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The diagnosis process of Collet-Sicard syndrome caused by skull base fracture: A case report

El proceso de diagnóstico del síndrome de Collet-Sicard por fractura de la base del cráneo: reporte de un caso

Sr. Editor:

Collet-Sicard syndrome caused by trauma is rare.[1] Vocal cord paralysis (VCP), difficulty swallowing, loss of taste in the posterior third of the tongue, tongue muscle and sternocleidomastoid muscle atrophy and paralysis are the typical clinical manifestations.[2] The patient complains to the doctor only about hoarseness and difficulty swallowing. They will ignore the change in taste and muscle. So some doctors misdiagnose it as simple VCP.

The female patient is 35 years old. The patient was unconscious when she fell from a height of 9 meters on June 24, 2020. The Glasgow Coma Scale score was 8/15 (E2, V1, M5). Computed tomography (CT) showed that occipital fractures on both sides and fractures of left transverse process of atlas. She underwent tracheal intubation twice because she removed the tube by herself. Her consciousness was clear and removed the tube on June 26. She was dysphonic, difficulty swallowing and given a nasogastric tube after swallowing assessment. Neurotrophic therapy was applied when the damage of laryngeal nerve was considered. She accepted swallowing rehabilitation and acupuncture treatment. The electronic fiber laryngoscope (EFL) showed complete bilateral vocal cord paralysis on July 23 (Fig. 1A). The hoarseness of the patient began to improve significantly on July 29. We found the tongue of patient was tilted to the left and the posterior third of the tongue had decreased taste on July 30. The EFL indicated that the right vocal cord was normal, and the left vocal cord was still completely paralyzed on August 21 (Fig. 1B). The patient passed the swallowing assessment successfully and the nasogastric tube was removed on August 28. One month later, the patient still had a hoarse voice, decreased taste, and a crooked tongue. The difficulty swallowing was disappeared. The EFL still showed complete paralysis of the left vocal cord (Fig. 1C).

The most common cause of Collet-Sicard syndrome is tumor metastasis of skull base, followed by vascular disease and trauma.[3] Collet-Sicard syndrome caused by skull base fracture had only been reported 8 times in English literature. The cranial nerves IX—XII near the jugular foramen are damaged when the skull base fractured.[3] Some scholars speculated that the pathogenesis was nerve edema or bone fragments directly compressing the nerve.[4,5] Since hoarseness and difficulty swallowing are the main symptoms, it is easy for doctors to misdiagnose it as simple VCP. Symptoms of VCP are hoarseness, difficulty swallowing and difficulty breathing.[6] Both tracheal intubation and trauma can cause VCP. Arytenoid dislocation or recurrent laryngeal nerve damage is the main cause of VCP after tracheal intubation.[7] EFL can be used to observe arytenoid cartilage to determine whether it is dislocated. In this case, the patient received tracheal intubations twice in a coma. So the tracheal intubation was first considered as the cause of VCP. The anesthesiologist reported that the tracheal intubations went smoothly. The EFL did not indicate arytenoid dislocation. Therefore, the anesthesiologist disagreed with the speculation that tracheal intubation caused VCP. Trauma can also lead to VCP. CT showed the left transverse process of atlas was fractured (Fig. 1D). But the transverse and longitudinal diameters of the atlas are large, the atlas fractures rarely cause VCP. VCP is possible exist when the atlas has a Jefferson fracture.[8,9] In this case, the degree of atlas fractures was mild. So it was not the responsible of this disease. We were confused until we discovered the tongue of patient was tilted to the left (Fig. 1G). And then, we tested the patient's sense of taste. The patient told us that she lost the taste in the posterior third of the tongue. It was a typical clinical manifestation of cranial nerve IX injury. Both cranial nerve IX and X are damaged at the same time, there will be hoarseness and difficulty swallowing. Cranial nerve XII injury will cause atrophy of the ipsilateral tongue muscles, and the tongue will be deflected to the affected side. There was no clinical manifestation of cranial nerve XI damage in this patient. We suspected that the cranial nerve XI damage might be mild, or the course of the disease might be short and had not been manifest. CT indicated that bilateral skull base fractures (Fig. 1E and F). Collet-Sicard syndrome was diagnosed successfully.

In conclusion, when patients with head trauma have hoarseness and difficulty swallowing, CT examination of the skull base must be performed. The doctor must check the

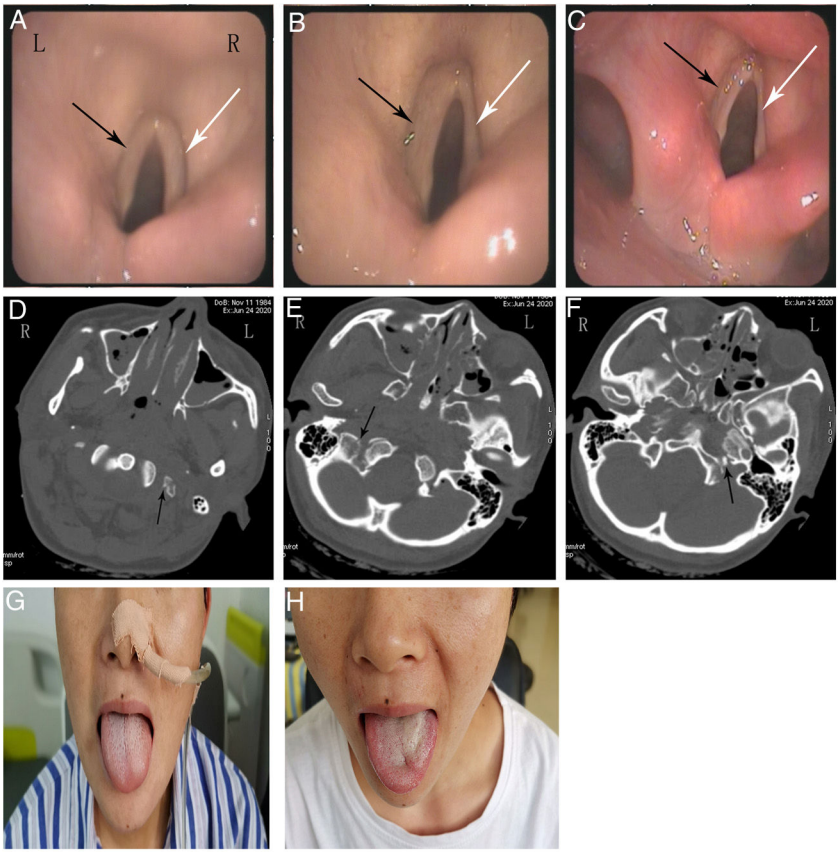

Figure 1 The electronic fiber laryngoscope shows that the bilateral vocal cords were fixed and the glottis could not move on July 23 (A). The left vocal cord (black arrow) was fixed and the right vocal cord (white arrow) was normal on August 21. There was a gap when the bilateral vocal cords closed (B). The left vocal cord (black arrow) was still fixed on September 21 (C). The computed tomography showed that the fracture of the left transverse process of the atlas and right skull base was mild (D, E). Bone fragments were found in the left occiput (F). The tongue tilted to the left on July 30 (G). The muscles of the left tongue were atrophy, and the tongue deviated to the left side on September 21 (H).

taste, tongue muscle and sternocleidomastoid muscle of patient. If the condition is stable, conservative treatment is recommended. Nutritional nerve therapy and swallowing rehabilitation are necessary.[2] This case reports our incorrect diagnosis process, in order to young doctors to have a deeper and comprehensive understanding of Collet-Sicard syndrome.

Disclosure

Informed consent was obtained for publication of the pafient's details described in this report.

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Improvement of restless legs syndrome with a plantar pressure device☆

Mejoría del síndrome de piernas inquietas con un dispositivo de presión plantar

Dear Editor:

Restless legs syndrome (RLS) is a sensorimotor disorder characterised by an urge to move the legs, frequently associated with unpleasant sensations in the legs, and typically presenting in the evening or night and at rest.[1] It often causes sleep alterations, and has a similar impact on quality of life to that associated with other chronic diseases.[2] The pathophysiological mechanisms of RLS are poorly understood; however, response to dopaminergic agents and iron supplementation has given rise to a number of theories on its aetiopathogenesis.[3] In addition to dopaminergic agonists and iron supplements, pharmacological treatment options also include $\alpha 2\delta$ ligands and some opioids.[4] Non-pharmacological options include physical therapy; magnetic, electrical, or vibratory stimulation; and pneumatic compression devices.[5] These may be used as complementary treatments when pharmacological treatment fails to achieve the desired response.

We describe the cases of 2 patients with RLS who presented partial response to pharmacological treatment. *Patient 1:* 70-year-old man with a 7-year history of RLS. In the previous year, he had been diagnosed with chronic kidney disease, generalised anxiety disorder, obstructive sleep apnoea syndrome, arterial hypertension, and obesity. During assessment at the sleep disorders clinic, chronic kidney disease was detected (glomerular filtration rate 30.77 mL/min), but no manifestations of neuropathy or anaemia were observed (Hb 14.7 g/dL), and ferritin levels were within the normal range (238 μg/mL). The patient was being treated with captopril, continuous positive airway pressure (CPAP), and escitalopram. To treat RLS, he was receiving pramipexole 0.25 mg (administered at bedtime), but significant discomfort and sleep problems persisted. *Patient 2:* 55-year-old woman with 4-year history of RLS and history of osteoarthritis and fibromyalgia; the latter 2 conditions were diagnosed at the age of 52 years and were under treatment with duloxetine. Pramipexole dosed at 0.125 mg/night achieved little improvement, and was subsequently discontinued due to visual hallucinations. The patient also showed low ferritin levels (65 μg/mL). She was prescribed pregabalin 450 mg/day and ferrous fumarate 700 mg/day. Serum ferritin increased to 112 μg/mL, and the patient improved slightly, but the symptoms delaying sleep onset persisted. Both patients were indicated to use a compressive foot wrap (Restiffic®) as a complement to pharmacological treatment; they were instructed to use the device at night, when discomfort started. They were also instructed not to make any changes in the treatments they were taking for RLS or other comorbidities. We evaluated the symptoms of RLS at baseline and each week after treatment onset for 4 weeks, using the RLS rating scale (RLSRS) and patient-reported data.

Symptom severity decreased remarkably in both patients, from very severe to moderate in the first patient (from 32 to 15 points) and from very severe to mild in the second (from 32 to 8 points). During follow-up, both patients reported not having used the device on some occasions, which led to symptom exacerbation (Fig. 1). They also reported transient irritation and oedema in the area covered by the foot wrap as the only adverse events.

Although several drugs are currently available for RLS, they may cause significant adverse effects (e.g., potentiation of the effects of dopaminergic agonists, especially levodopa) and may achieve poor or suboptimal results.

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