

## A Case Report on Reversible Horner's Syndrome as a Complication of Left Internal Jugular Vein Catheterization

Fadel Al-Rowaie<sup>1</sup>, Khalid Al-Matham<sup>1\*</sup>, Abdulaziz Al-Atmi<sup>2</sup> and Lina Al-Sharif<sup>2</sup>

<sup>1</sup>Department of Nephrology, King Fahad Medical City, Riyadh, Saudi Arabia

<sup>2</sup>Department of Medical Sciences, King Saud Bin Abdulaziz University, Riyadh, Saudi Arabia

\*Corresponding author : Khalid Al-Matham, Department of Nephrology, King Fahad Medical City, Riyadh, Saudi Arabia, Tel: +966563345878; Fax: +966112976600; E-mail: khalidosis@hotmail.com

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### Abstract

Horner's syndrome, characterized by clinical signs including ptosis, pupillary miosis resulting in anisocoria, and facial anhidrosis, is a rare ocular complication following internal jugular vein catheterization. We report a case occurring in a 23-year old female who developed acute left-sided Horner's syndrome after uneventful insertion of a left-sided permanent dialysis catheter. Ultrasound revealed no evidence of hematoma or carotid dissection. The catheter was left in place, and her symptoms resolved completely after 2 days. Clinicians should be alert for Horner's syndrome in these patients, to assure appropriate diagnosis, documentation, and follow-up are provided, aiming at greater understanding of risk factors for and potential consequences of this rare complication.

Establishing reliable vascular access is essential for successful hemodialysis in patients with end stage renal disease (ESRD). A cuffed, tunneled catheter (permacath) is often used for prolonged renal replacement therapy, with internal jugular vein access preferred due to its accessibility, low malposition rate, and overall low complication rate. However, up to 35% of internal jugular vein catheterization attempts across all indications may be unsuccessful, and complication rates up to 19% have been reported.

Horner's syndrome is a rare ocular complication that may occur after internal jugular vein catheter insertion, which usually resolves spontaneously. Because of its rarity, only a paucity of information is available about its specific etiology and course. Additionally, Horner's syndrome is considered to be underdiagnosed. Awareness of this condition is important to foster appropriate diagnosis, documentation, and follow-up for patients who develop its signs. Accordingly, we report a self-limiting case of Horner's syndrome that developed after internal jugular vein permacath insertion, and compare it with other cases reported in the literature.

**Keywords:** Horner's syndrome; Internal jugular vein; Hemodialysis

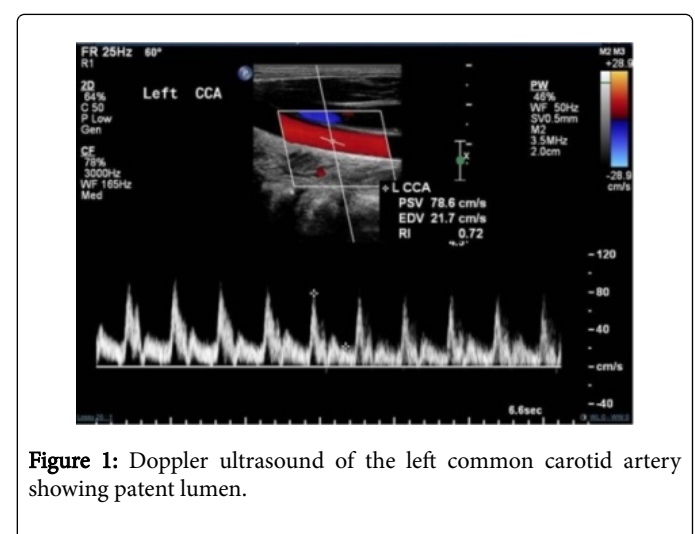
ultrasound ruled out carotid artery dissection and hematoma at the catheter insertion site (Figures 1 and 2).

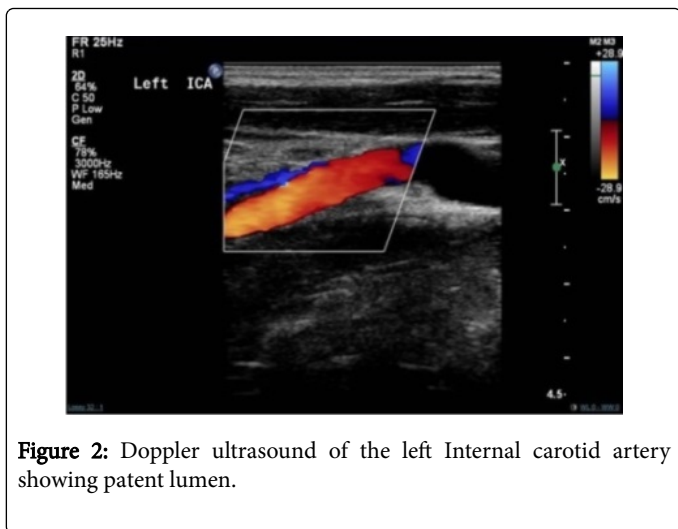
### Case Presentation

A 23-year-old Saudi female with ESRD of unknown primary cause was on regular hemodialysis 3 times per week through a right internal jugular permacath. Three weeks prior to her current admission to our hospital she was diagnosed with line-related sepsis and given empirical intravenous antibiotics as an inpatient at her previous hospital. Her permacath was removed 4 days prior to presenting to our hospital, following self-discharge against medical advice to seek a second opinion. On arrival at our hospital, she was vitally stable, with no erythema or discharge at the exit site of the previously removed permacath. She was initially dialyzed through a temporary right femoral catheter. After 3 days, a venogram revealed occluded right distal jugular and brachiocephalic veins. A left internal jugular permacath was inserted under fluoroscopic guidance. The procedure was uneventful, without arterial puncture. After 2 days the patient complained of left eye swelling [1-3].

### Clinical findings

The patient was examined by an ophthalmologist, who observed ptosis, miosis, enophthalmos, and anhidrosis in her left eye. An urgent





**Figure 2:** Doppler ultrasound of the left Internal carotid artery showing patent lumen.

### Diagnostic assessment

The ophthalmologist diagnosed the patient as having left-sided Horner's syndrome; that is, ipsilateral to the internal jugular vein catheter.

### Therapeutic intervention

No intervention was provided. In addition, in the absence of any evidence of complications at the site of catheter insertion, the catheter was left in place.

### Follow-up and outcome

Two days after observing signs of Horner's syndrome (4 days after catheter insertion), all signs were resolved.

### Discussion

With few exceptions, our understanding of Horner's syndrome following internal jugular vein catheterization has been acquired from case reports [4-6]. Many cases were associated with failed or difficult catheterization [6]. Dissection of the carotid artery, with associated risk of severe cerebrovascular complications, has been implicated as a possible cause of Horner's syndrome. Accordingly, it was essential to exclude carotid artery dissection in our patient. In addition, compression from a hematoma developing at the cannulation site may cause direct damage of the stellate ganglia or its associated sympathetic neurons that supply the eye, which are embedded in the carotid sheath with the internal jugular vein [5,6]. Ultrasound failed to show a hematoma in our patient. However, nerve compression may explain Horner's syndrome occurrence following otherwise successful insertions, if intraprocedural pressure from a misdirected cannula or excess rotation of the head and neck was adequate to elicit direct trauma to the sympathetic plexus [5,7,8]. Although insertion of large bore hemodialysis catheters may be associated with a greater risk, Horner's syndrome has also been reported following insertion of temporary catheters [9].

Recent studies support that ultrasound-guided internal jugular vein cannulation has advantages compared with anatomic guidance, suggesting that procedures using ultrasound guidance may be less likely to develop Horner's syndrome. [1] Catheter insertion in our

patient was not based on ultrasound guidance, but was performed using fluoroscopic guidance [6]. A small prospective study of 100 consecutive patients with central venous line insertion via the internal jugular vein examined the incidence and characteristics of Horner's syndrome based on an ophthalmologic exam performed within 5 days after catheterization [4]. All but 2 insertions were made with real-time ultrasound guidance. The mean number of insertion attempts was 1.49 (range 1-6). Pharmacologic confirmation was performed for eyes with a suspicion of Horner's syndrome by instilling a drop of apraclonidine 0.5% in each eye and observing after 60 minutes for reversal of anisocoria. Apraclonidine-induced dilation of  $\geq 1$  mm in the smaller pupil confirmed a diagnosis. Anisocoria of  $\geq 1$  mm was observed in 9 patients and ptosis in 4. The 5 cases with anisocoria without ptosis tested negative for Horner's syndrome using apraclonidine drops. Of 4 patients with both anisocoria and ptosis, 2 did not have reversal of anisocoria with apraclonidine. One affected eye was contralateral to the internal jugular catheter, and one tested negative twice on 2 days examined after 3 hours as well as 60 minutes. In the 2 patients who were positive for Horner's syndrome, catheterization was successful on the first, uneventful attempt. Two patients with carotid artery puncture and 4 with local bleeding did not develop Horner's syndrome. The authors compared their 2% incidence with another prospective study that also observed 2% incidence in 66 patients following catheter insertion without ultrasound guidance. Although the authors emphasized the inability to draw conclusions from the historical comparison and small sample sizes, they noted that Horner's syndrome continues to be a possible complication even in the era of ultrasound-guided catheter insertion.

All of the signs that have been associated with Horner's syndrome may not be present in an affected patient, and post-procedure diagnosis may be complicated for patients with reduced levels of consciousness [5]. In addition, signs may be misinterpreted, which may result in unessential investigations. For example, in one report of a pediatric patient, anisocoria on admittance following transfer from another hospital was believed to reflect unilateral left-sided pupil enlargement, which resulted in obtaining computed tomography of the brain to rule out intracranial pathology [10]. Further examination revealed the pupillary size difference was due to miosis of the right eye. A physical exam revealed numerous puncture marks on the right side of the neck, and the transferred medical records noted a failed attempt to catheterize the right internal jugular vein. This case exemplifies that not all cases are first identified in a recovery or ICU setting; accordingly, familiarity with Horner's syndrome and its signs is particularly important for clinicians in all healthcare settings. Another patient with ESRD admitted through the emergency department (ED) had a right internal vein catheter successfully inserted on the second attempt while he was unconscious, and proper catheter positioning was confirmed by chest x-ray [4]. After 4 hours of hemodialysis, the patient remained stuporous, and was noted to have anisocoria. A brain CT scan was performed to rule out an intracranial bleed secondary to heparin use during hemodialysis, which was unremarkable. When his consciousness improved, he was noted to have partial ptosis, miosis, and enophthalmos. Ultrasound did not reveal hematoma or carotid artery dissection. Complete resolution was achieved after 6 weeks. Our patient was also observed to have enophthalmos, which was initially considered to be a characteristic sign of Horner's syndrome with normal ultrasound findings. However, some recent opinions suggest that an artifact related to the reduced eyelid aperture makes the eye appear to be sunken [6].

Once Horner's syndrome develops, its natural course is not predictable. Our case was identified 2 days after catheterization, and resolved after 2 days. However, the exact onset is not definitively known in the absence of specific post-catheterization monitoring. Three other reported cases were first observed in the recovery room [3,8,9]. One was a surgery patient who had her internal jugular vein catheter removed and was discharged on day 2, with complete resolution of Horner's syndrome when she returned for a day 7 follow-up visit [9]. Improvement was noted after 18 days in another case, which was a 6-year old girl admitted for wound debridement under general anesthesia [8]. In the absence of a hematoma, the authors of both case reports believed direct trauma from the needle was responsible for Horner's syndrome development. The third case had a known hematoma, which was noted after several failed attempts finally resulted in successful insertion of a temporary hemodialysis catheter [3]. Although the patient noticed uncontrolled tears, ptosis, pain, and swelling in the eye immediately after catheter insertion, he did not report them and was sent home after dialysis without clinical staff being aware of his signs. Ptosis was noticed by healthcare staff when he presented for his subsequent hemodialysis after 3 days. Resolution was complete after 4 weeks. Another case, a 5-year old needing a central venous catheter for parenteral nutrition and antibiotics, had a right internal jugular vein catheter inserted on the fourth attempt, after the third attempt ended in carotid artery puncture [7]. Three days later, a diagnosis of Horner's syndrome was made after the child's mother noted anisocoria, with the right pupil smaller than the left, and subsequent identification of ptosis and miosis. There was no evidence of a hematoma or carotid artery dissection on MRI and magnetic resonance angiography. The ptosis improved after 1.5 months, with complete resolution after 5 months. Because of the delayed resolution, the authors believed the condition was the result of direct trauma from the needle. The varying time intervals until resolution was exemplified in a review of pediatric Horner's syndrome cases attributed to internal jugular vein cannulation, which reported that 9 cases had complete recovery after intervals ranging from 24 hours to 5 months [10].

## Conclusion

In conclusion, our case of this rare complication of internal jugular vein catheterization was of brief duration, and resolved while the patient remained hospitalized. As with our case, some reports were identified in response to patient complaints. We believe that monitoring for Horner's syndrome can easily be added to post catheterization management, and that diligent routine pupillary examinations should be performed for all patients after catheter insertion. With the current wide range of characteristics observed in reported cases, gaps in our understanding of the natural course of Horner's syndrome, and lack of information regarding its possible

association with subsequent events, better monitoring is warranted. Attention to diagnosing, documenting, and providing appropriate follow-up for Horner's syndrome cases will provide larger cohorts in which to investigate risk factors for and possible consequences of Horner's syndrome. More reports based on case series instead of single cases should become possible. Finally, in addition to awareness of Horner's syndrome in the recovery room, ICU, and ED, ophthalmologists with patients who present with signs of Horner's syndrome should ask about recent central vein catheterization to assist with their diagnosis and management.

## Conflict of Interests

The authors declare that there is no conflict of interests regarding the publication of this paper.

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