A case report of postulated ‘Barré Liéou syndrome’

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Abstract

The case history presented is of a 32 year old woman suffering with severe occipital and bilateral temporal pain together with autonomic disturbances affecting her vision, balance and breathing, symptoms which have been postulated as ‘Barré Liéou syndrome’. She complained of pain referred to the left arm and associated circulatory and sensory disturbance in keeping with the diagnosis of complex regional pain syndrome type I. Traditional Chinese and Western trigger point acupuncture techniques were used in order to treat her pain and autonomic dysfunction. Acupuncture was successful in reducing, but not totally alleviating, her pain, and was particularly effective in reducing the majority of autonomic symptoms.

Keywords

Acupuncture, sternocleidomastoid, Barré Liéou Syndrome, autonomic disturbance, trigger points.

Description of the case

The patient is a 32 year old woman who presented for treatment for physiotherapy, having been referred by her GP with severe occipital pain, visual disturbances, autonomic disturbances and breathing difficulties following a minor fall onto the left outstretched arm some two years previously. At the age of ten she fell out of a tree and fractured her skull, remaining unconscious for several weeks, but making a full recovery with no detected injury to the cervical spine. A recent x ray film showed evidence of loss of disc space at the C3/4 level. She was unaware of any damage or symptoms in her neck until the minor fall. She worked as a complaints clerk in a furniture business spending up to six hours a day at a computer. Her symptoms started with a feeling of constriction around her throat, occipital and temporal headaches, pain referred to the left arm, paraesthesiae along the distribution of the C6 nerve root and circulatory disturbances. Gradually she began to develop breathing difficulties especially on lying flat, severe nocturnal sweats confined to the left side of the face and arm, and an overwhelming physical inability to cope with even minor chores. She was referred to the Chronic Pain Clinic for a diagnostic block of the C6 nerve root. This reduced some of the referred pain but had little effect on the autonomic dysfunction, headaches or throat symptoms. Over the last 18 months she had been referred to two ENT specialists who could find no abnormality within the throat. She had received chiropractic and osteopathic treatment with little effect. She was referred for a chronic pain management protocol and acupuncture.

Symptoms

The patient reported a feeling of helplessness and loss of control, with impaired memory and cognitive skills. She was unable to work and had difficulty looking after her two children. She had nocturnal pain and insomnia. She had lost her voice, and had increasing difficulty in swallowing food because of a sore, swollen tongue (which had a thick black coating). She suffered with nausea, loss of taste and poor appetite. Her husband reported concern regarding her breathing at night – she appeared to stop breathing for several seconds before making a deep guttural sound. She had been referred to the sleep clinic to exclude sleep apnoea, but the tests were inconclusive. She slept in a semi-reclined position, which eased her breathing. She complained of severe occipital
pain, bilateral ear pain that was worse on the left, and tinnitus. Her arm symptoms were easier to cope with since the C6 root block.

Clinical examination
She sat with her head tilted in slight side flexion to the left and rotation to the right, her jaw was protracted and she had loss of cervical lordosis with increased thoracic kyphosis. She was tense and looked unwell, and there was excessive sweating over the left half of her forehead, face and chin, and her left eye was watering. She was dizzy on any movement of the head or change of body position, especially from lying to sitting. The left hand and fingers were swollen, shiny and sweating, and she protected these with the other hand, holding the left arm in adduction and medial rotation. Her voice was husky and hoarse and she constantly cleared the back of her throat. Her breathing was laboured and rasping although she did not appear to be aware of this. She scored her occipital and frontal pain at 9-10 on a verbal score of 0-10, and her left arm pain at 8-9.

She demonstrated a good range of cervical spine movement (RoM) with pain at the end of extension and right lateral flexion. Both sternocleidomastoid muscles (SCMs) appeared to have increased tone. This was particularly noticeable on the left. She became extremely distressed on palpation of the left SCM, which exhibited taut bands in both sternal and clavicular heads. Palpation of these trigger points immediately reproduced laboured breathing and swallowing difficulties with increased occipital pain.

She had a marked loss of intervertebral movement at T4 to T8, with an increased kyphosis and a protracted chin. She had pain on palpation at T4 to T6 with restricted rotation to the left. Any palpation over the thoracic spine between T2 and T8 invoked breathing difficulties and adverse sweating.

She showed the following autonomic changes in the left arm and hand: allodynia to light touch, heat and cold; circulatory disturbances; increased skin pigmentation and hair growth; and swelling of the arm and fingers.

Her CNS reflexes were normal and she did not have Horner’s Syndrome. She had been diagnosed with depression and anxiety by her GP since the onset of her symptoms, and reported panic attacks and feeling of inability to cope.

Treatment
The patient had previously undergone chiropractic treatment with little improvement, and it became apparent that she would not tolerate any manipulation at the cervical spine as the spasm in SCM prevented such treatment and may well have exacerbated her autonomic symptoms. The initial course of action was to reduce her anxiety and stress, induce a deeper sleep pattern, and gain some trust in this treatment approach. After discussion with the patient and gaining her consent, a TCM protocol aimed at relaxation and anxiety reduction was used. The patient was treated three times a week for two weeks with the protocol shown in Table 1.

After two weeks, her breathing and general anxiety levels were improved; her verbal pain score reduced to 6 out of 10. The next step was to

<table>
<thead>
<tr>
<th>Table 1</th>
<th>Traditional Chinese acupuncture approach in first nine treatment sessions</th>
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<tr>
<td>Treatment sessions</td>
<td>Points used</td>
</tr>
<tr>
<td>1 to 3</td>
<td>LR3, LI4, Yintang</td>
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<tr>
<td>4 to 6</td>
<td>LR2, LR3, LI4, LU10, LU7, BL13</td>
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<tr>
<td>7 to 11</td>
<td>LR2, LR3, LI4, LU10, LU7, BL13, LI11, LI20</td>
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deactivate the myofascial pain component and restore some postural alignment. The thick black coating to the tongue had changed to a thick, white coating.

Myofascial component

With the patient supine, a positive bilateral SCM compression test could be demonstrated, ie compression restored pain-free swallowing. A taut band was palpated in both SCM clavicular and sternal heads, and pressure reproduced the patient’s pain pattern.

Trigger points in the left sternal and clavicular heads of SCM were deactivated with a very fine gauge needle, 1.5cm in length, using a pinch grip to the SCM belly and searching for taut bands. A jump reaction was initiated by needling. The needle was left in situ for 30 seconds or until needle grasp was released. It was thus decided to deactivate the muscles individually for the safety and comfort of the patient. The scalene muscles tend to develop trigger points, especially if bilateral SCM trigger points are present for some time. The plan of deactivation is shown in Table 2.

Further pain management techniques were required to address stress management, relaxation and positive cognitive feedback. She requires daily myofascial stretch and exercise regimes, in order to keep pain and anxiety at reasonable levels. She will require a full ergonomic workstation assessment before returning to work, in order to prevent an exacerbation of the postural dysfunction.

On reassessment at 18 weeks, the patient was working full time, sleeping and maintaining an improved lifestyle with control of her pain pattern without opioids. However, on retesting for trigger points and taut bands, she was certainly uncomfortable on deep palpation. Although the trigger points were not active or referring pain, it was obvious that myofascial dysfunction was still likely. After further discussion, we decided that she would benefit from botulinum toxin injection to SCM with the aim of enhancing the deactivation and hopefully offering her longer-term relief.

Literature search

A computerised search of Pubmed, Medline and Amed was performed, and a hand search of Acupuncture in Medicine, The Journal of Chinese Medicine and Complementary Therapies in Medicine. Email and telephone were used to discuss the case with Dr Pearce, Consultant Neurologist at Hull Royal Infirmary and author of an article that questions the existence of Barré-Liéou Syndrome.1

This patient demonstrated symptoms in keeping with the Barré-Liéou Syndrome,2 a diagnosis that is disputed: some authors claim it is a figment and simply a repository for undiagnosed symptoms3,4 while others argue that it is genuine.4 Barré Liéou Syndrome was first described by Barré in 1926 when he identified a number of symptoms affecting the third and fourth cervical vertebrae that often followed minor neck trauma. The syndrome is believed to be caused by arthrosis of the cervical column or a protrusion of the skeletal border of the vertebral body irritating the cervical plexus, or even both, thus affecting the cervical arteries.5 The condition has been documented in children, and in occupations where people are forced to hold the neck in prolonged upward gaze. The result is increased cervical lordosis placing undue pressure on the vertebral arteries and reducing blood flow to the circle of Willis.6 The syndrome involves dysfunction of the posterior cervical plexus resulting in severe occipital pain, visual disturbance,1 and vertigo; it is also thought to involve the cervical sympathetic group.bmj.com on September 19, 2017 - Published by http://aim.bmj.com/Downloaded from
nerves, the blood supply to the cerebellum, and the cranial nuclei V, VIII and XI. Not all authors regard the syndrome as a single neurological entity: authors such as Randov et al, Del Torto, and Wight et al, describe a variety of symptoms including pain, visual disturbance, breathing and swallowing difficulties and cervical muscle spasm especially within the SCM muscle either unilaterally, or in most cases, bilaterally. Other symptoms involve anxiety, depression, insomnia, memory and cognitive disorders, with intermittent nocturnal respiratory arrhythmia.

Travell and Simons describe trigger points in sternal and clavicular heads of SCM giving rise to different patterns of autonomic phenomena or proprioceptive disturbances. The sternal division refers pain to the vertex, occiput and eye with constrictive symptoms in the throat, whilst the clavicular division involves the eye, sinuses and throat. Patients rarely present with neck pain or limited movement.

Discussion
This was a particularly complicated and challenging case presenting with a myriad of symptoms augmented by hyperactivity in the autonomic nervous system, reinforced by the patient’s distress and anxiety. It was particularly important to gain her co-operation and ultimate trust in order to make any changes to her symptoms. Acupuncture was the treatment of first choice as, although an invasive technique, it offers maximum TrP deactivation with minimal manipulation, compared with manual myofascial techniques. As was demonstrated, any amount of palpation and pressure increased the autonomic distress.

It became necessary to enhance some of the acupuncture treatment by offering some sort of orthopaedic cervical support. This was constructed in order to provide continuous posterior pressure at C5-8 to produce a cervical lordosis. This allows the patient to increase the lordosis when lifting her head, thus allowing release of the tight scaleni.

The patient continues postural re-education, muscle imbalance strengthening and relaxation techniques at home. She sleeps through the night, however, and can now manage minor household chores.

The clinical outcome of this case presentation was particularly satisfying, along with the treatment protocol used, as it enabled the physiotherapist to combine TCM and WMA for a holistic approach to management. This approach often benefits patients in chronic neuropathic pain who have had previous interventions with little effect. Acupuncture appears to have had an effect on this case especially in reducing some of the autonomic hyperactivity.

Although the diagnosis of Barré Liéou Syndrome appears contentious and is dismissed as a catch-all term for a variety of symptoms, this may be attributed to problems of vertebral instability affecting the nerve cell aggregations located in the neck. Ishikawa et al suggest that there may actually be a leak of cerebrospinal fluid into the epidural space at the thoraco-lumbar region, which will not show on MRI or CT scan but only on radionuclide cisternography.

Summary
Any therapist working in the field of chronic pain is often faced with a barrage of symptoms and may be tempted to approach the patient with aggressive manual techniques that only exacerbate the condition. Acupuncture may offer considerable sympathetic and central nervous system inhibition with a minimum bodily manipulation. The case history given here represents a relatively short treatment period in the overall management of this patient; ideally, longer-term follow up is needed to ascertain the overall effectiveness of the procedures used. It is clear that a larger, longer-term study would be useful in comparing the clinical effectiveness of acupuncture against a manual myofascial management regime.

Acknowledgements
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Case reports


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