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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

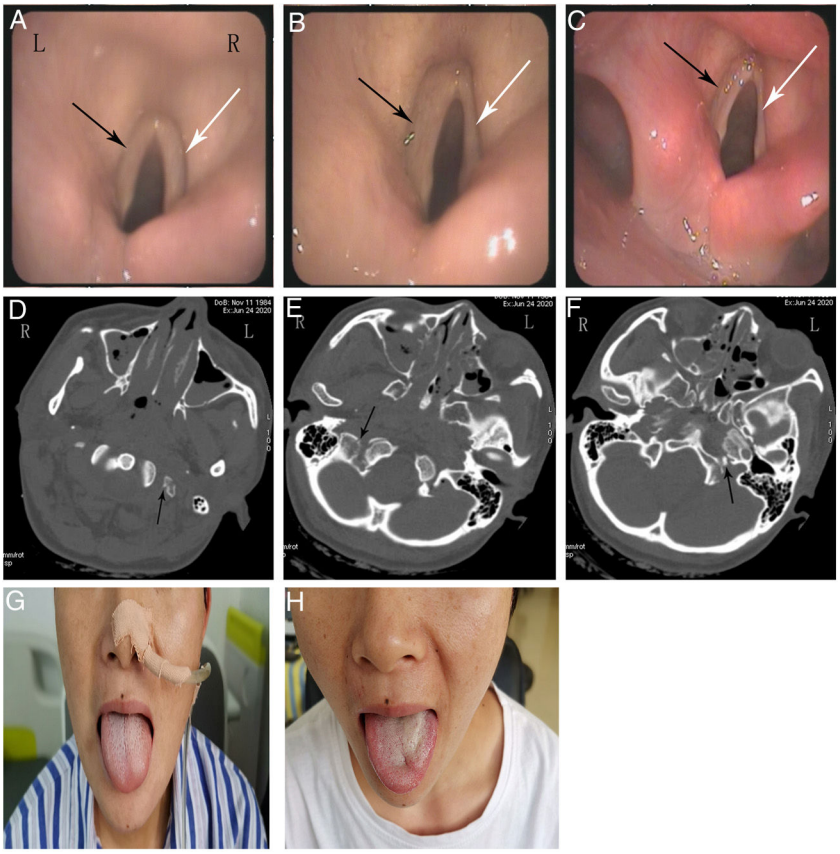


**Figure 1** The electronic fiber laryngoscope shows that the bilateral vocal cords were fixed and the glottis could not move on July 23 (A). The left vocal cord (black arrow) was fixed and the right vocal cord (white arrow) was normal on August 21. There was a gap when the bilateral vocal cords closed (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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# Mejoría del síndrome de piernas inquietas con un dispositivo de presión plantar

## Improvement of restless legs syndrome with a plantar pressure device

*Sr. Editor:*

El síndrome de piernas inquietas (SPI) es un trastorno sensoriomotor que consiste en la presencia de una necesidad imperiosa por mover las piernas habitualmente asociada con sensaciones desagradables en las piernas, las cuales suelen aparecer por la tarde/noche y en reposo[1]. Frecuentemente produce alteraciones del sueño y afecta la calidad de vida en un grado similar al de otras enfermedades crónicas[2]. El mecanismo fisiopatológico no es del todo bien comprendido, sin embargo, la respuesta a los agentes dopaminérgicos y a la suplementación con hierro han sido los principales impulsores de las teorías sobre los mecanismos etiopatogénicos subyacentes[3]. Además de los agonistas dopaminérgicos y la administración de hierro, dentro de las opciones de tratamiento farmacológico se encuentran los $\alpha 2\delta$ ligandos y algunos opioides[4]. Existen modalidades de tratamiento no farmacológico como la terapia física, la estimulación magnética, eléctrica y vibratoria, así como los dispositivos de compresión[5]. Estas opciones pueden utilizarse de forma complementaria cuando no se ha obtenido la respuesta deseada al tratamiento con fármacos.

Presentamos 2 casos de pacientes con SPI que presentaron respuesta parcial al tratamiento farmacológico. *Caso 1:* Varón de 70 años con SPI de 7 años de evolución. Asimismo, en el último año se le había diagnosticado enfermedad renal crónica, trastorno de ansiedad generalizada, síndrome de apnea obstructiva del sueño, hipertensión arterial y obesidad. Durante su atención en la clínica de sueño se constató la enfermedad renal crónica (tasa de filtración glomerular 30,77 ml/min), sin la presencia de manifestaciones clínicas de neuropatía y anemia (Hb 14,7 g/dl) con valores de ferritina adecuados (238 μg/ml). Se encontraba en tratamiento con captopril, terapia de presión positiva para vías aéreas (CPAP) y escitalopram. Para el SPI recibía pramipexol 0,25 mg/noche, sin embargo, persistía con síntomas significativos y notables problemas en su sueño. *Caso 2:* Mujer de 55 años con SPI de 4 años de evolución e historia de osteoartritis y fibromialgia diagnosticadas a los 52 años, y para las cuales recibía duloxetina. Se le prescribió pramipexol 0,125 mg/noche, pero percibió poca mejoría y presentó alucinaciones visuales por lo que interrumpió el fármaco; asimismo se obtuvieron niveles bajos de ferritina (65 μg/ml). Por lo anterior, se indicó pregabalina 450 mg/día y fumarato ferroso 700 mg/día. La ferritina sérica aumentó a 112 μg/ml y mejoró ligeramente, pero persistieron síntomas que prolongaban el inicio de sueño. A ambos pacientes se les propuso utilizar un dispositivo de presión plantar (Restiffic®) como medida complementaria al tratamiento farmacológico, instruyéndoseles para usar el dispositivo por la noche, al iniciar con las molestias. Así también, se les indicó no realizar modificaciones en los tratamientos que recibían para el SPI y otras condiciones médicas. Se evaluaron los síntomas de SPI al inicio y semanalmente por 4 semanas con la escala de gravedad de SPI (EGSPI), así como un registro de eventos.

La gravedad de los síntomas se redujo notablemente en ambos pacientes, en el primero la puntuación disminuyó de 32 a 15 y en la segunda de 32 a 8, es decir, pasaron de un padecimiento muy grave a una condición moderada y leve, respectivamente. Durante el tiempo de seguimiento, ambos pacientes reportaron haber realizado omisiones en la colocación del dispositivo, presentando exacerbación de los síntomas con la interrupción temporal (fig. 1); como eventos adversos describieron irritación local y edema transitorios.

Si bien existen fármacos eficaces para el tratamiento del SPI, algunos de ellos pueden producir efectos adversos significativos (por ejemplo, potenciación con agonistas dopaminérgicos, especialmente con levodopa), o bien la mejoría puede ser pobre o subóptima. Aunque los niveles séricos de ferritina < 75 μg/ml pueden en ocasiones asociarse con una menor respuesta terapéutica, solamente en un caso se documentó una concentración sérica baja y el tratamiento con hierro oral no modificó sustancialmente la respuesta. En estos casos, cobran mayor relevancia las modalidades de tratamiento no farmacológico. Los 2 pacientes que describimos así lo sugieren, pues habían tenido una