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Bilateral Tapia syndrome in teenager with post traumatic Hangman's fracture and carotid artery dissection

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ABSTRACT

Background: Tapia syndrome (TS) is a rare condition characterized by unilateral hypoglossal and recurrent laryngeal nerve palsy, leading to tongue deviation, swallowing difficulty and dysphonia.

Case report: We describe a case of a 17-year-old boy who reported a bilateral TS following head and neck trauma with Hangman's fracture and right common carotid artery dissection. The confirmation occurred only after complete cognitive and motor recovery, verifying the inability to protrude the tongue and swallow, associated with complete paralysis of the vocal cords, diagnosed with fiber optic laryngoscopy.

An initial recovery of tongue motility and phonation occurred after just over a month of rehabilitation.

Conclusion: In addition to the lack of awareness due to the rarity of the syndrome, the diagnosis of TS may be delayed in patients who are unconscious or who have slow cognitive recovery following head trauma. The case we present may help to increase awareness and avoid unnecessary diagnostic investigations.

Introduction

Tapia syndrome (TS) is a rare condition characterized by unilateral hypoglossal and recurrent laryngeal nerve palsy, leading to paralysis of the tongue and vocal cord, manifesting as tongue deviation, swallowing difficulty and dysphonia [1].

TS was first described by the Spanish otolaryngologist Antonio Garcia Tapia in 1904, as a peripheral lesion of the hypoglossal nerve and the recurrent laryngeal branch of the vagus nerve, in a bullfighter with neck trauma [2].

Over the course of over a century, very few cases of TS have been described, mostly of traumatic nature, often caused by direct compression or stretching of both nerves as a complication of orotracheal intubation [3,4].

Even more rare and dramatic is the simultaneous and bilateral involvement of both nerves such as to compromise the possibility of

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feeding, due to complete paralysis of the tongue, and breathing due to complete paralysis of the vocal cords [5,6].

This condition may occur after a damage of the nuclei of the X and XII cranial nerves [7] or a simultaneous involvement of both nerves along their peripheral route subsequently to a trauma of the upper cervical spine [8,9].

Here we present a case of bilateral hypoglossal and recurrent laryngeal nerve palsy in a 17-year-old boy following a road traffic accident that resulted in diffuse axonal injury, bilateral fracture of the pars interarticularis of cervical vertebrae 2 (C2) that caused spondylolisthesis of C2 (Hangman's fracture) and right common carotid artery dissection.

Case presentation

A 17-year-old boy with no significant past medical issues was brought to our emergency room after a *collision* involving a *motorcycle* and a transport *truck, a cervical collar correctly applied and oxygenation ensured with an oxygen mask*.

Upon admission to the emergency room the patient was unconscious (Glasgow Coma Scale <8), the *hemodynamic* condition was *stable*, *his pupils were* bilaterally *isochoric*, *isocyclic*, and reactive to light. Orotracheal intubation was performed with a video laryngoscope and the *cuff pressure* was measured using a cuff manometer and was set to approximately *20–25 cmH_2O*. Adequate and timely interventions allowed the SpO_2 never to fall below 90 %, while the mean arterial pressure was maintained over 65 mmHg.

A first total-body contrast enhanced computed tomography (CECT) was performed and a thin layer of left hemispheric subdural hematoma was found, along with intraventricular hemorrhage, bilateral fracture trasversing the pars interarticularis of C2 with an associated subluxayion of C2 on cervical vertebrae 3 (C3) (Hangman type fracture, Fig. 1), right common carotid artery dissection, and *right liver lobe* laceration without active bleeding.

Subsequently the sedated patient was transferred to the operating room and a cerebral intraparenchymal catheter was positioned to assess the intracranial pressure (ICP), and a value of 12 cmH_20 was shown.

A *carotid artery* and *vertebral angiography* was performed, which documented a right common carotid artery injury with intima invagination (Fig. 2), and an endovascular treatment with a metallic stent in the right common carotid artery was performed. Afterwards, the patient was admitted to the intensive care unit (ICU).

The next day, an increase in *ICP* refractory to standard *medical treatment* was observed. A total body CT scan showed severe general cerebral oedema, while the liver laceration was reported as stable. Therefore, the patient underwent bilateral decompressive craniectomy to facilitate the *control* of *ICP* and improve *cerebral perfusion* pressure.

In the following days, further evaluations regarding the C2-C3 spinal fracture-dislocation were carried out, including a cervical spine magnetic resonance imaging (MRI), which documented the anterolisthesis of C2 over C3, without signs of spinal cord suffering (Fig. 3), and conservative treatments were advised by neurosurgeons.

Somatosensory evoked potentials (SEPs) were performed, and described as normal in terms of amplitude and latency.

On the fifth day upon arrival the patient developed severe pneumonia due to methicillin-susceptible *staphylococcus aureus* (MSSA), successfully treated with cefazolin for 7 days.

On the 11th day upon arrival, after a neurological assessment, showing a left GCS 6 (E1-V1-M4) and absence of motor response on the right side, a brain CT scan was performed: reduced cerebral oedema and no ischaemic areas were found.

On the 13th day, with a persistent GCS < 8, a tracheostomy was performed.

In the following weeks, a gradual and slow cognitive and motor recovery was observed.

On the 42nd day upon arrival, the patient was awake, conscious, with normal limb movements, and in spontaneous breathing through the tracheostomy tube. However, during the neurological evaluation we acknowledged an inability to protrude the tongue, in lateral tongue movements, and in swallowing. Furthermore, a fiber optic laryngoscopy highlighted complete vocal cords paralysis with stagnation of saliva in the piriform recess.

Subsequently the patient was discharged to an acute care rehabilitation and after a month of physical therapy an initial recovery of tongue motility and vocalization occurred.

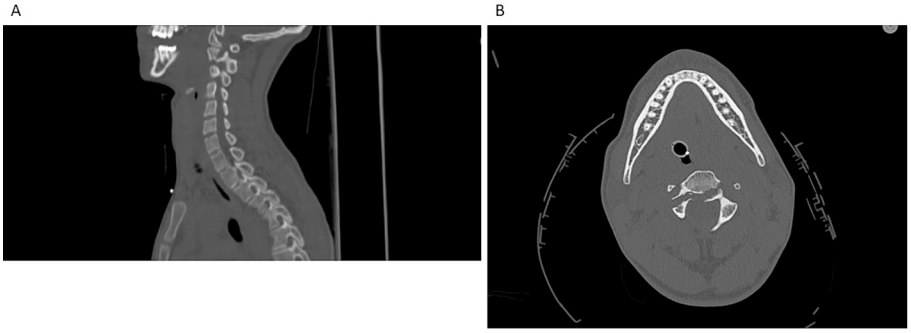

Fig. 1. Sagittal (A) and axial (B) CT scan showing a complex C2 fracture (Hangman's facture).

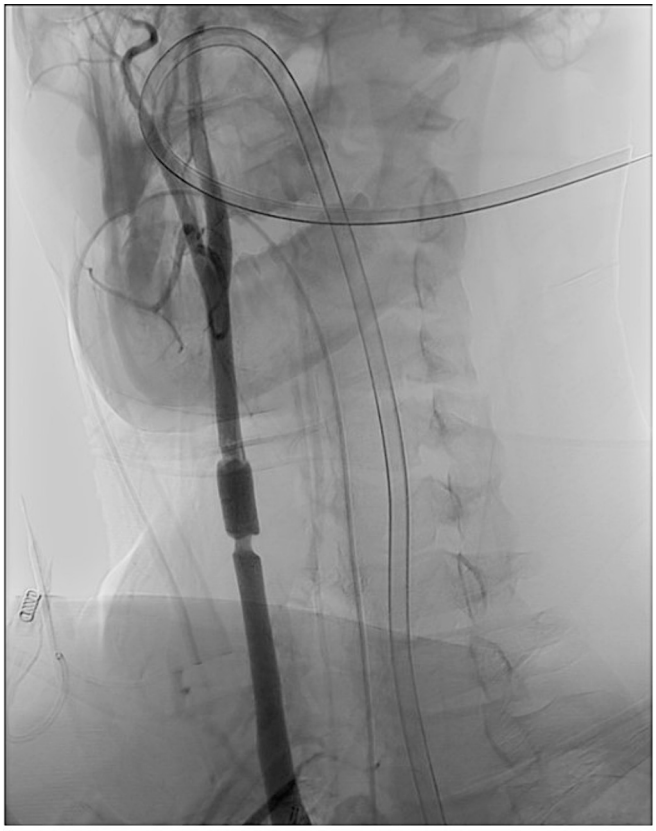

Fig. 2. *angiography* showing right common carotid artery injury with intima invagination.

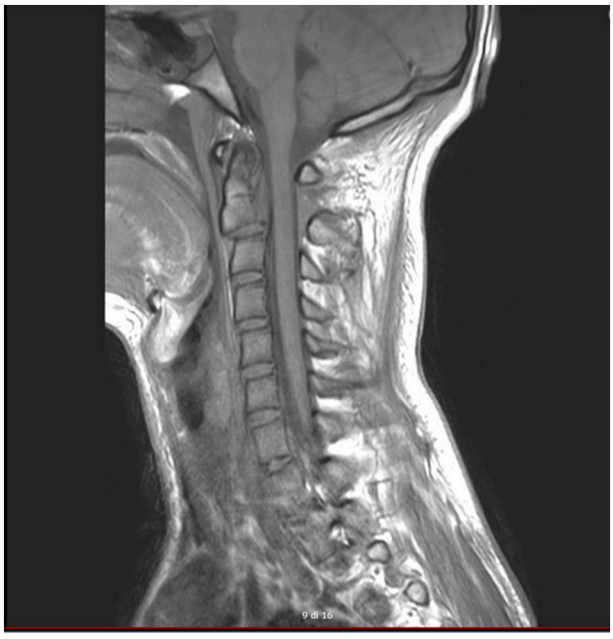

Fig. 3. Sagittal MRI showing anterolisthesis of C2 over C3.

Discussion

TS is today considered a rare but well-defined condition characterized by concurrent unilateral recurrent laryngeal and hypoglossal nerve palsy. <100 cases are described in literature and by far the most frequent cause is endotracheal intubation during general anesthesia [10].

The damage may occur both at a central level with involvement of the tegmentum and of the vagus and hypoglossal nuclei [7], and along the peripheral nerve route subsequently to a trauma of the oropharynx and the upper cervical spine [8,9].

However, the central form is extremely rare, as described in a recent review that analyzed TS cases between 1990 and 2020; the same review confirmed that peripheral lesions-related TS were mainly due to post-intubation edema (77 %) [11].

Tapia's early 19th century description of a bullfighter struck by a bull's horns included right side paralysis of the larynx and tongue and contralateral hemiplegia, and it was compatible with a central lesion, although the cause of the hemiplegia has not been established in Tapia's case, being potentially caused by embolism from a traumatized carotid artery or dissection of the vertebral artery with brainstem infarction.

Other rare cases of central TS have been described, related to vertebral artery aneurysm [12], brainstem infarctions [13,14], neurosyphilis [15], and brainstem tumor [7].

In peripheral TS, different injury mechanisms affecting the aforementioned nerve routes can be identified, and most commonly it occurs during surgery requiring general anesthesia and orotracheal intubation [17], due to stretching and compression of both nerves in the region of the oropharynx and hypopharynx [18], secondarily to difficult laryngoscopy [19], tracheal tube misplacement [20], *endotracheal tube* cuff *overinflation*, and extubation with an inflated cuff [1,17–19,21].

Prolonged intubation, repeated cycles of pronation, improper neck position during general anesthesia, are risk factors for peripheral TS, as shown in reports in intensive care units, cervical spine surgery, and shoulder arthroscopy [20,22–24]. Predisposing anatomical factors, such as an enlarged hyoid bone, cannot be excluded, as hypothesized by some authors [17,20].

Other causes include neck trauma, neoplasm or vertebral and carotid artery dissection [19].

While usually manifesting as dysphonia, tongue deviation, and swallowing difficulty, in the most severe forms TS cause permanent and irreversible damage with the need for tracheostomy and tube feeding [16].

The case described, due to the favorable outcome without cognitive and motor deficits, falls into one of the rare peripheral and bilateral TS, but identifying the precise mechanisms underlying the nerve damage is challenging for us. The patient was intubated upon admission to the hospital with the aid of a video laryngoscope to avoid neck mobilization, and subsequently tracheostomized, but the cuff pressure was constantly monitored and kept within an optimal range.

The right common carotid artery injury, as well as the neck trauma and the resulting Hangman's fracture, could have led to bilateral and simultaneous nerve damage.

Furthermore, our patient did not undergo anterior cervical spine surgery, having opted for conservative treatments, while other authors describe TS cases associated with spinal surgery [22–24].

However, the involvement of multiple mechanisms in determining nerve palsy, i.e., both carotid trauma, cervical fracture, and tracheal intubation, cannot be ruled out, although measures have been taken to prevent injury due to neck mobilization and cuff overinflation.

Furthermore, it is undoubtedly difficult to pinpoint the exact moment of the nerve damage, since the patient arrived at the hospital unconscious, a condition that continued even during his stay in intensive care. TS is a clinical diagnosis and therefore risks being made late in unconscious patients. In patients undergoing orotracheal intubation, the onset of symptoms usually appears immediate after extubation and includes hoarseness, difficulty speaking and swallowing, and tongue lateral deviation.

CT and MRI are useful in ruling out central causes of TS, while visual confirmation of nerve palsy can be obtained by fiber optic laryngoscopy and swallowing tests, during which vocal cords paralysis and accumulation of saliva in the pyriform sinus are evident.

In case of clinical suspicion, airway endoscopy can provide a safe and reliable diagnosis, thus avoiding the unnecessary use of CT, MRI, barium swallow test or electromyography [25].

For the correct management of this syndrome, a multidisciplinary approach that includes anesthetists, otolaryngologists, neurologists, physiotherapists and speech therapists is recommended. Tapia syndrome therapy is mainly rehabilitative, the timeliness of which can be decisive for a better and quicker result. Early involvement of speech and language therapy and the establishment of a structured swallowing rehabilitation program are crucial for a good outcome.

In conclusion, we believe it is useful to report our case to make a contribution to increasing awareness of TS and avoiding unnecessary diagnostic tests, as well as accidents during the extubation or decannulation phases, such as suffocation and aspiration pneumonia. Furthermore, the diagnosis is often delayed in comatose patients and a high level of suspicion should be maintained in patients with cervical spine trauma.

CRediT authorship contribution statement

Francesco Perrotta: Conceptualization, Investigation, Supervision. **Donato Piscopiello:** Conceptualization, Investigation. **Gaetano Iosa:** Conceptualization, Methodology. **Daniele Gemma:** Conceptualization, Formal analysis, Writing – original draft. **Daniela Rizzo:** Conceptualization, Investigation. **Francesca De Salvo:** Conceptualization, Investigation. **Davide D'Antini:** Writing – original draft, Writing – review & editing. **Emanuele Scarano:** Investigation, Supervision. **Fabio Colonna:** Investigation, Supervision.

Declaration of competing interest

The authors have no interests to declare.

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