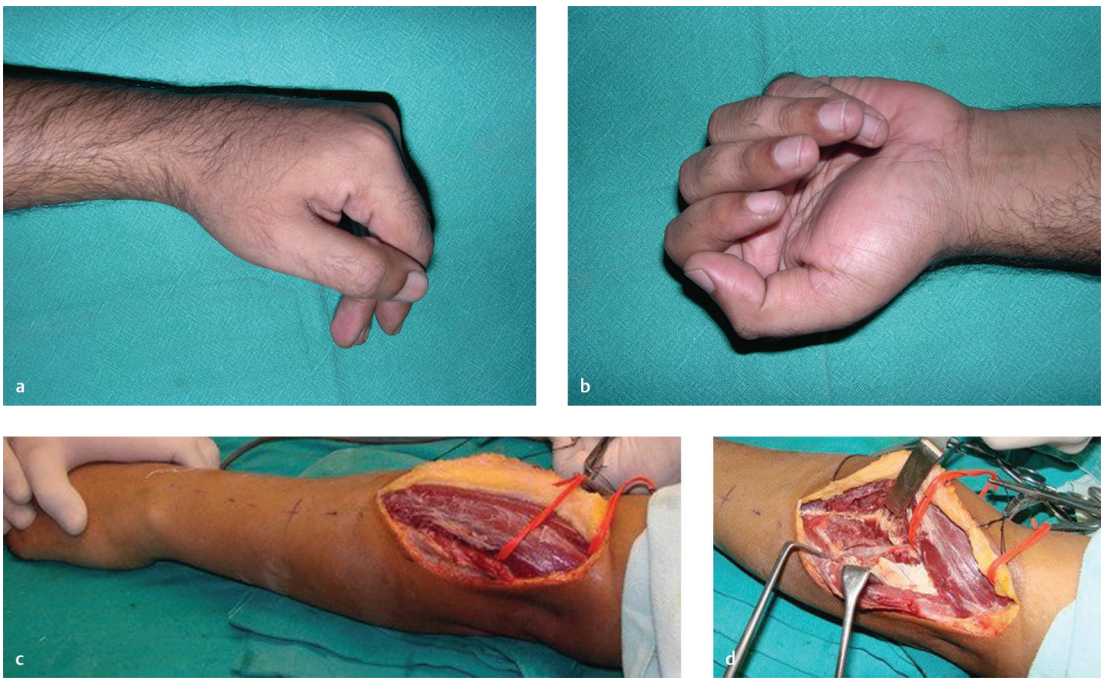

Fig. 7 (**a**) Wrist drop and finger drop deformity seen in case of posterior interosseous nerve syndrome. (**b**) Radial deviation on attempted wrist extension. (**c**) Posterior approach between ECRB and EDC. (**d**) Taped posterior interosseous nerve after division of Arcade of Frosche and part of superficial belly of supinator.

denervation in EDC (Fibs 1+, motor unit action potentials [MUAPs] small and prolonged).

Second case was of a lady in third trimester who presented with similar features of 11 months duration and more severe weakness in forearm (►**Fig. 8a**, **b**).

Anatomy

In the groove between brachioradialis and brachialis in lower lateral forearm, radial nerve divides into a superficial sensory branch and deep posterior interrosseous motor branch after it has innervated brachioradialis and extensor carpi radialis longus. The deep branch passes through two heads of supinator muscle to emerge on the posterior aspect of interrosseous membrane. This whole area is a constricting tunnel where the nerve can be compressed by fibrous edge of extensor carpi radialis longus, fibrous edge of proximal head of supinator called Arcade of Frosche, fibrous edge of distal head of supinator, or a vascular channel called leash of Henry. When the compression occurs proximal to the division of the radial nerve then it is called radial tunnel syndrome.

Management

External neurolysis after division of Arcade of Frosche and part of superficial belly of supinator was done through posterior approach between ECRB and EDC in upper forearm with good recovery in 3 months and near complete recovery by 15 months (►**Fig. 7c**, **d**).

Second patient was managed by exploration and was found to have anomalous deep branch of radial nerve.

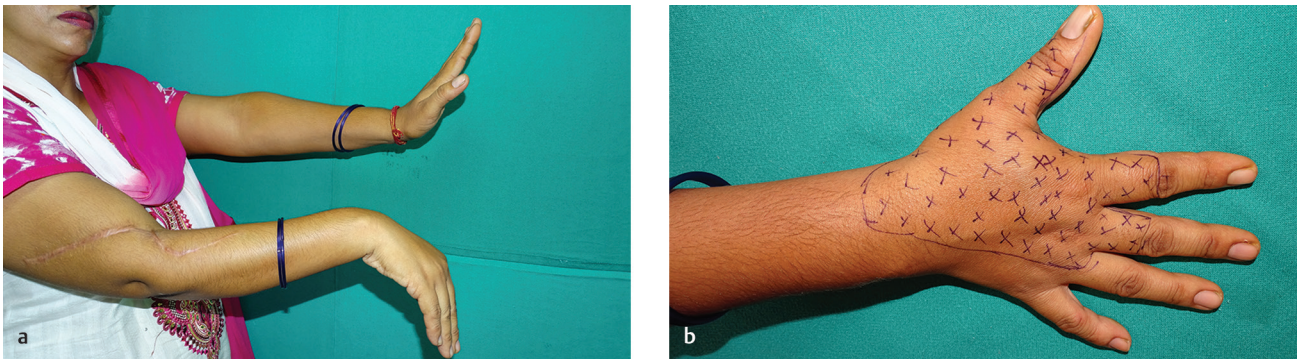

Fig. 8 (**a, b**) Second case of posterior interosseous nerve syndrome with history of 18 months and intraoperative finding of anomalous anatomy of deep branch of radial nerve. Postoperative image shows persistent wrist drop and radial sensory loss without any improvement.

Table 1 Summary of cases

S. no	Diagnosis	Patient no.	Age (y)	Sex	Duration (m)	Management	Outcome
1	Quadrangular space syndrome	1	32	M	5	Surgical release of teres insertion	Improvement in pain within weeks
		2	28	M	6	Surgical release and neurolysis	Improvement in pain
2	Anterior scalene syndrome	3	36	M	4	Scalenotomy of hypertrophied scalenus anticus muscle	Improvement in all symptoms in 2 months
3	Long thoracic nerve syndrome	4	26	M	5	Conservative	No visible recovery over 3 months
4	Suprascapular nerve syndrome	5	29	M	8	Release of suprascapular tunnel and neurolysis	Early improvement in pain with subsequent partial recovery of power
5	Cubital tunnel syndrome	6	23	F	2	Conservative	Complete relief in pain
		7	67	M	7	Surgical release of cubital tunnel retinaculum with anterior sub-muscular transposition of nerve	Marked relief in pain and partial improvement in clawing in 6 months
		8	55	M	5	Release & external neurolysis	Relief in pain by 2 months
6	Guyon's canal syndrome	9	42	M	2	Excision of ganglion	Complete relief in symptoms
		10	35	M	5	Surgical release and neurolysis	Improvement in symptoms
7	Posterior interosseous nerve syndrome	11	39	M	5	Release of Arcade of Frosche	Near complete recovery in 15 months
		12	43	F	11	Surgical release and exploration. Anomalous anatomy of deep branch.	No relief till 5 months. Tendon transfer being planned.
8	Radial tunnel syndrome	13	38	M	2	Conservative	No improvement in 3 months
9	Syndrome of superficial branch of radial nerve	14	68	F	7	Conservative including local steroid infiltration	Partial improvement

Syndrome of Superficial Branch of Radial Nerve

Presentation

Elderly lady presented with pain, parasthesia, and numbness around wrist and dorsum of hand precipitated by working as homemaker like sweeping or wringing cloths. Symptoms could be reproduced by forearm pronation and ulnar wrist flexion. There was also positive Tinels over distal radius ~ 6 cm proximal to styloid process. Tests for rheumatoid arthritis were normal and Quervain's disease was ruled out by appropriate tests.

Anatomy

Superficial branch of radial nerve originates proximal to supinator muscle in the groove between bravchioradialis

and brachialis, passing distally on radial side of forearm concealed in the belly of brachioradialis muscle and lying lateral to radial artery. In the middle third of the forearm, it lies behind brachioradialis and lateral to radial artery. It passes dorsally, beneath the tendon of brachioradialis, 7 cm above the styloid process of radius. It pierces the deep fascia to become subcutaneous as it passes dorsally and divides into lateral and medial branches. Lateral branch supplies the skin of radial side and ball of thumb. Medial branch supplies the ulnar aspect of thumb, dorsal aspect of index and middle fingers, and radial aspect of dorsum of ring finger.

Management

She reported improvement after restriction of precipitating activities and a local infiltration of injection triamcinolone (►**Table 1**).

Conclusion

Tunnel syndromes of upper limb usually present with vague and noncharacteristic features of insidious onset. It is of utmost importance to be aware of various types of compressive neuropathies and keep it as a differential diagnosis for such nonspecific complaints. Clinical investigations may be of some help only if the possibility of diagnosis of compressive neuropathies is considered. It is usually seen that before such patients present to a tertiary care center in our setup they had already exhausted most of the conservative measures and a lot of time. Since the final outcome after surgery depends upon the duration of nerve compression, surgery cannot be postponed indefinitely and satisfying results are obtained after early surgery on delayed presentation.

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