

# Pediatric Horner's syndrome secondary to internal jugular venous cannulation: case report

Síndrome de Horner pediátrica secundária à canulação da veia jugular interna: relato de caso

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## KEYWORDS:

Horner's syndrome; Children, Sympathetic nerve; Injury; Jugular vein.

## PALAVRAS-CHAVE:

Síndrome de Horner; Crianças; Nervo simpático; Lesão; Veia jugular.

## ABSTRACT

Horner's syndrome, a disorder associated with ipsilateral damage to the sympathetic pathway, is a rare complication of cannulation into the internal jugular vein. Depending on the intensity of the injury and location, the damage may be irreversible, even in young children.

## RESUMO

A síndrome de Horner, um distúrbio associado a lesão ipsilateral da via simpática, é uma complicação rara da canulação na veia jugular interna (VJI). Dependendo da intensidade da lesão e da localização, o dano pode ser irreversível, mesmo em crianças pequenas.

## INTRODUCTION

Horner's syndrome is classically described as a triad of miosis, blepharoptosis and anhidrosis, but may also be present enophthalmos, pseudo enophthalmos (due to ptosis of the upper lid and elevation of the lower lid), hyperemia of the cheek and iris heterochromia on the ipsilateral side of the lesion to sympathetic nerve along its course from the hypothalamus to the eye<sup>1</sup>.

In pediatrics, Horner's syndrome is classified as congenital or acquired. The diagnosis is important for subsequent etiologic diagnosis by providing therapeutic measures and follow-up<sup>1,2</sup>.

Clinical evaluation and neuroimaging (CT/MRI and angiography) are essential to differentiate inflammatory, neoplastic or traumatic causes<sup>3</sup>.

We report a rare case of pediatric and complete Horner's syndrome secondary to internal jugular vein cannulation where there was no regression of the sequelae even after one year.

## CASE HISTORY

A seven-month-old boy, preterm twin, was examined to our institution two months after admission to another hospital for bronchiolitis. According to the mother, the child needed central venous access due to the impossibility of peripheral access for medication administration. Initially, peripherally inserted central catheters (PICC) was tried without success, so the centrally inserted venous catheters (ICRC) were chosen. A day later, the patient's mother noticed her

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son's right eyelid ptosis. An ophthalmologist has been consulted and the child was referred to neuro-ophthalmology department. On examination: upper eyelid ptosis, facial anhidrosis (Figure 1), and miosis visible in scotopic environment [left pupil (4mm) greater than right (2mm) Figure 2] ipsilateral to the puncture site. Anisocoria was not shown in photopic environment. There were no alterations found both in the oculomotricity exam and biomicroscopy. The clinical evaluation concluded a diagnosis of Horner's syndrome. Despite the history of ICRC suggesting a traumatic mechanism, the investigation with imaging exams was performed. Two months after placement of CVC, cerebral and cervical magnetic resonance imaging (MRI) with angiographic sequences, chest CT and VJID Doppler US was also normal. No ocular pharmacological tests or urine catecholamine testing were performed. Excluding neoplastic and inflammatory causes, traumatic lesion of oculo-sympathetic pathway was admitted. We concluded that it

was a case of complete Horner's Syndrome induced by mechanical trauma during IJV cannulation with damage to a second order neuron of the sympathetic nervous supply. Conservative management with periodic monitoring was carried out. One year after diagnosis, there is no evidence of visual impairment or damage to the patient's psychomotor development when compared to other children of the same age group and his identical twin who does not have Horner's syndrome.

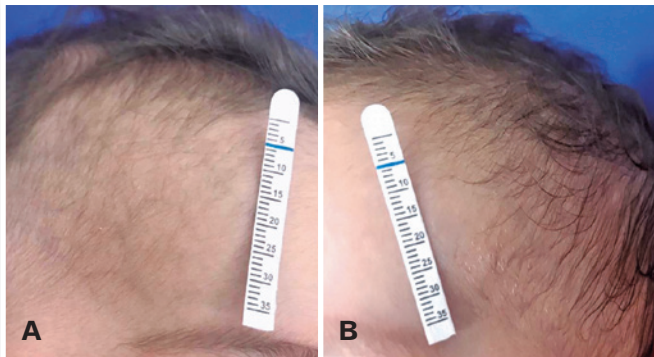
## DISCUSSION

Horner's syndrome is classically known as unilateral blepharoptosis, miosis with variable degrees of facial anhidrosis and enophthalmos may also be presente. The syndrome results from ipsilateral interruption of the sympathetic innervation<sup>4,5</sup>.

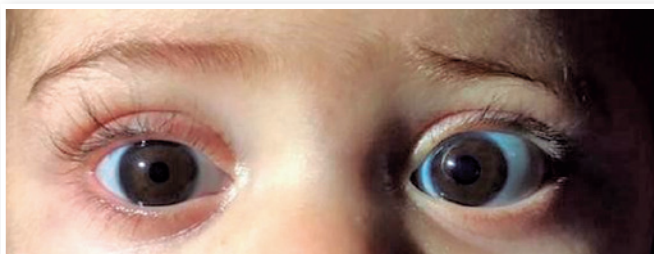
In youngsters the incidence of 1.42 per 100,000 had been estimated, where 40% were congenital, 42% were acquired after a surgical procedure in the thorax, neck, or central nervous system, and 15% were acquired from causes that included neuroblastoma, spinal cord tumors, rhabdomyosarcoma embryonal cell carcinoma, vascular malformations, intrathoracic aneurysm and trauma<sup>6</sup>.

In the present report, we demonstrate a case of pediatric Horner's syndrome induced by a central venous catheter, caused by direct trauma. Similar cases have been reported, usually secondary to anatomic abnormalities or complications of puncture technique.

During PICC, a peripheral vein is punctured and a device passed until reaching the distal third of the superior vena cava or proximal third of the inferior vena cava. This technique is advantageous due to its lower cost, ease of catheterization and lower risk of infection, but it is not preferable if large volume of fluid is required. In this situation, as in the present case, a large central vein is directly accessed (ICRC). The right internal jugular vein is preferred in children because of the lower risk of pneumothorax and easier compression in case of hematomas. However, in this case, the injury to the sympathetic pathway may have occurred due to respiratory distress caused by the acute bronchiolitis, psychomotor agitation and difficulty in sedating the child during insertion. In addition, US was not used during the procedure, a non-mandatory resource, which would reduce the risk of complications.



**Figure 1.** First day of follow-up. Right anhidrosis. Right, dry hair and skin (A) compared to left side with wet hair and skin (B). Schimmer tapes were used to try to demonstrate the difference, the tape is more rectified to the skin on the left side.



**Figure 2.** Three-month follow-up. Right-sided Horner's syndrome. Image captured with indirect lighting in the scotopic environment showing upper eyelid ptosis end miosis [left pupil (4mm) greater than right (2mm)].

The sympathetic trunk is closely related to the carotid sheath in its posterior wall and 18% of patients younger than six years have anomalous IJV anatomy, mostly IJV overlapping the carotid artery<sup>7</sup>. To avoid complications, the literature highlights factors that decrease the risk of damage to the sympathetic trunk: professional experience, patient positioning, sedation and use of ultrasound (USG). The right internal jugular vein (IJV) has several advantages as a cannulation site, having a predictable anatomy, even in children. Although the posterior approach is technically more difficult a 98% success rate of cannulation has been reported using the posterior approach in infants and children aged one day to 12 years<sup>8</sup>. Especially in pediatric cases, sedation is important to decrease psychomotor agitation.

The symptoms presented depend on the location of the sympathetic ocular pathway. In the exposed case, the presence of anhidrosis, absence of psychomotor damage or involvement of cranial nerve associated with the topography of the trauma suggests involvement in the second order neuron. The sympathetic nervous supply to the eye originates in the posterolateral hypothalamus. The first order neurons (central) descends to synapse, with second-order neurons at the ciliospinal center (Budge) between C8 and T1. These second order neurons (preganglionic fibers) pass over the pulmonary apex, ascend in the neck in the cervical sympathetic chain behind the common carotid artery outside the carotid sheath, ascend through the stellate and middle cervical ganglia and synapse with third order neurons (postganglionic) in the superior cervical ganglion, located at the angle of jaw<sup>1,3</sup>.

For confirmation diagnosis, localization of injury in the oculosympathetic pathway and etiologic elucidation, clinical evaluation, pharmacological and laboratory tests and neuroimage can be used. Especially in pediatric cases, ruling out malignancy is the main objective.

In this case, in which the clinical signs show a complete Horner's syndrome initiated after ipsilateral trauma by IJV cannulation, no ocular pharmacological tests (cocaine, apraclonidine, or hydroxyamphetamine) or urine catecholamine testing were performed. The neuroimaging study was performed to exclude other injuries and to identify complications with the insertion of a central catheter that could justify the syndrome.

The most recent protocol for image investigation in Horner's syndrome includes magnetic resonance

imaging of the head, neck and upper chest, the angiogram of the neck vessels in suspected post ganglionic involvement or without probable cause. CT is mostly indicated if a traumatic injury of the skull base is suspected<sup>3,5</sup>.

George et al. recommend only images of the head, neck and chest for patients over one year of age at the time of presentation<sup>2</sup>. For patients under one year of age, they recommend urinary catecholamine and examination for cervical or abdominal masses, but Jeffery et al. mentions that abdominal investigation should be acquired in cases without known injury (that is, surgical or birth trauma)<sup>9</sup>.

Corroborating with the neuroimaging study carried out in the present report, Barrea et al state that history of trauma or surgery may reduce the need for extensive systemic evaluation<sup>5</sup>. However, it's important make sure that require thorough evaluation should be imaged in patients in whom the clinical examination does not provide enough information to locate the lesion with certainty or there is no probable cause because of the frequent association of serious underlying disease.

Three months after IJV cannulation, maternal observation and clinical examination showed that the hair on the right side was smoother than the contralateral. During presentation or evaluation, no enophthalmos, pseudo enophthalmos, hyperemia of the cheek or iris heterochromia have been reported. At 12 months of age, the child maintains right ptosis, miosis and anhidrosis with no resolution, but the hair is bilaterally curly. Other studies without neuroimaging changes with probable direct damage of the needle in the sympathetic trunk have had different results. A six-year-old child had regression of ptosis and miosis 18 days after developing Horner's syndrome and a five-year-old boy had complete remission of the symptoms after five months<sup>3,5</sup>. In a younger child, seven-month-old, at the 12-month follow-up slight right eye ptosis could still be noticed and anisocoria was completely resolved<sup>10</sup>.

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