Clinical Neurophysiology Practice 6 (2021) 203–205

Contents lists available at ScienceDirect

Clinical Neurophysiology Practice

journal homepage: www.elsevier.com/locate/cnp

Case report

Jerks of the latissimus dorsi muscle and intercostal neuralgia after posterolateral thoracotomy

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ARTICLE INFO

Article history:
Received 22 February 2021
Received in revised form 7 June 2021
Accepted 17 June 2021
Available online 6 July 2021

Keywords:
Botulinum toxin
Clinical neurophysiology
Ehlers-Danlos syndrome
Iatrogenic neuropathy
Intercostal nerve
Involuntary movements
Jerks
Latissimus dorsi muscle
Neuropathic pain
Posterolateral thoracotomy
Post-thoracotomy pain syndrome
Thoracodorsal nerve

ABSTRACT

Introduction: Post-thoracotomy pain syndrome (PTPS) is a common complication related to intercostal nerve injury. During this type of surgery, although less frequently, thoracodorsal and long thoracic nerves can also be injured, and jerks of peripheral origins may appear. We report a case with intercostal neuralgia and latissimus dorsi muscle jerks after posterolateral thoracotomy.

Case report: A 55-year-old woman with Ehlers-Danlos Syndrome presented with a typical picture of PTPS along the right T5 dermatome following posterolateral thoracotomy at the level of the fifth intercostal space. Approximately six months after the surgery she developed frequent jerk-like involuntary movements of the right latissimus dorsi muscle. Neuropathic pain along the T5 dermatome was partially relieved with thoracic epidural block. No special attention was paid to the jerks until three years later. A neurophysiological study demonstrated a peripheral origin of these movements and the patient was then treated with periodic injections of botulinum toxin. In response, involuntary movements of the latissimus dorsi muscle disappeared.

Significance: To our knowledge, this is the first case with PTPS and post-thoracotomy latissimus dorsi muscle jerks in a patient with Ehlers-Danlos Syndrome. A correct diagnosis together with identification of iatrogenic neuropathic disorders allow the delivery of targeted treatments. In such cases clinical neurophysiology helps to determine a correct diagnosis.

1. Introduction

Post-thoracotomy pain syndrome (PTPS) is an underestimated complication that presents in approximately 50% of patients who undergo these surgeries (Rogers and Duffy, 2000). Intercostal nerve injury is the most likely predisposing factor for the development of severe chronic neuropathic pain. Intercostal nerve lesion is likely to happen during posterolateral thoracotomy and contributes to post-thoracotomy neuropathic pain. This clinical picture is characterized by spontaneous and/or evoked pain and sensory loss of any type along the area innervated by the injured thoracic nerve (Guastella et al., 2011). During these types of surgeries, although less frequent, thoracodorsal and long thoracic nerves can also be injured. As a consequence of partial nerve injury, jerks of the muscle innervated by the injured nerve may appear usually between several days and several years after the surgery (Aslam et al., 2009; Belluzzo et al., 2015; Carnero-Pardo et al., 1998; Hao and Clarke, 2002; Lagueny et al., 2014). Long series of post-thoracotomy intercostal neuralgia and several cases of post-thoracotomy jerks of the serratus anterior or of the latissimus dorsi muscle have previously been reported. To our knowledge, we describe the first case with PTPS and post-thoracotomy latissimus dorsi muscle jerks in the same patient.

2. Case report

A 55-year-old woman underwent a posterolateral thoracotomy at the level of the fifth intercostal space for resection of an unknown pulmonary nodule in the upper lobe of the right lung. The space occupying lesion turned out to be of benign origin. Her medical history included Ehlers-Danlos Syndrome (EDS) – genetically confirmed with features of thoracolumbar scoliosis, light dermatosis, hepatitis C virus (HCV) antibody-positive HCV RNA-negative without liver damage, hypertension and chronic pain. She previously had bilateral hip replacements as well as left foot

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https://doi.org/10.1016/j.cnp.2021.06.003
2467-981X/

arthrodesis, bilateral hand arthrodesis, left carpal tunnel syndrome, sympathectomy due to palmar hyperhidrosis and right apical pulmonary bullae resection. Shortly after the posterolateral thoracotomy, she was referred for intense spontaneous 'burning pain', allodynia and numbness distributed along the T5 right dermatome (from the scar area to her right nipple). Six months after the surgery she developed continuous involuntary contractions in the lateral portion of the right latissimus dorsi muscle near the thoracotomy scar (Video). This appeared regardless of the posture and seemed to worsen slightly with arm abduction. She could not be sure of the persistence of these movements during sleep. However, she did claim to be occasionally aroused from sleep due to a tightness from the axilla to the mammary area.

During the first year after the surgery, she received multiple treatments for post-thoracotomy neuropathic pain without any improvement. Finally, after epidural injection of ropivacaine 0.1% and betamethasone 12 mg at the T5-T6 level she reported partial relief of spontaneous and evoked pain. However, numbness and involuntary movements persisted. Four years after the posterolateral thoracotomy the patient was referred to the Neurology Department because of the persisting continuous involuntary movements. Brain magnetic resonance imaging was normal. Mild weakness and pain were noted with right shoulder movements. Upper limb deep tendon reflexes were normal and symmetrical. Scapular winging was not observed. She was treated with eslicarbazepine, levetiracetam and clonazepam but no therapeutic response was observed. The patient was then referred to Clinical Neurophysiology and an electromyographic study (EMG) was performed. Surface electromyography (EMG) and needle EMG showed bursts of continuous semi-rhythmic discharges in the right latissimus dorsi muscle. Bursts lasted 300–600 ms, with a frequency between 0.6 and 0.9 Hz (Fig. 1). Needle EMG of the right latissimus dorsi muscle showed evidence of chronic reinnervation (polyphasic motor unit action potentials with slight increase in duration). The needle EMG findings of other muscles supplied by the same nerve roots, including deltoid, biceps brachii, triceps brachii and abductor digiti minimi, did not show involuntary activity. Motor conduction studies of both thoracodorsal nerves were normal and did not show asymmetry. These findings suggested the diagnosis of right latissimus dorsi muscle jerks of peripheral origin in response to right thoracodorsal nerve injury.

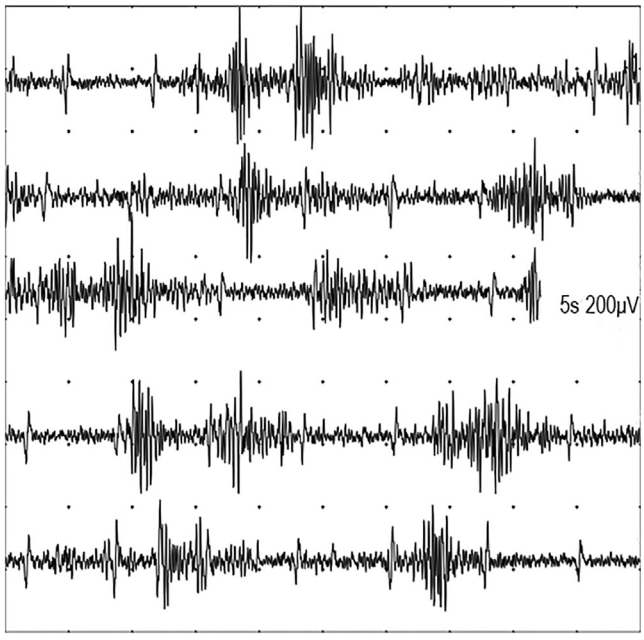

Fig. 1. Surface electromyography of the right latissimus dorsi muscle showing semi-rhythmic bursts of continuous discharges.

We offered the patient treatment with botulinum toxin injections and she accepted. Treatment with two injections of 20 units each of botulinum toxin type A (BTX-A) into the right latissimus dorsi muscle markedly reduced the continuous muscular contractions, which disappeared for eleven and half weeks. Her sensory symptoms did not show any therapeutic benefit. After the first injections, every three months, BTX-A injections were administered in the same way and the same favorable therapeutic benefit was obtained repeatedly.

3. Discussion

After posterolateral thoracotomy our patient developed two different neuropathic disorders. The first one to appear (just after surgery) was PTPS and the second one (six months after surgery) were jerks of the latissimus dorsi muscle. PTPS was well identified from the beginning and treatments were guided in order to mitigate neuropathic pain. On the other hand, no special attention was paid to the involuntary muscle contractions until three years after their onset. In retrospect we do not identify a clear reason for the belated attention paid to the latissimus dorsi muscle jerks. Between both iatrogenic neuropathic disorders (PTPS and the jerks) much more importance was given to PTPS. Both disorders should have been addressed simultaneously as soon as they coexisted. Also, during this period of time our patient had other numerous medical complaints related to her substantial medical history. Unfortunately, the delayed characterization of this involuntary movements impeded offering as soon as possible treatment with BTX-A injections.

PTPS appears frequently after thoracotomy and its chronicity and severity are related to the neuropathic damage caused during the intervention. As seen in our patient, the presence of pain and sensory symptoms located to the area corresponding to the damaged nerve can be enough for the diagnosis of intercostal neuropathy (Guastella et al., 2011). Clinical neurophysiology is also useful to support this diagnosis with dermatomal somatosensory evoked potentials and abdominal reflex electromyographic study (Benedetti et al., 1998). PTPS is very difficult to manage and its therapeutic approach is multimodal (Kelsheimer et al., 2019). Our patient was first treated with oral analgesia and capsaicin skin patches and did not report improvement until thoracic epidural block was performed, which was partially effective. Local injections of BTX-A may also be suitable for patients with focal painful neuropathies, where the extent of pain is limited – such as our patient's PTPS (Ranoux et al., 2008). In our case PTPS was relieved with thoracic epidural block before BTX-A was used, so we cannot assess if BTX-A injections played any role in reducing our patient's neuropathic chronic pain.

Six months after posterolateral thoracotomy and after the start of PTPS our patient presented a continuous involuntary jerk-like movement of the right latissimus dorsi muscle – its distribution was clearly defined by the right thoracodorsal nerve. Four years after these involuntary movements appeared, we proved that they showed clinical and electromyographic hallmarks of previously reported cases with appearance of jerks due to thoracodorsal nerve injury after thoracotomy (Aslam et al., 2009; Belluzzo et al., 2015; Carnero-Pardo et al., 1998; Hao and Clarke, 2002). The duration of these EMG discharges may range from 50 ms to 200 ms or, as it happens in our case, can even be longer. Sometimes these jerks are aggravated by voluntary contraction of the affected muscle. However, in our patient this was not clearly identified. The delay between surgery and the onset of myoclonus in our patient was

six months – consistent with the reported cases (usually between several days and several years). Longer latencies suggest more progressive compression. Once the neuroanatomical origin was known, the patient was offered treatment with BTX-A injections. She showed a successful response to periodic injections of BTX-A – latissimus dorsi jerks disappeared. BTX-A injections and surgical revision are the best therapeutic choices for jerk-like movements of peripheral origin (Lagueny et al., 2014). Surgical revision was not considered because our patient's EDS condition could lead to wound-healing problems (Malfait et al., 2010).

To our knowledge, to date, there have been no reports of a case with these two post-thoracotomy complications as described here (PTPS and post-thoracotomy jerks). The most remarkable diagnosis of our patient's medical history is EDS. Thereby our patient had probably greater predisposition of suffering iatrogenic nerve injuries. It is believed that EDS's genetic connective tissue defect is related to increased vulnerability of peripheral nerves to any mechanical stress (Voermans et al., 2006). In any case, these nerve injuries, either separately or together, may occur during posterolateral thoracotomies. In order to prevent them, the moments of the procedure in which nerve structures are at risk should be identified (Rogers et al., 2002). Hence, we believe these surgeries should be performed under intraoperative neurophysiological monitoring.

We want to highlight that although the symptomatology of intercostal and thoracodorsal neuropathy seen in our patient involved the same body area, this should not have obscured the diagnosis approach of the involuntary jerk-like movements. Our patient did not present with different symptoms within the same clinical picture. She presented with two different pathologies, each one with its own pathophysiology, onset latency and specific clinical features. As shown in our clinical case, the effective treatment was then specific for each disorder. Presumably, if the patient had been referred to Clinical Neurophysiology as soon as the jerks appeared the different neuroanatomical origin of each of these two neuropathic disorders (peripheral origin jerks and PTPS) would have been identified earlier. Clinical Neurophysiology tests allow evaluation beyond physical examination and lead to a correct categorization of neuropathic disorders – so neurophysiological tests provide guidance to specific treatment approaches (Caviness, 2019; Merchant et al., 2020).

Informed consent

A written informed consent was obtained from the patient for the publication of the video in which she appeared.

Author contributions statement

S.C-S and T.B-H performed drafting and revising of the manuscript. All authors contributed to the data acquisition and approved the final manuscript.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.cnp.2021.06.003.

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