Clinical Case Report

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Neck-to-shoulder pain as an unusual presentation of pulmonary embolism in a patient with cervical spinal cord injury

A case report

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Abstract

Rationale: Information on referred pain can be helpful for diagnosing diseases of the visceral organs. Here, the authors report a patient with cervical spinal cord injury (SCI) who had referred pain at the right side from the neck to shoulder, as a presentation of pulmonary embolism (PE).

Patient concerns: A 55-year-old man with complete tetraplegia, due to cervical SCI after C5 and C6 vertebral body fracture, complained of right neck-to-shoulder pain (numerical scale rating: 6). Despite pain medication (meloxicam 15 mg, gabapentin 400 mg, and propacetamol HCl 1 g), the pain was not reduced. Along with right neck-to-shoulder pain, he presented mild fever (37.8°C) and mildly elevated respiratory rate (20 breaths/min). D-dimer level was also mildly elevated to 6.09 mg/mL (normal value: < 0.5 mg/mL).

Diagnoses: Computed tomography pulmonary angiography revealed PE in the right lower lobe pulmonary artery.

Interventions: For managing PE, rivaroxaban was administered for three days.

Outcomes: After the administration of rivaroxaban, the patient's pain completely disappeared.

Lessons: This study shows that pain at the neck-to-shoulder area can occur following unexpected causes such as PE. Not limited to PE, the evaluation of diseases in the thoracic or abdominal organs is recommended if patients with cervical SCI present refractory pain in the dermatomes innervated by high cervical nerve roots.

Abbreviations: AIS = Association Impairment Scale, CT = computed tomography, CUS = compressive ultrasonography, DVT = deep vein thrombosis, PE = pulmonary embolism, SCI = spinal cord injury.

Keywords: pulmonary embolism, referred pain, spinal cord injury

1. Introduction

Visceral pain is characterized by being poorly localized and is usually described as deep, squeezing, or colicky. It is mediated by discrete nociceptors in the cardiovascular, respiratory, gastrointestinal, and genitourinary system.[1] On the other hand, somatic pain is characterized as well localized, intermittent, or constant, and is described as aching, throbbing, or cramping. It arises in somatic structures, such as skin, tissue, and muscle.[2] Frequently, visceral pain is perceived as pain arising from somatic sites.[3–5] Clinicians term this type of pain as "referred pain."[3–5] The most acceptable theory for this phenomenon was proposed by Ruch and Patton.[6] They suggested that visceral and somatic afferents converge onto common dorsal horn neurons; subsequent activation of nociceptors in the viscera results in the perception of pain arising from a somatic source.[6] Although sometimes referred pain causes confusion for clinicians to locate the exact source of pain, the understanding of referred pain can be helpful in diagnosing visceral pathologies. For example, the knowledge that shoulder pain can be a sign of ischemic heart disease can prevent the misdiagnosis of heart disease as having a shoulder problem.[7] Therefore, understanding and awareness of referred pain are necessary for the accurate diagnosis of the pain source.

In the current study, a patient with cervical spinal cord injury (SCI) who had referred pain at the right side from neck to shoulder as a presentation of pulmonary embolism (PE) is reported.

2. Case report

A 55-year-old man, who had no previous history of pulmonary or cardiac disease, had cervical SCI due to C5 and C6 vertebral body fracture and dislocation after car accident; he underwent posterior cervical laminectomy and posterolateral fusion on C3–7 at the neurosurgery department in a university hospital. He developed complete tetraplegia and was transferred to the rehabilitation department in the same university hospital 13 days after the onset of SCI. Pharmacologic prophylaxis for deep vein

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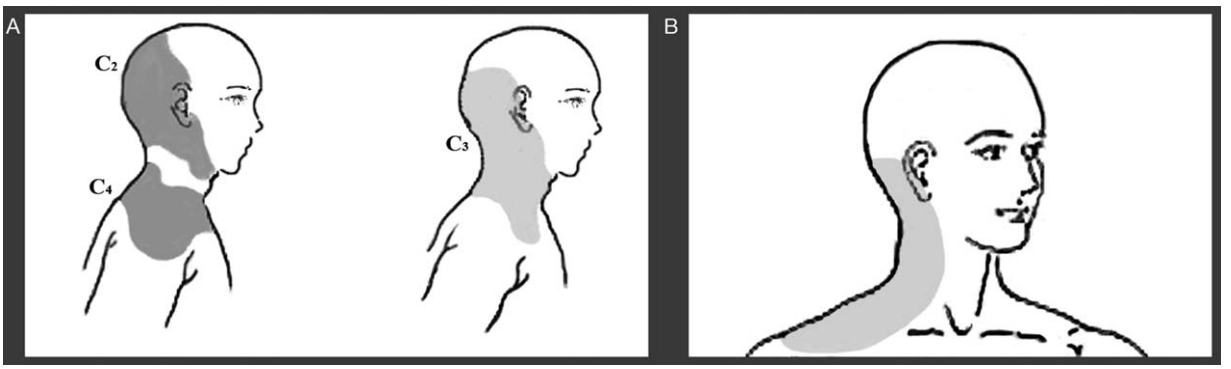

Figure 1. A, The dermatomal distribution of the C2–4 nerve root. B, The location of the patient's perceived pain (right neck-to-shoulder area).

thrombosis (DVT) and PE was not provided. At the time of transfer to the rehabilitation department, his neurological level of injury was C5, and the American Spinal Injury Association Impairment Scale (AIS) was grade A; motor level C5/C5, sensory level C5/C5, motor score 8/8, sensory score 20 for light touch, and 20 for pin prick. He had no perianal sensation. Two weeks after the onset of SCI, the patient complained of dull pain at the right side from the neck to shoulder (numerical scale rating: 6) (Fig. 1), together with mild fever (37.8°C) and mildly elevated respiratory rate (20 breaths/min). For the management of the patient's pain, pain medication (meloxicam 15 mg, gabapentin 400 mg, and propacetamol HCl 1 g) was administered. Despite pain medication, the patient's pain did not decrease (numerical scale rating: 6). The white blood cell count was elevated to 14,860 cells/μL (normal value: 4000–10,000/μL), and the C-reactive protein level was at 7.0 mg/dL (normal value: < 1.0 mg/dL). The other laboratory and radiology tests performed to locate the source of mild fever did not show any abnormality: urine leukocyte (−), urine WBC 0~1 cells/high-power field, blood culture (−), urine culture (−), and chest X-ray (−). Because D-dimer was mildly elevated to 6.09 μg/mL (normal value: < 0.5 μg/mL), compressive ultrasonography (CUS) was performed for locating DVT and computed tomography (CT) pulmonary angiography for PE. CUS did not reveal any evidence of DVT; however, CT pulmonary angiography presented with PE in the right lower lobe pulmonary artery (Fig. 2). The capnography conducted for 1 hour showed that his average SpO_2 was 94% and his average pCO_2 was 42 mm Hg in room air. For the treatment of PE, 30 mg rivaroxaban was orally administered for 3 weeks, followed by 20 mg rivaroxaban for 3 months. Three days after the administration of rivaroxaban, the patient's pain completely disappeared. The patient provided informed consent for the participation in the study. The study was approved by the Institutional Review Board of Yeungnam University Hospital.

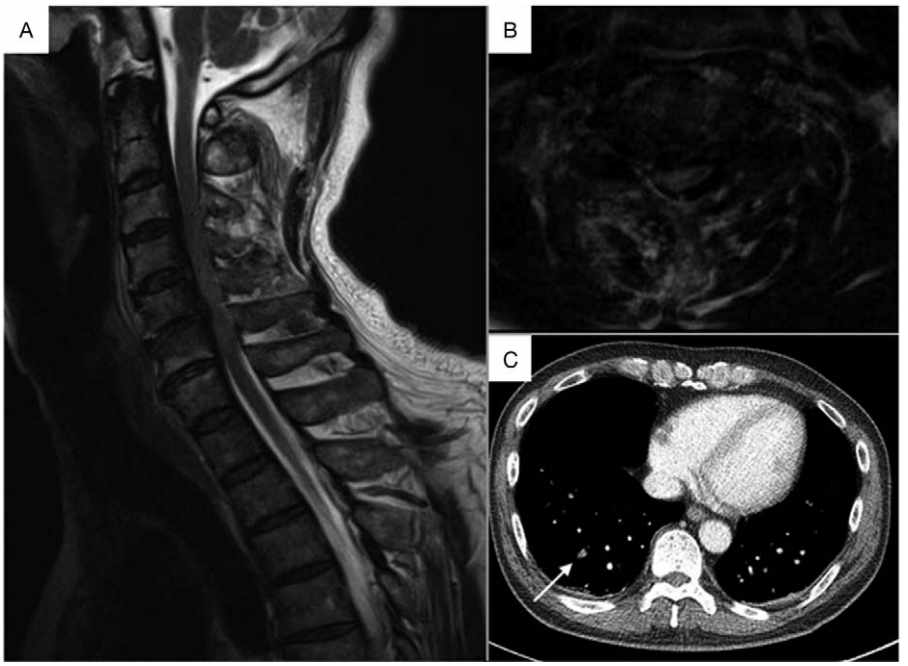

Figure 2. Cervical spinal magnetic resonance imaging performed after the injury. A, Sagittal T2-weighted image shows cervical cord compression with central canal stenosis at the C5–6 level. B, Axial T2-weighted image at the C5–6 disc level shows compressed spinal cord with high signal intensity. C, Computed tomography pulmonary angiography shows intraluminal filling defect in the right lower lobe pulmonary artery (white arrow).

3. Discussion

The current study reports a patient who complained of right neck-to-shoulder pain, which seems to be referred pain from PE. First, we thought that the patient's pain was neuropathic pain or musculoskeletal pain, such as myofascial pain syndrome, rotator cuff syndrome, or cervical facet joint syndrome. The pain was not reduced after the administration of pain medication; however, it was completely relieved after the treatment of the PE. Furthermore, the dermatome corresponding to the location of the patient's pain was at C2–4 (Fig. 1).[8] Because the patient's last intact level was C5, his pain was not neuropathic pain from SCI.[9] Considering the responses to the treatment and the clinical symptoms, the patient's pain on the right side from the neck to shoulder was referred pain from PE in the right lower lobe pulmonary artery.

Referred pain is the pain perceived at a location adjacent to or a distance from the location of origin.[3–6] Although the mechanism of referred pain has not been elucidated, the most acceptable theory is the convergence-projection theory. It states that multiple nerves converge into a single shared neural pathway; hence, the central nervous system is unable to differentiate the origin of pain.[6] The bronchial branch of the vagus nerve innervates the lung, and the nociceptive input from the lung transfers to the spinal trigeminal nucleus via the vagus nerve.[10] The spinal trigeminal nucleus descends as far caudally as C3 or C4 level; together with the input via the vagus nerve, the afferent input from the upper cervical segments also synapses with the spinal trigeminal nucleus.[11,12] Considering that neck-to-shoulder area corresponds to C2–4 dermatome,[8] irritative information from PE on the right lung seemed to have converged with the sensory information from the right neck-to-shoulder area. Through this sharing in the spinal trigeminal nucleus, the patient seemed to have perceived neck-to-shoulder pain, even though there was no structural problem in that area.

PE is the potentially life-threatening condition causing morbidity and mortality in patients with acute SCI.[13] Venous stasis and alterations of various regulatory proteins following paralysis increase the incidence of thromboembolic disorders, DVT, and PE.[14] In the acute phase of SCI, the incidence of PE has been reported as 4.5% with a mortality rate of 3.5%.[15] If patients present high suspicious clinical presentation of PE, a thorough examination for diagnosing PE is necessary. CT pulmonary angiography is the first-line imaging test for acute PE.[16] If patients show symptoms that indicate PE, CT pulmonary angiography should be conducted for the diagnosis of PE. Usually, clinical symptoms of PE manifest as dyspnea, tachypnea, syncope, or chest pain; however, these symptoms are indistinctive from the symptoms that typically present after cervical SCI.[17] The symptoms from PE can be easily misjudged as the symptoms from weakness of respiratory muscles, orthostatic hypotension, or neuropathic pain. Therefore, diagnosis of PE in patients with cervical SCI is challenging. This report suggests that pain in the neck-to-shoulder area in patients with SCI can be a potential clue for the diagnosis of PE. Additionally, in the case of complete cervical spinal cord injury, patients cannot receive sensory inputs from the dermatomes below the level of injury to the spinal cord. Considering that the vagus nerve carries nociceptive afferent input from the viscera of thorax and abdomen, the referred pain in the neck-to-shoulder area is not limited to PE.[18] The refractory pain in the neck-to-shoulder area (the dermatome of upper cervical nerve distribution) may be an important sign of internal organ diseases.

Our patient did not receive prophylactic treatment for DVT and PE. Previous studies reported that Asian patients have a significantly lower incidence of DVT as compared with western patients.[19,20] Therefore, pharmacologic thromboprophylaxis with either warfarin or heparin is not routinely provided to Asian patients with SCI. However, recently, several studies have reported that the incidence of DVT after SCI in Asian patients is comparable with that in western populations.[21–23] On the basis of the results of these recent studies, we think Asian clinicians should consider the routine use of pharmacologic thromboprophylaxis during the acute stage after SCI.

Previously, some studies reported referred pain along with the dermatome from the upper cervical levels induced by lung diseases.[24,25] Petchkrua et al[24] reported a patient who had a referred pain in the right shoulder area due to pneumonia in the right lower lobe. Furthermore, van der Bruggen et al[25] reported a patient with referred pain in the bilateral shoulder areas following a large central tumor in the right lung. The authors of aforementioned studies suggest that the patient's referred pain was induced by diaphragmatic irritation transmitted via the right C4 sensory nerves in the phrenic nerve, which shares the same dermatome as the shoulder area.

In this study, the patient had referred pain in the neck-to-shoulder area, which seemed to have been induced by PE; the pain was dramatically reduced after the treatment of PE. This case study shows that pain at the neck-to-shoulder area can occur following unexpected causes such as PE. This is the first study to show that PE can cause pain in the neck to shoulder area and pain in this area can be a sign of PE. In addition, not limited to PE, the evaluation for the existence of disease in the thoracic or abdominal organs would be necessary if patients with cervical SCI present refractory pain in the dermatomes innervated by high cervical nerve roots.

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