Scandinavian Journal of Pain 3 (2012) 238–240

Contents lists available at SciVerse ScienceDirect

Scandinavian Journal of Pain

journal homepage: www.ScandinavianJournalPain.com

Educational case report

Treatment of post-traumatic pain, and autonomic and muscular dysfunction by ganglion impar block and medial branch block of the facet joints: A case report

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ARTICLE INFO

Article history:
Received 11 May 2012
Received in revised form 11 July 2012
Accepted 13 July 2012

Keywords:
Autonomic dysfunction
Facet joints
Ganglion impar
Case report
Whiplash
Pain

ABSTRACT

Introduction: Patients exposed to whiplash trauma are at risk of developing pain and dysfunction of the neck and shoulder. Although rarely discussed in the literature, some patients also develop autonomic dysfunction.
Case presentation: A previously healthy 41-year-old woman was involved in a "head-on" car crash. During the following 3 years she developed severe and complex post-traumatic pain syndrome, which consisted of neck pain, lumbar pain, sensory-motor dysfunction, and myoclonic muscular contractions. Despite pharmacotherapy, physiotherapy, and rehabilitation, her condition worsened, resulting in severe disability. Fourteen years after the car crash, an interventional pain therapy program was started, which consisted of sympathetic ganglion impar block and medial branch blocks of facet joints at different levels. These treatment strategies ultimately normalized her sensory-motor dysfunction, reduced her autonomic dysfunction, and stopped the myoclonic muscular contractions.
Conclusion: This case highlights a possible interaction between the pain-generating facet joints, the somatosensory nervous system, and the autonomic/sympathetic nervous systems. The case also highlights the importance of identifying autonomic dysfunction in patients with persisting pain syndromes.
Implications: This complex case shows that many clinical phenomena cannot be explained using our present knowledge of pain mechanisms. We hope that readers who have observed similar cases can learn from our case, and are encouraged to publish their observations.

1. Introduction

Persisting pain after neck trauma is an important public health problem. However, there are several unanswered questions regarding the mechanisms causing the injury, the clinical symptoms, radiological findings, and alternative treatments. These issues were recently discussed in a "State of the art" supplement in the journal *Spine* [1].

Reports describing the interaction between the somatosensory and the autonomic nervous systems in persisting post-traumatic pain syndrome are rather sparse. However, this interaction was discussed by Passatore and Roatta [2], who used whiplash trauma as a model to describe autonomic dysfunction and persisting pain. Some reports have also described the effects of different types of blockade on complex regional pain syndromes (CRPS), which further strengthens the theory of a possible relationship between the autonomic nervous system and persisting pain [3–7].

2. Case presentation

A 41-year-old previously healthy female was involved in a "head-on" collision. While she was driving at about 60 km/h, another car heading towards her lost control on a curve of the road and hit her car. She complained of neck pain, radiating pain affecting her right arm, and headache after the crash. She also developed partial post-traumatic amnesia. Upon returning to work as an assistant nurse, all of her symptoms worsened. Over several years, she was examined repeatedly by several different specialists, who recommended various pharmacotherapies, including gabapentin, amitryptiline, and opioids, to manage her symptoms. Her problems continued to worsen, as her pain spread from the neck/arm region to the lower thoracic and lumbar region. The pain also radiated out to her right leg, which impeded walking.

By 2 years after the crash, the patient developed myoclonic muscular contractions, which progressively increased in frequency, eventually resulting in severe disability. The myoclonic muscular contractions started suddenly, initially in the areas of pain (e.g., the lumbar region, leg, and shoulder), and then spread to the rest of her body. She lost all motor control during the contractions, but maintained consciousness. Repeated electroencephalography,

DOI of refers to article: http://dx.doi.org/10.1016/j.sjpain.2012.09.003.
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1877-8860/$ – see front matter
http://dx.doi.org/10.1016/j.sjpain.2012.07.002

psychological examinations, and other tests were normal, resulting in divergent opinions between psychiatrists/psychologists and neurologists. The patient's clinical findings raised the question of whether she had a dissociative or somatoform syndrome, or a neurological disorder. However, the results of clinical examinations did not seem to support dissociative or somatoform syndromes. Anti-epileptic drugs and benzodiazepines were ineffectual, as were physical rehabilitation strategies.

Fourteen years after the car crash the patient was referred to The Pain Clinic, Umeå, for an interventional pain therapy program with diagnostic blocks and treatments. At presentation, she reported having widespread problems. She reported having a constant pressure in the neck that radiated to the lateral aspect of the head on both sides, and then forwards to the eyes. She also complained of pain in the upper part of the thoracic spine that radiated out to the shoulders, the lateral part of the upper arms, the dorsal aspect of the forearms, and fingers 1, 4, and 5. She suffered from pain in the lumbar region, which radiated out to the dorsal part of both hips and then down the outsides of both legs, including the lateral part of both feet. She also complained of numbness in both arms and hands. She could do little without triggering pain and contractions; a reduction in her activity did not attenuate her symptoms.

She also reported symptoms consistent with autonomic dysfunction. For example, her hands were swollen and hot, and her sudomotor activity was increased. Her feet were swollen, cold, and cyanotic in colour. She also had problems urinating. There was a temperature difference between left and right foot. Her muscles were constantly in a state of tension, which transformed into contractions several times each day. She experienced tachycardia and fever during these contractions.

Her life was limited by her constant need for assistance because she could not control, on her own, the dystonia and subsequent contractions. During the attacks, she had difficulty breathing and could not talk. Additionally, she usually fell to the floor during each attack. She received care from her husband overnight and from a full-time nurse during the day. Her social life was therefore very limited.

The initial interventions focused on trying to locate the cause of the lumbar pain, because the myoclonic muscular contractions were considered too difficult to understand or to treat. Starting caudally on the right side, medial branch blocks (MBB) were performed at the levels of L5 and L4 with 5 mg/ml bupivacain (1 ml per nerve). These injections, however, triggered myoclonic muscular contractions in the right leg. After extending the MBB to the levels L3, L2, and L1, and finally to Th12, the contractions disappeared. This was the first time in many years that the patient experienced some relief from the whole-body muscle tension. Since 80% of her lumbar pain disappeared following administration of the local anaesthetic, the test was repeated for confirmation. Although the block induced transient myoclonic muscular contractions, as before, her pain improved during the repeated MBB procedure. Therefore, radiofrequency denervation of the medial branches at L2–L5 was performed.

Six months later, the lumbar pain was approximately 50% of the pre-treatment level. However, towards the end of this period, her symptoms affecting the right leg started to deteriorate, with loss of coordination and strength. The leg was also cold, suggestive of autonomic dysfunction. Similar but less severe symptoms were also affecting her left leg. Based on these symptoms, we performed a sympathetic block. Since the problems affected both legs and because she also complained of pain when sitting, we started with a ganglion impar block. This was performed with 4 ml of 5 mg/ml bupivacaine [5]. Although the patient did not consider the injection painful, the block triggered myoclonic muscular contractions that spread throughout the body. According to the patient's nurse, the contractions triggered by this block were similar to the patient's regular contractions. Within 10 min, the contractions had diminished and only involved the neck and face. At this time, the patient could speak a little, and said that the contractions in the head and neck could persist for several hours, which was confirmed by the patient's nurse.

From these findings, we suspected that the contractions were modulated by the sympathetic nervous system (the ganglion impar in this case) as well as the facet joints and medial branches, since a similar reaction was observed after MBB. Because we were able to stop the myoclonic muscular contractions originating in the lumbar region by performing MBB there, we decided to perform similar blocks in the cervical region.

While the patient was experiencing myoclonic muscular contractions in the face, neck, and arms, she was placed on the examination table, in a side position with her right side uppermost. MBB of C3, the third occipital nerve, C4, and C5 was performed under fluoroscopic control. The contractions stopped once the local anaesthetic started to act, and the patient could move and talk without any problems. She reported that her contractions had never diminished this rapidly before.

The patient did not experience any contractions for 3 days after the procedure. At an examination several weeks later, she was hopeful of becoming free of the contractions. The MBB in the neck was repeated but now these injections triggered the contractions. Ganglion impar block was repeated because the combination of MBB at C3–C5 and ganglion impar block stopped the contractions at previous visits. This procedure stopped her myoclonic muscular contractions and pain for 5 days – the first such relief in 15 years.

When she visited the clinic 1 month later, a third test was performed. Ganglion impar block trigged the contractions again, while MBB at levels C3–C5 stopped the contractions. Therefore, we performed radiofrequency denervation of the medial branches C3–C5. Following this procedure, the general myoclonic muscular contractions that previously affected both sides of her body disappeared completely, although she occasionally developed contractions affecting the left side of her body, including the face, neck, arm, and leg.

Subsequent studies revealed the involvement of different pain generators in the lumbar, thoracic, and cervical facet joints on the left side. These were treated by radiofrequency denervation, which subsequently reduced the contractions.

Since performing radiofrequency denervation of the left MB at C3–C5 in August 2007, the patient remained free from contractions until her follow-up in June 2011. The patient reported that, when she is active and triggers pain in the spine, she can feel something happening in the muscles, but this does not progress to myoclonic muscular contractions.

At her most recent follow-up in February 2012, she reported that triggering pain, mainly in the lumbar region, sometimes resulted in myoclonic muscular contractions in the right leg. However, she could control them by using analgesics and relaxing in an ergonomic chair, which was not possible before treatment. Her need for nurse care had reduced from 40 to 16 h each week, and she could perform light work in the garden, and take care of herself and the household in a manner that she had not been able to do for many years.

The spinal interventions (i.e., MBB, radiofrequency denervation, nerve root blocks, and sympathetic blocks) were performed over a period of 4 years (from February 2005 to April 2009). A total of 43 nerve blocks and 8 sessions of radiofrequency denervation at different levels were performed during this time, involving joints C2–C3, C7–Th2, and Th9–S1 on the left side, and C2–C7, and Th12–S1 on the right side. Before the interventions, her pain varied between 1.6 and 9.1 on a visual analogue scale (VAS). After the nerve blocks, she reported that her pain level had reduced by at least 80% on two different tests before radiofrequency denervation was considered. The

patient has not reported any side effects of these treatments. Three years after the last intervention, her EQ-5D (time trade-off index) and EQ-VAS were 0.691 and 0.5, respectively, which are slightly lower than the mean scores in Swedish women of her age [8].

3. Discussion

With this case we would like to describe a possible interaction between the somatosensory pain and the autonomic nervous systems, which may evoke different and sometimes obscure symptoms [9]. We would also like to emphasize the importance of carefully evaluating the patient's descriptions and experiences, as this is essential to fully understand the magnitude of the patient's problems.

Our patient developed persisting pain, sensory-motor dysfunction, myoclonic muscular contractions, and autonomic dysfunction after a car crash. The development of symptoms illustrates the situation discussed by Passatore and Roatta, where symptoms can develop a long time after the accident [2]. Of course, the casual relationship is uncertain and impossible to prove. Nonetheless, prior cases with similarly strange reactions to treatments have been reported [10]. In our case, the onset of myoclonic muscular contractions and autonomic dysfunction had a devastating effect on the patient's daily activities, and rendered her dependent on constant assistance.

After the patient's initial presentation, her clinical status deteriorated rapidly, but allowed us to perform elaborate therapies. It is well known that the facet joints are particularly vulnerable to injury after trauma, especially during car crashes causing whiplash [11,12]. The facet joints are richly innervated and are close connected to the sympathetic nervous system [13–15]. Thus, we considered that the patient's symptoms affecting her limbs could be due to sympathetic dysfunction, similar to some types of CRPS [16]. A positive feedback system, as proposed by Passatore and Roatta [2], could be responsible for her syndrome. In this way, pain originating in the facet joints leads to an increase of sympathetic outflow causing muscular contractions, sensitization of the nociceptive pathways, and increased pain (see Fig. 6 in [2]). This could also explain why MBB or sympathetic block triggered the muscular contractions, and why the symptoms that were more recently reported by the patient (i.e., raised muscular activity) could be stopped with analgesics.

Our working hypothesis was that the interaction between the persisting pain and the pain in the facet joints caused abnormal activation and dysfunction of the sympathetic nervous system. Even if we do not fully understand the underlying mechanisms, there was a clear time-dependent relationship between the intervention (i.e., radiofrequency denervation of the MB stopped the painful stimuli arising in the facet joints) and the patient's recovery, which may support the hypothesis.

Of course, we cannot ignore the possibility that the patient experienced a psychological problem, such as "somatization." However, our clinical perception during the 4 years of repeated examinations and treatments did not support such possibilities. Instead, we rather agree with Merskey's conclusion in a Commentary in *Pain* that "*. . . notions of somatization (and also of the DSM-IV idea of a pain disorder) increasingly lack validity*" [17].

4. Conclusion

This case highlights a possible interaction between the pain-generating facet joints, the somatosensory nervous system, and the autonomic/sympathetic nervous systems. The case also highlights the importance of identifying autonomic dysfunction in patients with persisting pain syndromes.

5. Implications

This complex case shows that many clinical phenomena cannot be explained with our present knowledge of pain mechanisms. We hope that readers who have observed similar cases can learn from our case, and that they are encouraged to publish their observations.

Consent

Written informed consent was obtained from the patient for publication of this case report. A copy of the written consent is available for review by the Editor-in-Chief of this journal. The patient has read the article and did not wish to add further details.

Competing interests

The authors declare that they have no competing interests.

Authors' contributions

JH analysed and interpreted the patient data, performed the medical examinations and interventions and wrote the manuscript. HW reviewed the paper. Both authors read and approved the final paper.

Acknowledgement

We wish to thank Sherdil Nath, MD, FRCA London, for initial discussions concerning this case in the early intervention period.

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