Delayed Onset and Prolonged Horner Syndrome in Two Children After Single-Shot Ultrasound-Guided Infraclavicular and Subclavian Perivascular Brachial Plexus Blocks for Upper Extremity Surgery: Case Reports

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Horner syndrome is a known complication of cervical approaches to brachial plexus blocks due to local anesthetic-induced oculosympathetic paresis. It has rarely been described in relation to ultrasound-guided brachial plexus blocks in children. This syndrome is usually self-limiting but may cause apprehension to the child, parents, and the treating physicians until its resolution. We report delayed manifestations of Horner syndrome in 2 children following ultrasound-guided infraclavicular and subclavian perivascular blocks.

Keywords: Brachial plexus block, delayed presentation, Horner syndrome, ultrasound.

Horner syndrome is a neurologic condition that consists of a triad of miosis, ptosis, and anhidrosis (Figure 1) The cause stems from paresis of the oculosympathetic nerves supplying the head and neck region. Horner syndrome is a common sequela of the more proximal approaches to brachial plexus blocks (BPBs) such as interscalene and periscalene blocks because of their proximity to paravertebral sympathetic fibers, which are easily blocked by medial diffusion of the local anesthetic (LA).[1,2] For similar reasons, other complications such as hemidiaphragmatic paralysis, hoarseness, and central neuraxial spread of LA are also known risks of these approaches. Hence, distal approaches to BPBs, such as supraclavicular and infraclavicular blocks, are preferred to provide analgesia or surgical anesthesia for upper extremity procedures other than shoulder surgeries.[2] Recent widespread availability of ultrasonography machines has permitted direct visualization of the nerve, needle, and LA spread and has ensured improved success rates (> 95%) with minimal complications.[1,3]

There is a paucity of literature describing Horner syndrome after ultrasound (US)-guided BPB in children. We describe an atypical presentation (delayed onset and prolonged effect) of Horner syndrome following US-guided distal approaches to BPB in 2 children. Written informed consent was obtained from the parents of both children for the publication of their clinical details and photographs.

Case Summaries

• *Case 1.* A 9-year-old boy, ASA physical status 1, weighing 30 kg, presented with complaints of weakness and deformity of his left hand. He had suffered a sharp cut to

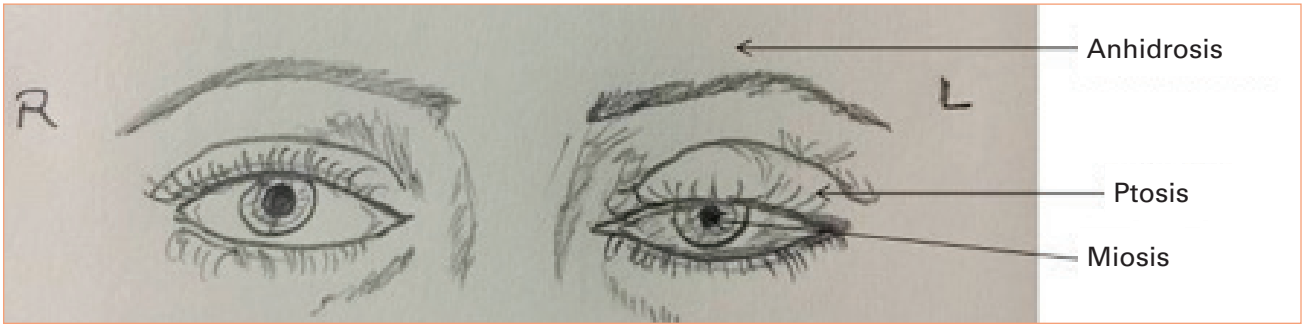

Figure 1. Diagrammatic Representation of the Features of Horner's Syndrome (Left Eye)

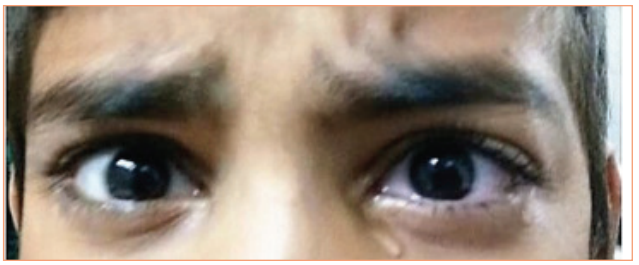

Figure 2. Horner's Syndrome Post Infraclavicular Block

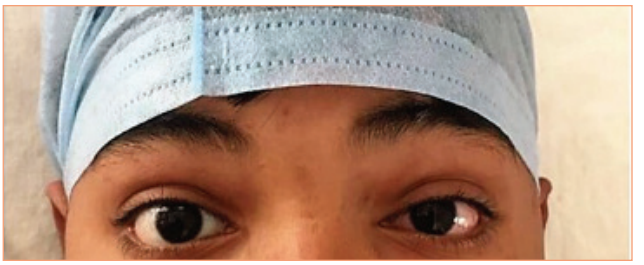

Figure 3. Horner's Syndrome Post Subclavian Perivascular Block

his left forearm 6 months earlier. Based on the findings of the clinical examination and nerve conduction tests, a diagnosis of ulnar nerve injury was made. He was scheduled for ulnar nerve exploration. The anesthetic plan was discussed with the surgeon and the parents, and it was decided to perform general anesthesia with infraclavicular BPB. Written parental consent was obtained.

A 20-gauge intravenous (IV) cannula was secured on the patient's nonoperative hand, and electrocardiogram, pulse oximeter, and noninvasive blood pressure monitors were attached. The child received fentanyl, 60 µg, and general anesthesia was induced with propofol, 60 mg. After adequate jaw relaxation was ensured, a classic laryngeal mask airway (size 2.5) was inserted, and spontaneous respiration was maintained.

Under aseptic precautions, a linear array probe (L25, 6- to 13-MHz transducer) of the ultrasonography machine (M-Turbo, SonoSite) was placed in a parasagittal plane immediately below the clavicle, medial to the coracoid process. A short-axis view of the axillary artery was obtained, and a 19-gauge (5-cm) Tuohy needle (Portex, Smiths Medical) was inserted in-plane from a cranial to caudal direction; its tip was positioned at around the 6-o'clock position in relation to the artery. Five milliliters of normal saline was used for hydrodissection to navigate the needle tip close to the posterior cord of the brachial plexus, and 12 mL of 0.25% bupivacaine with 15 µg of fentanyl was administered, with intermittent aspiration. Following this, a 20-gauge epidural catheter was threaded and secured in place using sterile adhesive dressing for further top-offs as required. Thereafter, the tourniquet was applied (baseline systolic blood pressure of 96 mm Hg and tourniquet inflated to a pressure of 180 mm Hg for 90 minutes). The surgery lasted 2 hours.

The child was extubated and was transported to the recovery room. He was pain free (visual analog pain score of 0) and discharged to the inpatient ward after 2 hours. Six hours after the block, partial ptosis developed along with miosis, conjunctival congestion, and watering in the ipsilateral (left) eye (Figure 2). He also complained of nasal stuffiness and warmth on the left side of his face. The parents and child became anxious about these side effects and were counseled about the self-limiting nature of the condition. The catheter was removed, and 30 mg of IV diclofenac was prescribed as needed. Four hours after the onset of symptoms, the ptosis and erythema decreased, and the next morning (16 hours after the block) the symptoms had subsided completely. Throughout this period, the child was pain free and did not require any other analgesics. His vital signs were stable, and he did not have any respiratory symptoms.

• ***Case 2.*** A 12-year-old boy, ASA physical status 1 (weighing 61 kg, 164 cm in height), was scheduled to undergo implant removal from the left elbow. Anesthetic options were discussed with the child and his parents, who consented for supraclavicular BPB.

On the morning of surgery, the patient was administered alprazolam, 0.25 mg orally. With use of aseptic techniques, a linear array L38, high-frequency (10- to 12-MHz) ultrasonography probe (Imagic Agile, Kontron Medical) was placed in midclavicular area above the clavicle. The brachial plexus was visualized as a "bunch of grapes" posterolateral to subclavian artery. A 22-gauge, 50-mm insulated nerve block needle (Stimuplex A, B Braun) was then inserted in-plane from a lateral to medial position. Hydrodissection with 3 to 4 mL of normal saline was done to ensure the placement of the tip of the needle in the corner pocket (between the subclavian artery and the first rib). Sixteen milliliters of 0.75% ropivacaine with 6 mg of dexamethasone was injected. The onset of motor and sensory blockade of the limb was appreciated immediately. The patient remained comfortable during the surgery and no additional analgesics or sedatives were required.

In the postanesthesia care unit, the patient complained of ipsilateral ptosis (approximately 3 hours from administration of the block) along with conjunctival erythema. On examination, ipsilateral miosis and warmth of facial skin was noted (Figure 3). There were no complaints of hoarseness or respiratory distress. The family and the surgeon were apprehensive about any nerve injury and were counseled. The boy remained pain-free and did not request any analgesics until the next morning. The ptosis and erythema subsided completely on the next day (16-18 hours from block administration), and this coincided with the sensory and motor block regression.

Discussion

Horner syndrome (oculosympathetic palsy) is character-

ized by miosis, ptosis, anhidrosis, enophthalmos, conjunctival injection, and nasal congestion. It may occur in 20% to 90% of adult patients after an interscalene approach to the brachial plexus because of interruption of the sympathetic outflow to the ipsilateral head and neck by diffusion of the LA in the prevertebral spaces.[1] The complication has infrequently been reported with more distal approaches. A recent study of US-guided infraclavicular block using the coracoids and a retroclavicular approach reported no case of Horner syndrome, whereas another study, describing outcomes of US-guided supraclavicular BPB, reported an incidence of 1% in 500 patients.[4,5] Moreover, this syndrome has rarely been described in relation to pediatric regional anesthesia practice.[3,6] In the present report, we reported 2 atypical cases of Horner syndrome in children following subclavian perivascular and infraclavicular approach to BPB.

The incidence of Horner syndrome is variable despite the same approach and dose of LAs.[1,2,4,7] This variability may be due to the maldistribution of LA in the brachial plexus sheath.[8] It has been proposed that a fascia might be dividing the brachial plexus sheath into 2 compartments facilitating unidirectional block and cephalad spread.[8] Recent anatomical studies have suggested that after supraclavicular BPB, the LA migrates both above and below the clavicle, whereas the LA remains confined to infraclavicular space following the infraclavicular approach.[9]

Traditionally, the axillary approach to BPB has been preferred for upper extremity analgesia in children because of a perceived higher incidence of complications with other approaches. In the era of ultrasonography, however, both supraclavicular and infraclavicular BPB have been revisited because of high success rates and improved safety profiles.[3] A large prospective regional anesthesia survey did not find any complication related to 997 upper limb blocks and suggested that these blocks are extremely safe in children.[6]

Ultrasound guidance reduces the LA requirement because of precise placement of the drug around the nerve bundles. This decreases the LA available to escape medially and cephalad toward the paravertebral space and, hence, reduces the incidence of Horner syndrome.[1,3,4,5] The use of ultrasonography was found to decrease the incidence of Horner syndrome compared with a conventional nerve stimulator–guided technique.[2] In a recent trial comparing US-guided supraclavicular with infraclavicular block in children above 5 years of age, no case of Horner syndrome was recorded.[3]

We had a rare experience of Horner syndrome in 2 cases involving US-guided BPB. The total doses of LA used in the present cases were well below accepted limits of LA (0.5-1 mL/kg) for supraclavicular and infraclavicular BPB in children.[3,10] In infraclavicular BPB, the deeper location of the plexus makes real-time visualization of the needle tip and the cords challenging. An injection of small amounts of a saline solution (hydrodissection) may improve the visualization of the nerve bundles and the needle tip. This can be particularly useful for BPB in which nerve bundles lie adjacent to hypoechoic vessels, in that demarcation of the nerve borders and their separation from the vessels improves the safety of the procedure.[11] In children, the total volume of LA that can be administered is limited by their body weight, and its use for hydrodissection may lead to its waste in case the needle tip is not in the correct place. However, the use of normal saline for hydrodissection might have added to the total volume and probably facilitated cephalad migration of LA.

In the patient who received supraclavicular BPB, a high concentration of LA (0.75% ropivacaine) might have facilitated the blockade of sympathetic fibers and led to Horner syndrome. Also, we had used dexamethasone to prolong analgesia, and this may have potentiated the blockade of the sympathetic fibers too, thereby prolonging their blockade. There has been some concern about the neuronal toxicity of dexamethasone as an adjuvant to LA.[12] To our knowledge, however, there is no evidence of any long-term neurapraxia with use of dexamethasone in the literature. A recent meta-analysis has concluded that perineural use of LA with dexamethasone prolongs the effect of BPB without any adverse effects.[13]

A recent trial assessing the development of Horner syndrome after interscalene BPB found a significantly higher incidence in younger patients.[1,2] They concluded that a higher water content of the prevertebral spaces of children facilitates the diffusion of ropivacaine and subsequent development of Horner syndrome. This mechanism might have contributed to the development of Horner syndrome in the second child.

In our case reports, the use of a tourniquet in the patient who was administered an infraclavicular BPB and a history of humeral fracture in the other patient might have obliterated the axillary sheath distally and increased the proximal migration of LA. This may have contributed to the prolonged duration of oculosympathetic paresis.

Horner syndrome typically occurs within 1 hour of the block and remits in 1 or 2 hours, but in our cases, the syndrome manifested late (3 hours and 8 hours) and lasted 8 hours and 18 hours after the block, coinciding with the dissipation of the motor and sensory block. Continuous LA infusion through a catheter may lead to delayed manifestation of Horner syndrome, as reported by Salengros et al,[14] who noted its onset approximately 50 hours after an infraclavicular block. In our case, infusion was not started through a catheter, and the catheter was removed after the onset of the symptoms.

Horner syndrome has been reported to persist for prolonged periods (up to 6 months) in a few cases.[15,16] In these cases, the facial distortion due to ptosis can lead to medicolegal litigation and unnecessary treatments.

The unexpected development of these symptoms can

be distressing, especially for the children, and pose a diagnostic dilemma for their clinicians. Despite the increased safety of the BPB with the advent of ultrasonography, all the risks associated with these blocks including Horner syndrome should be explained in detail at the time of obtaining informed consent. In case this complication occurs, the family should be thoroughly counseled regarding the self-limiting nature of the symptoms, and reassurance should be provided.

To conclude, despite the recent availability of ultrasonography, practitioners should be aware of the possibility of Horner syndrome and its delayed presentation, even with the distal approaches to brachial plexus block. Also, the volume of saline used for hydrodissection should be carefully regulated to prevent Horner syndrome resulting from the cephalad migration of LAs.

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DISCLOSURES

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