Ultrasound-guided Infraclavicular Brachial Plexus Block for Supracondylar Humerus Fracture in a Patient with Central Obstructive Sleep Apnoea due to Charcot-Marie-Tooth Disease

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ABSTRACT
The use of regional anaesthesia techniques in the presence of degenerative neurological disease is a relative contraindication. The subsequent reticence of anaesthesiologists is perhaps reflected in the absence of any published cases of a Charcot-Marie-Tooth (CMT) patient that received a peripheral nerve blockade for surgery. We report a CMT patient, who presented with a supracondylar distal humerus fracture that benefited from an ultrasound-guided infraclavicular brachial plexus block.

Keywords: Brachial plexus, Infraclavicular.

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INTRODUCTION
Charcot-Marie-Tooth disease (CMT) is a heterogeneous group of inherited, progressive peripheral neuropathies that affect both sensory and motor nerves. It affects one in 3,300 individuals and represents one of the most common causes of inherited neuromuscular diseases, most often diagnosed between mid-childhood and within 30 years of age.1

Although classified into several types, based on clinical characteristics, nerve conduction velocity and inheritance, the most common types are the autosomal dominant CMT1 and CMT2. The former characterized by a decrease in motor and sensory nerve conduction velocities with demyelination, while the latter characterised by relatively normal nerve conduction but a histopathology of axonal loss.2 Clinically, most patients with CMT have disabilities due to weakness of distal small muscles and associated deformities, but relative sparing of proximal muscles.

Due to its compromising effects on respiratory function, general anaesthesia, muscle relaxation and ventilation with intubation, this poses concern with regards to recovery of nerve conduction, muscle power and respiratory function. Furthermore, a potential association of anaesthetic agents as malignant hyperthermia triggers and the risks of further injury to already deteriorating peripheral nerves due to regional anaesthesia or incorrect positioning, all add to the anaesthetic challenges posed by CMT.3,4

The literature search did not reveal any reported cases discussing the use of peripheral nerve blockade to address these challenges in a CMT patient.

CASE REPORT
We present a 53-year-old male patient, weighing 90 kg, who was diagnosed with CMT type 1 at the age of 3, who presented with a left supracondylar humerus fracture. He had significant deformities of all four extremities associated with bilateral lower extremity, mid-calf level, stocking type sensory neuropathy with significant muscular wasting level. Despite significant contractures in both hands, motor weakness and decreased sensation in the right hand, he was able to ambulate with the help of a walker and splints. He was prescribed nocturnal (BiPAP) Bi-level positive airway pressure, for central obstructive sleep apnoea (OSA), progressive phrenic nerve latency and respiratory muscle weakness. Recently, he had been non-compliant for 2 months due to aerophagia causing nausea. Subsequently, he had reported frequent nocturnal awakening with shortness of breath, without orthopnoea, but needing to sleep with two pillows due to significant gastro-oesophageal reflux disease (GERD), for which he was using pantoprazole. He had no known drug allergies and pre-operative laboratory tests were all within the normal range.

An open reduction-internal fixation of supra-condylar fracture was planned. The patient agreed to regional anaesthesia for the surgery, to avoid post-operative respiratory compromise due to sedatives, opiates and muscle relaxants with intubation, obviated by significant GERD, all of which were likely to require intensive care unit (ICU) admission. It was decided to perform an ultrasound (US) guided infra-clavicular brachial plexus (BPB) block, which would provide sufficient surgical anaesthesia to the surgical site, distal humerus and elbow region, and theoretically avoid any proximal spread of the injected local anaesthetic above the clavicle level and avoid any effects on the ipsilateral phrenic nerve. We aimed to monitored this complication with serial bed side spirometry (PFT).
The PFT restrictive pattern revealed; FVC: 1.88 l (52% predicted—prd), FEV1: 1.50 l (51% prd) as expected with numbers, such as, PEF: 263 ml (57% prd), FEV1/FVC 80%.

Ultrasound (US) guided brachial plexus block (BPB) was performed with the patient supine, after aseptic preparation of the infra-clavicular skin with deciding 0.5 (0.5% chlorhexidine and 70% isopropyl alcohol). The axillary artery was visualised with a SonoSite MicroMaxx, 3.8 cm 13-6 MHz linear US transducer (SonoSite Inc, Bothell, WA), scanning in the infra-clavicular, para-sagittal plane, medial to the coracoid process. The skin entry point, between the clavicle and the rostral end of the US transducer, was infiltrated with 2% lidocaine. A 100 mm 21 G isolated short bevel tip needle (Stimuplex, B Braun Medical Inc, Bethlehem, PA) was introduced under ‘in-plane’ direct US visualisation. The needle was advanced from the superior aspect so that its tip was located posterior to the axillary artery. Without neuro-stimulation, following negative aspiration for blood, 20 cc of local anaesthetic (LA) solution (ropivacaine 0.375 % + lidocaine 0.6%) was injected, with negative aspiration after each 5 cc. The distinctive view of LA spread, covering the posterior border of the axillary artery, extending towards the lateral and medial sides, was achieved (Fig. 1). The needle was repositioned for further 5 cc injections of the same LA solution around the lateral and medial cords, located respectively to the axillary artery (Fig. 2).

After completion of the LA injections, US of the supraclavicular fossa, confirmed no LA spread above the clavicular level (Fig. 3). Total surgical anaesthesia was achieved within 25 minutes in the territories of the four major nerves of the brachial plexus (musculocutaneous, radial, median and ulnar nerves). Due to some preserved sensation, intercostobrachial nerve infiltration across the proximal medial side of the arm was performed with a 22 G needle, and 10 cc of LA (bupivacaine 0.25% + lidocaine 1%) solution.

Pulmonary function test (PFTs) performed 45 minutes after completion of the block revealed very similar results to the pre-block PFT: FVC: 1.77 l (49% prd), FEV1: 1.39 l (47% prd), PEF: 231 l (50% prd). In the operating room, the patient was positioned on a bean bag in the lateral position with careful protection of pressure areas. Vital signs were all stable for the 2.5 hours surgery, during which the patient required no analgesic medication or sedatives. Upon completion of the surgery, the patient was transferred to the recovery room, with no pain or difficulty in breathing. He requested opioids only once; which was 9 hours after the block, when he received 10 mg oral oxycodone. Return of full sensation and motor activity on his left hand and arm, on the four major nerve territories occurred in the 25th hour after BPB. He did not experience any neurological or respiratory sequelae and was discharged home the next day.

**DISCUSSION**

Charcot-Marie-Tooth disease type 1 is the most common form of CMT, which accounts for more than two-thirds of the cases. The typical phenotype presents with distal weakness, atrophy, sensory loss and foot deformities with predominance of motor symptoms. The onset is in first or second decade of life, with most patients maintaining ambulation and normal life expectancy. Restrictive PFTs are associated with respiratory muscle weakness in up to
30% of CMT patients but in symptomatic pulmonary involvement, FVC ranges between 0 and 30% of predicted. In the present patient, initial FVC was 52% of the predicted value, obviating his nocturnal BiPAP therapy for the last year.

According to the ‘double-crush’ phenomenon, suggested as a possible mechanism of further neurological injury, patients with pre-existing neural compromise may be more susceptible to injury when exposed to a secondary insult at another site. Secondary insults may include mechanical (needle or catheter induced trauma), ischemic (epinephrine-induced vasoconstriction) or toxic (LA neurotoxicity) risk factors, often associated with regional anaesthetic techniques. Accordingly, anaesthesiologists have been reluctant to perform regional anaesthesia techniques in patients with pre-existing neurological disorders for fear of worsening of the neurological state.

Hebl et al reviewed 139 patients with a pre-existing neurological disorder with a mean duration of 23 ± 23 years, receiving neuraxial anaesthesia with standard doses of local anaesthetics. Zero new or progressive postoperative deficits were reported. 74% of the patients reported active neurological symptoms (sensori-motor deficits, parenthesis, dyesthesias or hyperreflexia) at the time of the surgery. The reports of peripheral nerve blocks in patients with pre-existing neurological disorders are limited to one case report but not to any case series.

Various intraoperative factors can precipitate acute respiratory failure in CMT patients: First of all, the supine position will not be tolerated, especially by the patients with severe orthopnoea. Second, these patients are sensitive to the effects of central depressants, opioids and neuromuscular blocking drugs. Third, loss of accessory respiratory muscle function due to a regional block affecting the intercostal muscles will produce acute ventilatory failure in the conscious patient dependent upon these muscles for normal tidal breathing. Hirsch suggested careful assessment of diaphragmatic function for patients with severe CMT type 1, presenting for anaesthesia. A decrease of 30% in forced vital capacity between the erect and lying position and paradoxical movement on inspiration would be basic indicators of diaphragmatic involvement with CMT.

We postulate a number of advantages of the US-guided infraclavicular BPB block for our patient. Firstly, it provided sufficient surgical anaesthesia and prolonged analgesia for the surgical area, avoiding supplementary opioids in our patient with central OSA. The ultrasound facilitates avoidance of pulmonary and vascular structures, allows precision of delivery and confirmation of LA spread, while confirming the absence of LA spread above the clavicle level, which could have resulted in the blockade of the phrenic nerve. Among the different approaches to blockade of BPB, LA distribution both above and below the level of the clavicle is expected following interscalene and supraclavicular blocks. However, Rodriguez et al demonstrated that with the infraclavicular approach, the spread of the local anaesthetic was limited to the infraclavicular region, without extension to the supraclavicular area, which results in the very low rate or absence of side effects related to the unintended blocks of the phrenic, recurrent laryngeal or cervical sympathetic nerves. This prevents phrenic nerve block, hoarseness and Horner’s syndrome respectively. Again, in the light of incidences of 67 to 80% paralysis of phrenic nerve with the supraclavicular block and 100% with interscalene block, the same group demonstrated that no significant change in PFTs occurs following an infraclavicular block and concluded that the block did not produce a reduction in the respiratory function. Accordingly, our post-block PFTs resulted in very little difference from the pre-block PFTs, with no respiratory complaints from the patient. The recovery of the normal sensation and hand muscle function within 25 hours showed that we did not worsen the patient’s neurological status.

**CONCLUSION**

The authors believe that ultrasound guidance during infraclavicular brachial plexus block helped to provide optimum anaesthetic care for our patient with Charcot-Marie-Tooth disease type 1. Perhaps the wider use of peripheral nerve blocks might avoid prolonged intubations and ICU admissions in patients with pre-existing neurological conditions.

**REFERENCES**


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