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CASE REPORT
Hand/Peripheral Nerve

Anatomical Variability Predisposed a Child to Permanent Brachial Plexopathy following Incidental Trauma

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Summary: Anatomic variations in peripheral nerves and the perineural environment are common and can contribute to acute or chronic neuropathy in certain individuals. Awareness of these variants is relevant to understanding both the etiopathogenesis and the increased susceptibility to nerve injury in some patients. We present a 4-year-old boy who sustained a permanent injury to the upper brachial plexus from a relatively minor trauma. Surgical exploration revealed a variation in upper trunk anatomy that likely contributed to this outcome. *(Plast Reconstr Surg Glob Open 2020;8:e2804; doi: 10.1097/GOX.0000000000002804; Published online 24 April 2020.)*

INTRODUCTION

The brachial plexus (BP), formed by the complex confluence of the cervical ventral rami (C5–C8) and the first thoracic nerve (T1), innervates the shoulder girdle and the upper extremity. BP injuries can be debilitating and may require operative treatment.[1,2] Numerous anatomic variations have been described throughout the BP.[3-6] These differences can increase the risk of developing nerve injury along the BP nerves, and may impact its treatment. The following case is illustrative of this concept. The following clinical information, including digital media, is presented after obtaining consent from the patient and his family.

CASE REPORT

An otherwise healthy 4-year-old boy presented to the emergency room with a sudden onset weakness, involving his proximal right upper extremity (RUE) following a fall onto his right arm and shoulder. There was no activity in the bicep or deltoid muscles, and the triceps were discernably weak; finger and wrist movements were normal. The patient denied any notable pain, numbness, or tingling. A radiograph demonstrated no signs of fracture or other abnormalities, and the patient was discharged.

Two months later, he presented to an orthopedic clinic, with no improvement in RUE strength. Physical examination revealed that the patient's proximal RUE was atrophied; the magnetic resonance imaging (MRI) completed 1 month later (3 months postinjury) confirmed diffuse atrophy involving the musculature of the right shoulder girdle, an abnormal signal without clear nerve disruption in the right BP, and an increased T2 signal intensity in the right proximal humeral physis suggestive of epiphyseal edema (Fig. 1). The patient was given a 14-day course of oral steroids for posttraumatic BP inflammation, but weakness of the shoulder girdle, arm elevation, and elbow flexion persisted. An electromyogram, a month later, demonstrated severe RUE brachial plexopathy localized in the upper trunk and lateral cord. He exhibited minimal activation of the right axillary, musculocutaneous, and median nerves in response to the deltoid, biceps, and abductor pollicis brevis muscles, respectively.

He was observed for several months but had persistent inability to flex his elbow or raise his arm past shoulder height. At 10 months postinjury, operative exploration of the BP revealed an injured C5 root that did not join with C6 into a formal upper trunk, but instead tracked distally and independently branched simultaneously into an anterior branch (AC5), a posterior branch (PC5), and the suprascapular (SS) nerve. The C6 root also continued distally and eventually divided into an anterior branch (AC6) and a posterior branch (PC6). Just distal to this branching, AC5 and AC6 combined to form an anterior division (AD), and PC5 coalesced with PC6 to form a posterior division (PD). Thus, a formal upper trunk was never formed (Figs. 2, 3). To prevent unnecessary trauma to the nerve structures, we elected not to follow the 2 divisions

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Received for publication December 24, 2019; accepted March 6, 2020.

DOI: 10.1097/GOX.0000000000002804

Disclosure: *The authors have no financial interest to declare in relation to the content of this article.*

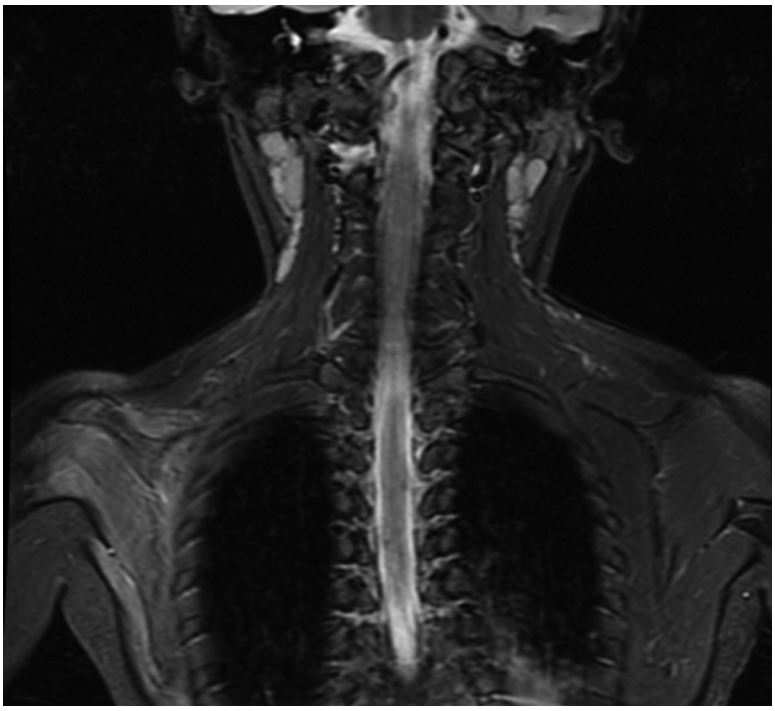

Fig. 1. An MRI completed 3 months postinjury. Imaging revealed defused atrophy involving the musculature of the right shoulder girdle, an abnormal signal of the infraclavicular portion of the right BP, and a right proximal humeral physis T2 hyperintense signal suggestive of epiphyseal edema.

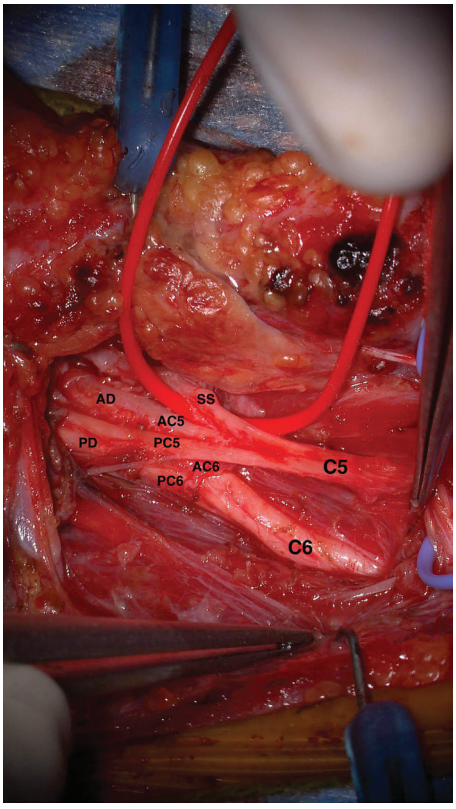

Fig. 2. An intraoperative photograph showing the abnormal brachial plexus anatomy. C5 independently branches into an anterior branch (AC5), the posterior branch (PC5), and the SS nerve. The C6 root also divides into an anterior branch (AC6) and a posterior branch (PC6). Distally, AC5 and AC6 combined to form an anterior division (AD), and PC5 coalesced with PC6 to form a PD.

under the clavicle to discern whether the PD combined with the C7 root.

Fascicular bundles of the spinal accessory nerve were transferred to the SS and PD, and the large superior cervical sensory branch was used as a multifascicular, end-to-end interposition graft from C5 to the AD and PD. Shortly following his operation, the patient's family moved out of state, therefore making follow-up challenging.

DISCUSSION

Anatomic variations in the BP can have clinical significance. These differences may increase the risk of intraoperative injuries (ie, during radical neck dissection),[7] compressive neuropathies,[8,9] obstetric brachial plexopathy, and traumatic injuries. Furthermore, these variations are more commonplace than previously thought, with reports in the literature showing variations as high as 34%,[10] 47.7%,[11] and 53.4% in cadaveric studies.[12] Malformation of the superior trunk, similarly to our patient, has also been reported, with rates ranging from 1% to 1.4% in fetal cadaveric studies.[6,13,14] Interestingly, there have been no previous reports of prolonged functional recovery due to undiagnosed BP injury in patients with anatomic variants. It is our belief that these cases often go undiagnosed and that this case report will raise awareness during the initial work-up of patients with persistent upper extremity weakness after trauma.

Early diagnosis and treatment are paramount for optimizing clinical outcomes of BP injury and preventing irreversible nerve/muscle disability. In situations like this, when diagnosis and treatment are delayed by anatomic

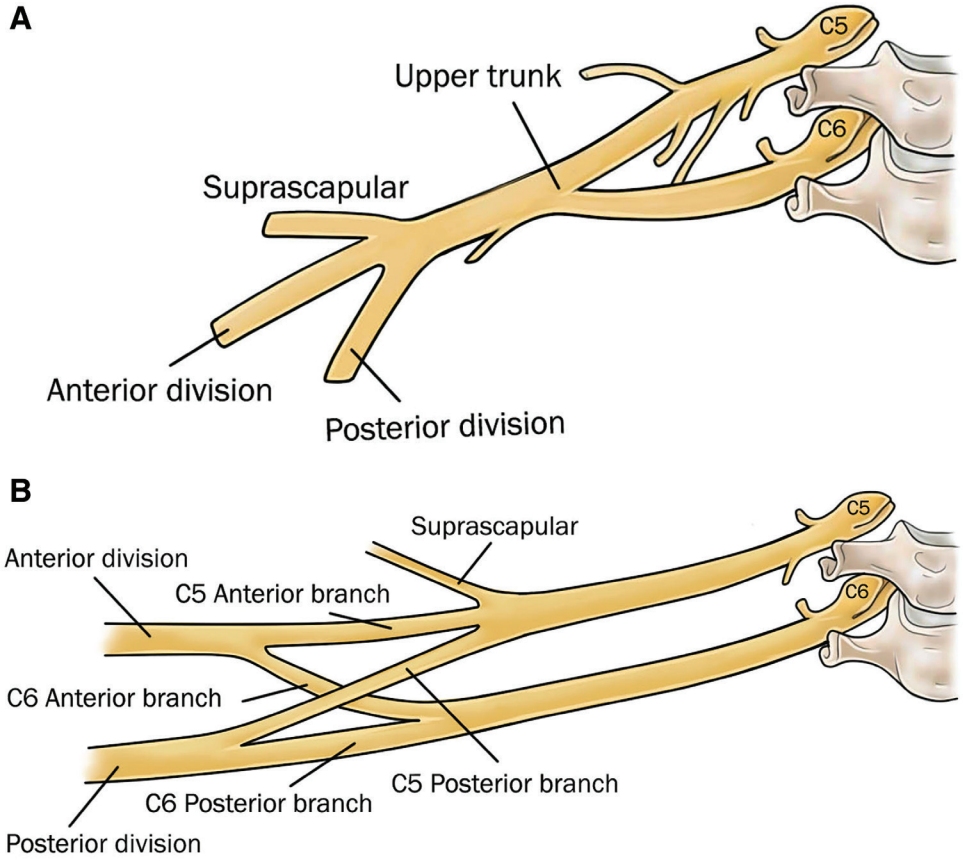

Fig. 3. A diagram illustrating the normal brachial plexus anatomy (A) compared with the abnormal brachial plexus anatomy in our patient (B). Note that in the latter image, C5 and C6 branch independently and do not form an upper trunk.

variations and an abnormal clinical presentation, patients are at higher risk for long-term deficits. Missing and delaying diagnosis in these settings has been shown to correlate with worse outcomes, specifically in elbow flexion and strength as well as shoulder range of motion (ROM).[1,15] This is unfortunate because sensory recovery and functional recovery are better in younger children[16,17] and when surgical intervention occurs early.[2] Our patient had some delay in treatment because his MRI showed no clear nerve root or trunk disruption, a finding confirmed at operative exploration, and MRI findings can be nonspecific in the context of trauma when inflammation can obscure findings of clear pathoanatomy.[18] The absence of a discrete nerve injury on MRI coupled with the relatively benign mechanism of injury prompted us to observe this patient longer than usual for signs of recovery. In retrospect, the unique structure of his BP negatively impacted his chance for such a favorable outcome.

Our patient's severe BP injury resulted from a very minor trauma—jumping onto a pile of young children. Although the MRI raises the possibility of a concomitant proximal humeral physeal injury, the force was insufficient to create a frank osseous or muscular injury. Moreover, the vast majority of traumatic BP stretch injuries resolve with observation. We hypothesize that the severity of our patient's injury is the direct result of his unusual BP anatomy. Typically, the C5 and C6 nerve roots coalesce soon after exiting the neural foramina, thus creating a larger and structurally reinforced upper trunk (UT). The SS nerve, anterior division (AD), and posterior division (PD) arise from this larger, more blended nerve unit. In our patient, the C5 root traveled alone until it approached the clavicle and split into 3 relatively small branches: SS, AC5, and PC5 (Figs. 2, 3). The isolated C5 contribution would, in our opinion, be more prone to stretch injury than the normal situation, and the effect of any damage would more directly affect muscles innervated by the SS and AD because of the lack of observable C6 contributions to these branches.

This case report emphasizes the importance of considering anatomic variability in the context of BP injury. Variations of nerve anatomy, constitution, and the perineural environment (surrounding structures, special orientation, systemic influences, etc) likely account for the observed disparities in the risk of peripheral nerve injury and the prognosis of both nonoperative and operative treatments.

A limitation of our report is the lack of a long-term functional follow-up. The family moved to California

shortly after the initial BP reconstruction, and according to his mother (via phone), the patient underwent an Oberlin procedure within several months. Although it is reported that the patient moves the arm "well," we are unable to objectively measure his function, and any attempt would be confounded by the additional reconstructive procedure. Nevertheless, the focus of this report is to raise awareness of this condition, and similar, rare anatomic variations in the BP and the potential clinical implications consequent to such anomalies.

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