

THORACIC LONGUS NERVE LESION IN A PATIENT WITH ANKYLOSING SPONDYLITIS

Vesna Budišin¹, Marijan Cesarik², Davorka Rosić³, Borislav Radić¹, Marica Topić³

¹Polyclinic Medikol, Mandlova 7, 10000 Zagreb, Croatia

²General County Hospital Požega, Osječka ul. 107, 34000 Požega, Croatia

³Special hospital for medical rehabilitation Lipik, Ul. Marije Terezije 13, 34551 Lipik, Croatia

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ABSTRACT

The paper presents a 33 - year - old patient who has been suffering from ankylosing spondylitis since the age of 28. Pain in his right shoulder and weakness in his right arm developed after more hours of walking with a backpack. The performed procedure diagnosed a lesion of the long thoracic nerve without of sensory damage. Peripheral nerve injuries long thoracic nerve lead to weakness of the muscles -serratus anterior muscle and result in a protrusion of the medial side of the scapula (scapula alata). It is difficult to raise the arm in the shoulder joint above the horizontal line, that is to raise the shoulder from the chest when the arm is extended and pressed against a fixed object in front of the patient. Due to its long, relatively superficial course, long thoracic nerve is susceptible to injury, either through direct trauma or stretching. The long thoracic nerve, also called Charles Bell's external respiratory nerve, is a rare isolated nerve damage. The nerve is often injured from carrying a load on his shoulder, with supraclavicular and axillary injuries, blows in the neck area. Injury has been reported in almost all sports, usually occurring from a blow to the ribs with an outstretched arm. Long thoracic nerve can be damaged during breast cancer surgery, especially radical mastectomy that involve the removal of axillary lymph nodes. It is a common lesion in spinal surgeries.

Key words: Lesion, long thoracic nerve, ankylosing spondylitis

Correspondence person:

Vesna Budišin, MD

E-mail: vesna.budisin@medikol.hr

INTRODUCTION

Nervus thoracicus longus (C5 to C7), isolated nerve damage is rare. Due to its long, relatively superficial course, n. thoracicus longus is sensitive to injury. Possible mechanisms of injury are trauma from supraclavicular and axillary blows (injury), (1), heavy loads on the shoulder such as carrying heavy loads in construction workers (2), constant maximum extension from the shoulder with lifting of the upper limb as in many sports: weight lifting (3), golf, tennis, archery (4-6). Nervus thoracicus longus can be damaged during many operations, after breast amputation (especially radical mastectomy that includes the removal of axillary lymph nodes), after thoracotomy (7), during surgery of the front part of the spine that caused an external compressive force on the nerve (8).. Until nerve lesions can occur as a complication of parvovirus infection (9), with compression of the C7 spinal root (10) and idiopathically (11). Damage to the n. thoracicus longus and n. phrenicus after neck chiropractic manipulation (12).

Lesion n. thoracicus longus is manifested by paresis of the serratus anterior muscle and the appearance of a winged scapula - scapula alata (the medial edge of the scapula on the paretic side is located closer to the spine and protrudes from the chest like a

wing). In paresis of the serratus anterior muscle, the scapula moves away from the back. It is difficult to raise the arm in the shoulder joint above the horizontal line, that is, to raise the scapula from the chest when the arm is stretched out and pressed against a fixed object in front of the patient. In the case of an isolated lesion of the thoracic longus nerve, there is no loss of sensation. Paresis is more common on the right side of the body (13).

Ankylosing spondylitis is a chronic, progressive inflammatory rheumatic disease, which affects the sacroiliac joints and the spine with all associated connective structures, costovertebral and costotransverse joints. It often affects the hips and shoulders, less often peripheral joints, and some visceral organs.

Neurological complications are common in inflammatory rheumatic diseases, and peripheral neuropathies are one of the neurological manifestations. Peripheral neuropathies represent a heterogeneous complex of diseases that arise as a result of damage to the peripheral nervous system.

The aim of this work is to present a patient who, after physical load and effort, had an isolated lesion of the n. thoracicus longus and the patient also has an inflammatory rheumatic disease, ankylosing spondylitis.

CASE REPORT

A 33-year-old patient was diagnosed with ankylosing spondylitis (B 27 positive) when he was 28 years old. The mother is being treated for seronegative spondyloarthropathiae (HLA B 27 positive) and the father is a carrier of the positive HLA B 27 antigen. The patient has no comorbidities.

After several hours of walking on uneven and hilly terrain with a backpack weighing about 10 kg, which he carried over his right shoulder, a patient with a medium osteomuscular build developed pain in his right shoulder, with difficulty in mobility in the shoulder joint with a protrusion of the scapula. The patient works as a merchant and was lifting heavy loads at work.



Picture 1. Scapula alata – winged scapula in lesion of n. thoracicus longus

During the clinical examination, the patient complained of difficulty in raising the right

arm above the horizontal line, during which the scapula protruded from the thoracic wall (winged scapula - scapula alata).

An X-ray examination of the cervical and thoracic spine and the right hemithorax was performed, the findings were normal. X-ray and ultrasound of the right shoulder were also normal. MR findings of the cervical spine were normal.

In our patient, electromyoneurography (EMNG) of n. thoracic longus, which showed that it was a severe neurogenic lesion of the nerve, and electromyography (EMG) showed paralysis of the n. serratus anterior. Treatment was carried out using physical therapy: individual kinesiotherapy (shoulder range of motion exercises, exercises to strengthen the serratus anterior muscle and other muscles that stabilize the scapula: rhomboideus major and minor, levator scapulae and trapezius muscles).

We also used electrostimulation to strengthen the mentioned muscles. From electroanalgesic therapy to reduce pain we used TENS - transcutaneous electrical nerve stimulation.

Due to his rheumatic disease of ankylosing spondylitis, which affected the axial skeleton (the entire spine and sacroiliac joints), the patient was treated with the antirheumatic drug ibuprofen in doses that

depended on the inflammation and pain. After the lesion of n. thoracicus longus, in addition to ibuprofen, he also took the opiate tramadol for pain.

Physical therapy procedures are repeated several times over a period of up to 2 years, during which a possible recovery of the nerve is expected.

Electroneuromyography n. thoracicus longus was performed several times in a period of up to 2 years and showed that there was no recovery of the nerve n. thoracicus longus, because it was a more serious nerve injury - neurotmesis, which has a worse prognosis.

The patient worked as a merchant, and since he could no longer do his job as a merchant, he completed his education and continued to work on a computer.

DISCUSSION

N.thoracicus longus (NTL), long thoracic nerve, so named because of its length of up to 25 cm. It was first described and called the external respiratory nerve by the Scottish surgeon Charles Bell (1774-1842). The nerve starts from the anterior ramus of three spinal nerves of the cervical spine, C5, C6 and C7. The long thoracic nerve is formed as the upper part originates from the C5 and C6 roots, and the lower part comes from the C7 root. The roots from C5 and C6

pass through the scalenus medius muscle, while the C7 root passes in front of the muscle. The joining of these two parts takes place in the axilla. The nerve descends behind the brachial plexus and the axillary artery and vein, lying on the outer surface of the serratus anterior muscle. It runs along the side of the chest to the lower border of this muscle, supplying fibers to each of its branches. The long thoracic nerve descends downward and passes anterior to the scalenus muscle, descends distally and laterally deep to the clavicle and superficial to the first and second ribs, and then descends down the chest wall along the mid-axillary line on the outer surface of the serratus anterior muscle (14, 15). The long thoracic nerve is a motor nerve and innervates the serratus anterior muscle. The name serratus comes from the Latin term serrare (saw) because it resembles a saw, as it consists of 8 to 10 separate slides that connect the lower surface of the inner edge of the scapula with each of the ribs. Serratus anterior connects the lower surface of the inner edge of the scapula with the rib cage.

M. serratus anterior, the "serrated muscle", starts from the first 8 to 10 ribs and attaches to the medial edge of the scapula. From an



Picture 2. Scapula alata – The protrusion of the scapula is best seen when the arm is extended forward

anatomical point of view, the muscle can be divided into the upper part, which is supplied with C5 and C6 fibers, and the lower part, which is supplied with C7 root fibers. The upper part is responsible for scapular protraction (moving the scapula forward along the thoracic wall), and the lower part for scapular stabilization (15, 16).

M. serratus anterior, known as the "boxer's muscle", acts to pull the scapula forward towards the chest, enabling anteversion of the arm and raising the ribs, helping in breathing, so it is also responsible for the anterolateral movement of the scapula, which enables raising the arms. Weakness or paralysis of the serratus anterior muscle results in a "wing" or bulging "fluttering" of the scapula (scapula alata). As the serratus anterior muscle under normal

circumstances pulls the scapula forward, the weakness of the muscles results in the movement of the lower edge of the scapula closer to the spine, resulting in protrusion of the medial side of the scapula (16, 17).

The first description of the scapular wing was published by Dr. M. Winslow in 1723 from France (18). N.thoracicus longus and its blood supply are sensitive to both compression and stretching of the anterior part of the lower segment of the scapula. The subscapular artery through the inferior angle of the scapula supplies blood to the greater part of the anterior serratus muscle, as well as to the peripheral part of the thoracic nerve longus.

Nerve damage can occur after poor epineuria blood flow as a result of compression or stretching of the subscapular artery. N.thoracicus longus and its blood supply are anchored in the serratus



Picture 3. Scapula alata - it is less visible when the hand is held at the side

anterior muscle, and together with the muscle participate in the movement of the muscle. With arm adduction, the scapula moves forward, simultaneously bringing the nerves and blood vessels forward, out of the way of the scapula. During arm extension, the serratus anterior muscle prevents the winging of the scapula, preventing stretching of the nerves and blood vessels (17). The anterior serratus muscle is innervated exclusively from the thoracic longus nerve, which after damage results in muscle paralysis, a phenomenon known as the winged scapula. The serratus anterior is primarily responsible for adducting the scapula, a movement that occurs when executing a kick. The serratus anterior works together with the trapezius muscles to allow the continuous upward rotation of the scapula, which is necessary to raise the arm overhead (19).

Compared with other nerves of the brachial plexus, the long thoracic nerve has a relatively smaller diameter which, together with its minimal connective tissue and superficial course along the surface of the serratus anterior muscle, increases its susceptibility to operative or nonoperative damage with subsequent development of the scapular wing. The long thoracic nerve travels to the level of the eighth or ninth rib and is relatively unprotected from the axilla downward. As such, the nerve can suffer

exposure to external injuries, through direct external trauma or compression (carrying loads on the shoulders such as a backpack, heavy wood...). and arm-raising shoulder extension caused by athletics, golf, tennis, weightlifting (20, 21).

Passive arm extension and uncoordinated scapular movements during sports, such as a sudden shoulder snatch, can stretch the long thoracic nerve and its blood vessels, resulting in a winged scapula. It has been observed that with increasing proximity of the thoracic longus nerve to the lower corner of the scapula, less scapular movement is required to compress or stretch the nerve (17, 21).

Procedures in the thoracic region, including radical mastectomy, transthoracic sympathectomy, transaxillary thoracotomy, misplaced intercostal drain, axillary lymph node dissections, thoracic catheter placement, first rib resection, increase the risk for proximal injury to the long thoracic nerve. Understanding the anatomical course of the thoracic longus nerve is important for reducing the risk of unintentional nerve damage and the success of the surgical procedure (21).

Physiological variations in the course of the long thoracic nerve are positioned on the median scalene muscle and can lead to iatrogenic nerve injuries and the formation

of "wings" of the scapula, because the nerve roots can be located anteriorly, posteriorly, or across the median scalene muscle. With the formation of the winged scapula, the serratus anterior muscle does not function synchronously with the other muscles of the shoulder girdle (22).

In our patient, electromyoneurography (EMNG) was performed, which showed that it was a more severe neurogenic lesion of the thoracic longus nerve, and electromyography (EMG) showed paralysis of the n. serratus anterior.

In case of damage to the n. thoracic longus, abnormalities are limited to m. serratus anterior. Other muscles of the upper extremity were also examined, which receive the same innervation from the C5, C6, C7 roots (m.biceps, m.deltoideus, m.supraspinatus, m.infraspinatus, m.triceps, m.pronator teres) cervical radiculopathy was excluded, brachial plexopathy and affection of other proximal nerves. The cervical paraspinal muscles were also examined, a proximal lesion of the cervical roots was excluded (23, 24).

Peripheral nerves are divided into three groups according to the severity of the damage: neuropraxia, axonotmesis and neurotmesis.

Neuropraxia is a disorder of the peripheral nervous system in which there is a temporary loss of motor and sensory function due to blocked nerve conduction, usually lasting six to eight weeks before full recovery. This condition is caused by blunt nerve injury due to external blows or impact injuries to the muscle fibers and nerve fibers leading to repeated or long-term accumulation of pressure on the nerve. As a result of this pressure, ischemia occurs, neural lesions occur, and the human body naturally reacts with edema that extends in all directions of the pressure source. Microscopic evidence shows damage to the myelin sheath but not to the axon. Therefore, distal nerve fibers do not degenerate, and myelin damage can be repaired (23, 24).

Almost all cases of neuropraxia can be completely treated by non-operative means. In cases of neuropraxia, nerve function is temporarily impaired. Recovery begins within two to three weeks after the injury and ends within six to eight weeks. (23, 25).

Axonotmesis is characterized by the interruption of the axon, but the sheath is preserved, the surrounding connective tissue around the axon is preserved. The distal part of the axon degenerates and sometimes it is possible to achieve recovery with conservative treatment, which requires

treatment for up to 18 months, depending on the height of the lesion. These injuries heal about 1 mm/day, slower than neurapraxia, but recovery is possible.

Neurotmesa (greek tmesis means "cutting") is the most serious nerve injury. In this type of injury, both the nerve and the nerve sheath are damaged. Although partial recovery may occur, full recovery is impossible. Neurotmesa occurs in the peripheral nerves and most often in the upper extremities, and accounts for 73.5% of all cases of peripheral nerve injuries (25).

Injuries resulting from repetitive activity or carrying heavy loads are incomplete and generally resolve spontaneously within 6 to 24 months (25). In these cases, physical therapy and exercise are crucial for maintaining the range of motion and strengthening the surrounding muscles (26).

Therapeutic procedures are to strengthen the serratus anterior muscle and other muscles that stabilize the scapula (m. rhomboideus major et minor, m. levator scapulae and m. trapezius), thereby reducing muscle atrophy and shoulder progression towards shoulder instability. Electrostimulation is used to improve muscle strength. From electroanalgesic therapy to reduce pain, we used TENS - transcutaneous electrical nerve stimulation.

Patients who have nerve damage, neurotmesis, often face a poor prognosis, and the damaged nerve never fully recovers. If the recovery is weak, a surgical procedure can be applied, transplanting nerves from another part of the body. Surgical operations give people a better chance of regaining certain body functions. (27).

In the last few years, successful surgical procedures have begun to be performed, which perform endoscopic minimally invasive neurolysis of the thoracic segment of the long thoracic nerve, with the assistance of a robot (28).

In our patient suffering from inflammatory rheumatic disease, ankylosing spondylitis, the lesion of the n. thoracic longus in the period of 2 years, by which time it is expected that the nerve could recover. Electroneuromyography showed that our patient had a severe nerve injury - neurotmesis.

Neurological complications are common in inflammatory rheumatic diseases, and peripheral neuropathies are one of the neurological manifestations. Peripheral neuropathies represent a heterogeneous complex of diseases that arise as a result of damage to the peripheral nervous system.

The general clinical classification of neuropathy is polyneuropathies (when several nerves are affected),

mononeuropathies (when one nerve is affected) and mononeuritis multiplex or multifocal mononeuropathy when two or more isolated nerves in different parts of the body are affected. Of all forms of peripheral neuropathies, mononeuritis multiplex is considered a specificity of inflammatory rheumatic diseases because according to some analyses, 31% of patients with this form of PN were diagnosed with rheumatic disease (29).

Peripheral neuropathies are most often found in systemic lupus erythematosus (SLE), systemic vasculitis, rheumatoid arthritis (RA), and primary Sjögren's syndrome (pSS) (26, 28). They can also be found in systemic sclerosis, a mixed connective tissue disease (29, 30).

The nervous system is most often affected in vasculitis of medium-sized blood vessels (for example, polyarteritis nodosa), but it often appears in other vasculitis as well. The frequency of peripheral neuropathies in patients with polyarteritis nodosum is up to 70%. The disease can initially be presented with peripheral neuropathy, which most often affects the legs. The onset is very acute, pain and paresthesias appear in the area of the affected nerve in a very short time (several hours), motor disturbances develop up to the complete loss of nerve function. Nerve damage is the result of involvement of the vasa venorum and

consequent ischemia and nerve infarction. (30).

According to a retrospective analysis of 1533 patients with SLE, damage to the peripheral nervous system is present in about 14% of patients (31). The most common diagnoses are sensorimotor and pure sensory polyneuropathy (56%), as well as motoneuritis multiplex (9%), where asymmetric manifestations predominate and the distal (sural and peroneal) nerves of the lower extremities are affected (31).

Damage to the peripheral nervous system is best studied in primary Sjögren's syndrome, where sensory polyneuropathy dominates, but sensorimotor polyneuropathy and mononeuritis multiplex are also present (32). In 25% of patients with Sjögren's syndrome, polyneuropathy is the leading or first clinical manifestation of the disease (33).

In rheumatoid arthritis, PNs are usually associated with nerve compression by rheumatoid nodules, swollen synovium, aponeurosis, bony exostoses or vasculitis. Peripheral neuropathies are manifested in RA as entrapment neuropathies, motoneuritis multiplex, sensory-motor and sensory neuropathy, and motor neuropathy is not rare either (34).

Peripheral neuropathies are found in many inflammatory rheumatic diseases, even in

ankylosing spondylitis (35). Gündüz and associates conducted a study on the frequency of peripheral neuropathy in patients with ankylosing spondylitis, without a single symptom of neuropathy, but it was shown that peripheral neuropathy can also be present in asymptomatic patients with this disease (36).

CONCLUSION

A lesion of the n. thoracicus longus occurred in our patient with inflammatory rheumatic disease, ankylosing spondylitis, after carrying a load on the right shoulder. As it was a case of severe nerve damage - neurotmesis, even after two years of treatment, the nerve did not recover.

In patients with ankylosing spondylitis, there are several studies in the literature that have investigated the relationship between peripheral neuropathy and ankylosing spondylitis, but further studies with larger samples and longer disease duration are awaited to confirm the results and unravel its clinical relevance. We cannot speak with certainty about the connection of the lesion n. thoracicus longus with ankylosing spondylitis, but our patient could have a predisposition for the development of peripheral neuropathy through his inflammatory rheumatic disease, considering that PN are found in many inflammatory rheumatic diseases.

In severe nerve damage, as in our patient with a lesion of n. thoracicus longus the current treatment is focused on new ways of nerve regeneration and new surgical techniques that have been successfully performed in recent years with the help of robots.

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LEZIJA NERVUS THORACICUS LONGUSA KOD BOLESNIKA S ANKILOZANTNIM SPONDILITISOM

Vesna Budišin¹, Marijan Cesarik², Davorka Rosić³, Borislav Radić¹, Marica Topić³

¹Poliklinika Medikol, Mandlova 7, 10000 Zagreb, Republika Hrvatska

²Opća županijska bolnica Požega, Osječka ul. 107, 34000 Požega, Republika Hrvatska

³Specijalna bolnica za medicinsku rehabilitaciju Lipik, Ul. Marije Terezije 13, 34551 Lipik, Republika Hrvatska

SAŽETAK

U radu je prikazan bolesnik star 33 godine, koji od 28 godine života boluje od ankiлоzantnog spondilitisa. Bol u desnom ramenu i slabost desne ruke razvio je nakon više sati hodanja s naprtnjačom. Učinjenom obradom dijagnosticirana je lezija nervusa thoracicus longusa bez ispada osjeta. Periferne ozljede živca n. thoracicus longusa dovode do slabost mišića seratus anteriora a za posljedicu imaju izbočenje medijalne strane skapule (scapula alata). Otežano je podizanje ruke u ramenom zglobu iznad horizontalne linije, odnosno podizanje lopatice od grudnog koša kada je ruka ispružena i pritisnuta o fiksirani objekt ispred bolesnika. Zbog svog dugog, relativno površinskog tijeka, n. toracikus longus osjetljiv je na ozljede, bilo kroz izravnu traumu ili istežanje. Nervus thoracicus longus, koji se naziva i vanjski respiratorni živac Charlesa Bella, rijetko je izolirano oštećenje živca. Često je povređen pri nošenju tereta na ramenu, kod supraklavikularnih i aksilarnih povreda, udaraca u području vrata. Ozljeda je zabilježena u gotovo svim sportovima, obično se pojavljuju od udarca u rebra pri ispruženoj ruci. Dugi torakalni živac može biti oštećen tijekom operacije raka dojke, posebno radikalne mastectomije koje uključuju uklanjanje aksilarnih limfnih čvorova. Česta je lezija kod operacija kralješnice.

Ključne riječi: lezija, nervus thoracicus longus, spondylitis ankilosans

Kontakt osoba:

Vesna Budišin, dr. med.

E-mail: vesna.budisin@medikol.hr