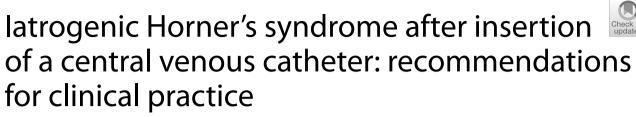
CASE REPORT

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Abstract

Background: Horner's syndrome is a rare complication of central venous catheter (CVC) insertion. It comprises a triad of unilateral ptosis, ipsilateral meiosis with a normally reactive pupil, and ipsilateral facial anhidrosis of variable spread.

Case presentation: A 25-year-old female who underwent CVC insertion developed ptosis, reverse ptosis, meiosis, dilational lag, and enophthalmos after her procedure. The patient was diagnosed with a case of Horner's syndrome attributed to occulosympathetic damage due to CVC in the catheter, after exclusion of all other sources. The patient was counseled regarding the etiology of the disease and advised monthly follow-up.

Conclusions: Horner's syndrome is an uncommon complication that can easily be prevented by corrections in technique and experience. The study emphasizes the establishment of diagnostic and preventive principles for HS.

Keywords: Occulosympathetic palsy, Bernard-Horner Syndrome, Anisocoria, Ptosis, Meisis, Anhidrosis

Background

Horner's syndrome (HS), typically described as a triad of unilateral ptosis, ipsilateral meiosis with a normally reactive pupil, and ipsilateral facial anhidrosis of variable spread, which occurs due to a disruption in the sympathetic supply of the eye [1]. The Swiss ophthalmologist, Johann Friedrich Horner, was the first person to fully describe the condition in humans and attribute it to oculosympathetic paresis [2].

Catheterization by the central venous catheter (CVC) is a common procedure typically done with a Swan-Ganz catheter, mostly inserted into the internal jugular vein to assist in intravascular resuscitation. HS is one of the rarest complications of CVC insertion [3]. Here, we present a case of possible iatrogenic injury due to CVC insertion,

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leading to oculosympathetic palsy or HS. To our knowledge, this is the first case report of iatrogenic HS outlining recommendations to reduce the incidence of this complication.

Case presentation

A 25-year-old primiparous woman was admitted to the high dependency unit (HDU) after a lower segment cesarian section (LSCS) under spinal anesthesia at 36 gestational weeks. Per-operatively, the patient had an uncontrolled post-partum hemorrhage, ultimately requiring a partial hysterectomy. As a preparatory measure for her surgery and anticipated post-partum hemorrhage, an intensivist inserted a single-lumen CVC (7 Fr x 20cm) in the patient's right internal jugular vein (IJV) under ultrasound guidance.

On her first post-operative day, the patient noticed that her right upper eyelid is drooped and her right eye

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appeared smaller than the left one. Owing to this, neurology and ophthalmology consults were made.

On examination, her unaided visual acuity was 6/6 bilaterally. She had right-sided enophthalmos of 3 mm on Hertle's exophthalmometer, right-sided blepharoptosis, and reverse ptosis (MRD1 = 3 mm, MRD2 = 2.5mm), a PFH of 5.5 mm and a mild right-sided periocular swelling, as seen in Fig. 1. In the left eye, the PFH was 7.5 mm, MRD1 was 4 mm and MRD2 was 3.5 mm. Pupillary examination revealed anisocoria in room light (OD: 2 mm, OS: 3 mm) which increased in the dark room (OD: 3mm, OS: 4.5 mm). On direct light reflex, there was a dilation lag in the right eye; however, there was no relative afferent pupillary defect (RAPD). The Hirschberg corneal reflex test was bilaterally central. The coveruncover test showed bilateral esophoria. Her extra-ocular movements were within her full range of motion bilaterally, and she had no diplopia in any of the gazes. The rest of her anterior and posterior segment examinations were



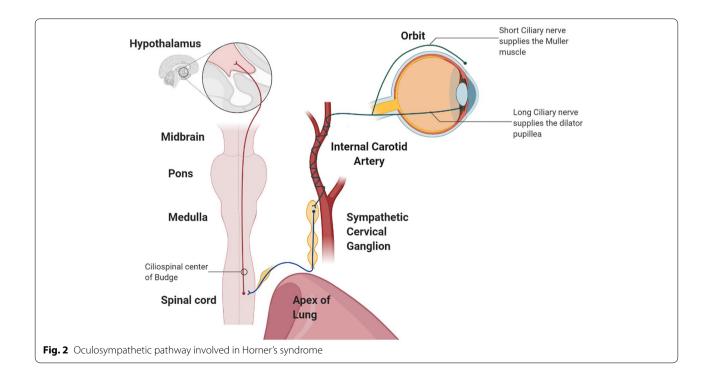
unremarkable. The general physical examination revealed a central venous line in the right internal jugular vein and associated neck swelling at the insertion site. No other neurological deficit including abnormal eye movements, ipsilateral limb ataxia, or dissociated sensory loss was noted.

The MRI of the brain and cervical spine came out to be unremarkable in establishing any pathology. A diagnosis of HS due to the central venous line was based on the exclusion of other pathologies. The patient was counseled regarding the etiology and possible course of the disease. She was advised to come for a monthly follow-up at our neuro-ophthalmology clinic.

Discussion

CVC insertion is performed on millions of patients per day with well-documented complications [1, 3]. The incidence of HS due to CVC is reported to be 2% by Butty et al. in his prospective study [3]. Any disruption within the oculosympathetic tract causes HS, which may occur within the first-order (central), second-order (preganglionic), or third-order (postganglionic) neurons as seen in Fig. 2 [1].

The sympathetic first-order neurons originate in the posterolateral hypothalamus and run through the lateral brainstem and into the intermediolateral gray column of the spinal cord at C8-T1, where they are part of the ciliospinal center of Budge. The second-order neurons pass across the pulmonary apex into the cervical sympathetic



chain in the carotid sheath. The third-order neurons originate in the superior cervical (stellate) ganglion from where they travel up the adventitia of the internal carotid artery into the cranium. Here, the fibers divide into long and short ciliary nerves to reach the target tissues [1].

There are multiple causes of HS divided into those affecting first-order neurons (central tumors, strokes, demyelinating disorders, syringomyelia, neck trauma, etc.), second-order neurons (Pancoast tumor, schwannomas, trauma, etc.), and third-order neurons (trauma, tumors, infection, cluster headaches, etc.) [1]. They are enlisted in detail in Table 1. Maloney et al. reported that in about 450 patients with HS, only about 65% (270 individuals) had an identifiable etiology, of which 13% were central, 44% preganglionic, and 43% postganglionic [5].

The right IJV is the preferred side for CVC insertion because it has predictable anatomy, a high success rate, and a low risk of complications [6]. These complications include carotid artery aneurysm (most common), aortic dissection, thrombosis, arteriovenous fistula formation, pneumothorax, and HS [6]. Ways in which a CVC may cause HS include direct damage to the sympathetic chain, direct damage to the peri-subclavian neural loops, and hematoma of associated vasculature (especially if the hematoma is trapped within the carotid sheath) [3–6].

Clinical features of HS include ptosis (with or without reverse ptosis) and meiosis (more apparent in darkness), which occur due to paresis of muller and dilator pupillae muscles, respectively. In addition, anhidrosis may occur due to the involvement of sympathetic innervation of facial sweat glands [1]. Features in addition to these may be seen with respect to the underlying cause as seen in Table 1. Diagnosis of HS is made by pharmacological tests including 5% cocaine (normal pupil dilates, Horner pupil unresponsive), 0.5% Apraclonidine (normal pupil minimally constricts, Horner pupil dilates), 0.5% hydroxyamphetamine (normal pupil dilates, postganglionic Horner pupil unresponsive), and 1% phenylephrine or 2% epinephrine (normal pupil unresponsive, postganglionic Horner pupil dilates) [4, 7].

Once a diagnosis of HS is considered probable, depending on the clinical features, a multitude of tests may be carried out, including but not limited to, X-ray cervical spine, MRI of the brain, CT scan of the chest, and/or CT/ MRI angiography, to identify the underlying pathology [1, 8].

Here, we enlist the following recommendations to reduce the likelihood of developing HS after CVC:

- A CVC should be inserted under the guidance of direct ultrasound, as it has been shown to reduce the likelihood of developing HS. The cervical sympathetic chain itself can be identified via ultrasound [3]. The chain may be identified lying medial to the scalene muscles; lateral to the longus colli, esophagus, and trachea; superior to the subclavian artery; and posterior to the pleura and vertebral vessels [9].
- 2. Avoid keeping the needle angle too steep; otherwise, the needle may pass behind the carotid artery to damage the sympathetic chain [6].
- 3. Once the head is tilted to the left by more than 40°, the risk of IJV overlapping the carotid artery is increased significantly, leading to greater chances of developing HS. Tilting the head within 30° and keeping the neck muscles relaxed leads to higher success rates and minimizes the patient's discomfort [5].
- 4. Testing for HS can be positive with apraclonidine as soon as 3 h after the insult. Pharmacological testing

 Table 1
 Causes and clinical manifestations with respect to level of lesion for Horner's syndrome (derived from multiple sources [1, 4, 5])

Level of the lesion	Possible cause	Additional clinical features
Hypothalamus	Stroke, tumor, trauma	Contralateral hemiperesis, Contralateral hyperesthesia
Thalamus	Stroke, tumor, trauma, demyelination	Contralateral ataxic hemiperesis, Contralateral hypoesthesia, vertical gaze paresis, dysphagia
Dorsal mesencephalon	Stroke, tumor, trauma	Contralateral trochear nerve paresis
Pons	Stroke, tumor, trauma	Ispilateral abducens nerve paresis (sometimes bilateral)
Lateral medulla	Stroke, tumor, trauma	lpsilateral ataxia. contralateral hyperalgesia, vertigo, dysphagia, nystagmus, facial nerve paresis
Spinal cord	Infarction, tumor, trauma, demyelination, myelitis, syringo- myelia, arterioveneous malformation	Ipsilateral or contralateral sensory dissociation, systemic dysautonomia
Apical lung	Pancoast tumor, schwaanoma, subclavian artery aneu- rysm, mediastinal tumor, cervical rib, trauma	Ipsilateral shoulder pain, paresthesias along the upper limb, Atrophy of small muscles of the hand
Internal carotid artery	Tumor, trauma, aneurysm, dissection, arteritis, thrombosis	Incomplete HS (absence of anhidrosis)
Skull base	Nasopharyngeal carcinoma, lymphoma, trauma	Incomplete HS, 'lump in neck', bloody rhinorrhoea, hearing loss
Cavernous sinus	Tumor, trauma, inflammation, thrombosis	Incomplete HS, headache, eye pain, diplopia

should be done as soon as possible and once HS is diagnosed and attributed to catheter insertion; the CVC should be removed by a specialist to limit further damage [5].

5. Posterior approach to IJV underneath the sternocleidomastoid muscle should be avoided as the likelihood of damaging the sympathetic chain increases with this approach. An anterior approach is recommended for the ease of procedure, limited complication rate, and an overall better outcome [6].

Conclusions

Conclusively, Horner syndrome is an uncommon complication of a common procedure, CVC insertion, that can easily be prevented with experience and correction in technique. The study also emphasizes the establishment of diagnostic and preventive principles for HS.

Abbreviations

HS: Horner's syndrome; CVC: Central venous catheter; LSCS: Lower segment cesarian section; IJV: Internal jugular vein; MDR1: Marginal-reflex distance at 1mm; MDR2: Marginal-reflex distance at 2mm; PFH: Palpaberal fissure height; OD: Oculus dexter; OS: Oculus sinister; MRI: Magnetic resonance imaging; CT: Computed tomography.

Acknowledgements

None to declare.

Authors' contributions

UI and TAK were the core members of the primary treating team of the patient and selected the case report. SZ did the extensive literature review and interpreted the patient data. SZ and NUZ wrote the first draft. UI, TAK, and MAZ critically reviewed the draft and corrected the manuscript for the final version. The authors approved of the final paper and take accountability for all aspects of the report.

Funding

None to declare.

Availability of data and materials

The dataset generated or analyzed during the current study is not publicly available due to patient confidentiality but is available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

An informed written consent to participate in the study was taken from the parents of the patient who are his legal guardians. Confidentiality was maintained at all levels of the study. Ethical approval was also obtained from the Hospital Ethics Review Committee.

Consent for publication

An informed written consent for publication was taken from the parents of the patient who are his legal guardians.

Competing interests

The authors declare that they have no competing interests.

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Received: 19 April 2022 Accepted: 24 April 2022 Published online: 15 July 2022

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