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Case Report

Atypical presentation of forearm compartment syndrome in a child

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ABSTRACT

Background: Compartment syndrome (CS) in children is rare in the paediatric population and diagnosis is challenging. Atypical or delayed management of CS has been known to have poor outcomes. We report an 8-year-old girl with an atypical presentation of CS and her management.
Case report: In this case report, we discuss the case of an 8-year-old girl with an open fracture of the midshaft radius and ulna fracture managed with Titanium elastic nailing system (TENS). She presented 2 weeks post injury with significant pain, flexed fingers and agitated. Following an urgent forearm fasciotomy and carpal tunnel decompression, her symptoms improved. 72 hours later, her fasciotomy wounds were closed and her symptoms continued to improve. 9 weeks post injury her forearm fracture had radiological union. 6 months post injury her TENS nails were removed.8 months post injury, she had no functional motor deficit but residual sensory deficit in median nerve distribution.
Conclusion: CS requires a high index of suspicion in the paediatric population. In addition to cardinal 5P's signs of CS, a triad of 3A's has been proposed for diagnosis of CS in children. Long term outcomes are favourable in atypical presentations or delayed diagnosis cases.

Introduction

Compartment syndrome (CS) is a recognized complication characterized by increased pressure within a closed anatomical space, which can affect any myofascial compartment in the body, including the extremities. Trauma is the most common cause of compartment syndrome in children [1]. CS is primarily a clinical diagnosis, often established based on patient history, examination findings, and in some cases, the measurement of compartment pressures. The diagnostic criteria for CS in children may mirror those in adults, however reaching this diagnosis in the paediatric population can be particularly challenging, and often results in significant delays [1,2] Despite these delays in diagnosis, which naturally lead to a delay between injury and fasciotomy, most paediatric patients surprisingly achieve excellent clinical outcomes [3]. We present a case in which the diagnosis was due to the delayed presentation, yet the clinical outcome was favourable.

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Case report

A 9-year-old girl presented to our hospital in April 2024 after falling from a trampoline onto her left outstretched arm. She was right-hand dominant and had no significant medical history. Upon initial evaluation, she reported severe pain in her left forearm, along with tingling sensations in the index, middle, and ring fingers of her left hand. She was reluctant to move her fingers due to the pain. The initial assessment revealed a visibly deformed forearm, and a puncture wound on the volar aspect of her forearm, measuring 0.5 cm × 1 cm overlying the ulna. X-rays of her left forearm indicated midshaft fractures of both the radius and ulna (Fig. 1). Her arm was immobilised in an above-elbow cast, and the wound was cleaned with normal saline, followed by the application of damp gauze. She also received intravenous antibiotics as part of the management for her open fracture.

Operative intervention

She received general anaesthetic for the procedure and had an arm tourniquet applied. The wound was initially debrided and thoroughly washed with 0.9 % saline. The wound was subsequently closed at the end of the procedure. The fracture was reduced using closed reduction manoeuvres and titanium elastic nail system was used for stabilisation. Under an image intensifier (II), the entry point along the lateral aspect of the distal radius was identified. A small stab incision was made over the entry point and care was taken to protect the superficial radial nerve. The distal radius entry point was opened using a sharp awl and a 2 mm titanium elastic nail was inserted under II guidance. Next, the entry point for the ulna was identified over the proximal ulna 1 cm distal to the apophysis. The ulna cortex was opened using a sharp awl and 2 mm titanium elastic nail was inserted under II guidance. Final II images showed a satisfactory reduction of the fracture with all implants in situ (Fig. 2). The procedure lasted for 59 min with an arm tourniquet pressure of 180 mmHg. An above elbow backslab was applied for comfort and pain relief.

In the post-operative period, she was comfortable with minimal pain, and able to flex her thumb and index finger. The altered sensation over her thumb and index finger remained the same as before the surgery. The rest of the neurological and vascular examination was normal. She was discharged the next day with a follow up appointment at the fracture clinic. Two days after the operation, she returned to the emergency department complaining of pain in her left forearm. She was seen by a physician who provided oral analgesia and closely monitored her. No cardinal signs of compartment syndrome were observed and her neurovascular examination showed no change from her previous visit. Her pain improved with oral analgesia, and she was discharged home with follow-up instructions. She missed the 1-week clinic appointment due to a family vacation but attended the 2-week follow-up (15 days from injury) appointment. During this visit she reported ongoing significant pain in her left forearm which had worsened in the last few days, along with the tingling in her thumb, index, and middle fingers. The backslab was removed and examination the forearm did not feel tense, persistent pain, painful passive stretch of the fingers, limited supination and fingers resting in a flexed position. She was very anxious and agitated during the examination, displaying signs of atypical compartment syndrome. X-rays taken at this visit showed no secondary displacement or further fractures (Fig. 3).

Following a team discussion, she underwent a left forearm single incision fasciotomy with carpal tunnel decompression. All the 3 compartments were decompressed, and the neurovascular structures were visualised and protected. Intraoperatively, the flexor carpi radialis, palmaris longus, flexor pollicis longus and pronator quadratus were noted to be dusky and all the muscles were observed to bulge immediately following decompression of the compartments. A small amount of haematoma was also found in the deep volar compartment. The hematoma was washed with saline and all the muscles were examined and noted to be responding to electrical stimulation. At the end of the surgery, the wound was packed with saline soaked gauze and an above-elbow back slab was applied. In the immediate post-operative period, she reported significant improvement in pain and her analgesia requirements were significantly reduced. She was closely monitored over the next 2–3 days and 72 h later she was taken back to the theatre for wound debridement and

Fig. 1. Initial X-ray of left forearm.

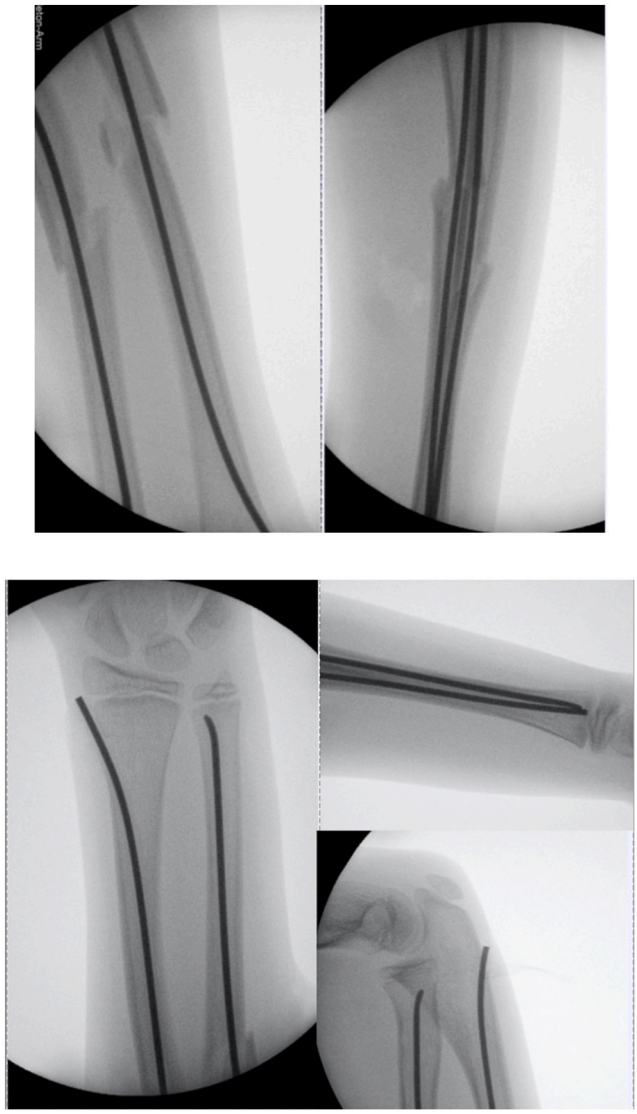

Fig. 2. Intraoperative images for left forearm TENS nailing.

Fig. 3. X-rays taken at 2 week appointment prior to forearm decompression.

closure. The muscles had regained their colour and were responsive to electrical stimulation and the wounds were clean. Following discharge, she was reviewed weekly and over a period of 4 weeks her motor function of the median nerve was back to normal. She continued to experience altered sensation in her index and middle finger. She was subsequently followed up every 4 weeks until the fracture united. Signs of fracture union were seen on x-rays 9 weeks post injury (Fig. 4).

9 months post injury, she continued to experience a burning sensation on the proximal aspect of the fasciotomy wound and altered sensation in the index and middle finger. She had no functional motor deficit, however her grip strength was lower on the left side (11kgs) compared to the right side (15kgs). The fracture had united well without any signs of any residual deformity as demonstrated in Fig. 5. She underwent prolonged physiotherapy and desensitisation therapy. 12 months post injury, her sensation in the index and middle finger was restored, the burning sensation over the wound had significantly reduced and her grip strength had been restored equally to the contralateral side.

Discussion

Compartment syndrome is an orthopaedic emergency that requires urgent surgical decompression to prevent long term morbidity [4]. Children with compartment syndrome can present in various ways, which often makes diagnosis more challenging. Although it is typically considered a condition that presents acutely, it can also manifest subacutely or with a delayed onset, particularly in children who often present later with compartment syndrome compared to adults, leading to a delay in diagnosis. Various reasons have been suggested for this delayed presentation and diagnosis. Children might present with more subtle symptoms such as irritability, reluctance to use the affected limb, or behavioural changes, particularly in younger or non-verbal children. Those who are very young or have developmental delays may have difficulty articulating pain or other symptoms associated with compartment syndrome. Often these symptoms are seen non-specific and are usually attributed to the initial injury and general post-injury discomfort rather than compartment syndrome.

The traditional hallmarks of compartment syndrome for both adults and children include disproportionate pain, pulselessness, pallor, paraesthesia and paralysis; through a paediatric lens, a new triad of 3A's has been proposed: increased anxiety, increased demand for analgesics and agitated behaviour to establish the diagnosis of CS [2]. Furthermore, these behaviours were observed to frequently foreshadow the development of more clinically indicative signs of compartment syndrome [4]. Whilst pain is often the primary symptom in the majority of cases, there are reports of paediatric ‘silent’ compartment syndrome indicating this condition may present without marked pain [5,6].

Delayed presentation may occur hours to days after the initial injury, often resulting in a more challenging diagnosis. This aetiology in children is often linked to less obvious trauma or subtle vascular compromise, and it can be exacerbated by delayed management of fractures or casting that is too tight. It has been noted that children may have more pliable fascia, potentially leading to a slower progression of symptoms and thus a delayed diagnosis [7]. The differences in tissue compliance of children, are that they are more elastic compared to adults [8]. This means that the compartment can expand more, possibly delaying the onset of symptoms and signs until the condition has become more severe.

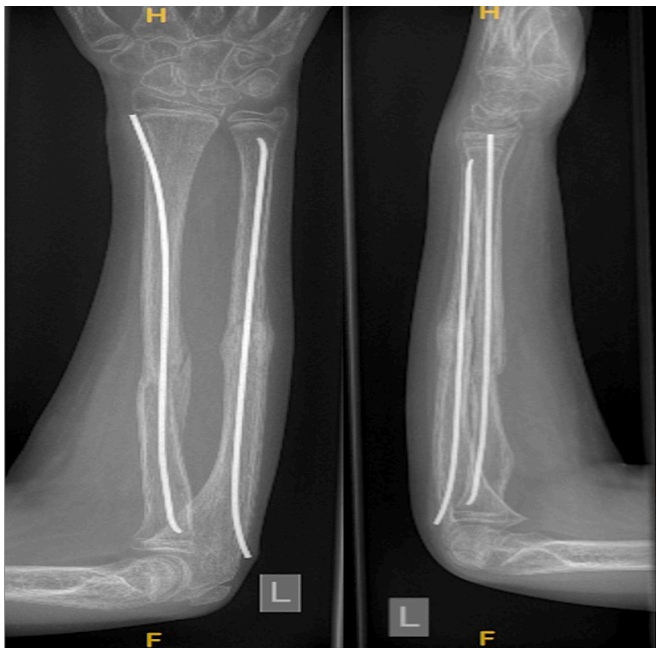

Fig. 4. X-rays taken 2 months post injury showing callous at fracture site.

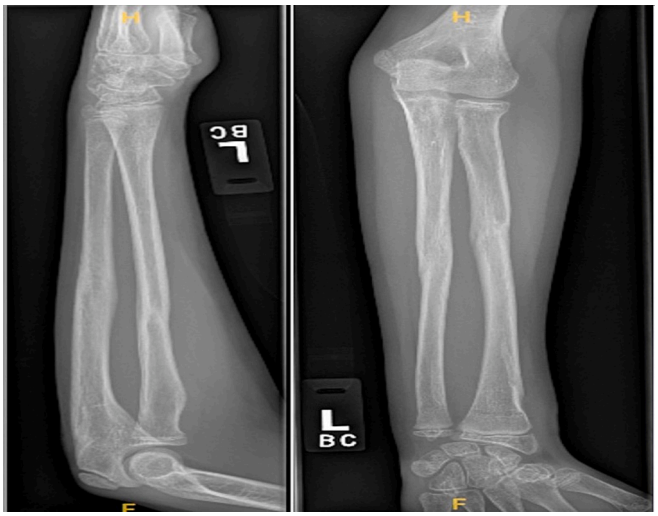

Fig. 5. X-rays taken 8 months post injury and removal of TENS nail show fracture union and no residual deformity.

Compartment syndrome is a clinical diagnosis but there are a few adjuncts for identifying CS such as needle manometry (NM) and near infrared spectroscopy (NCS). NRS has been utilised to detect muscle ischemia in both adults and children, but the lack of standardisation in threshold values makes this tool impractical [9]. NM is still a widely practiced method to measure intra-compartmental pressures. In the paediatric population, an absolute compartment pressure of 30-45 mmHg or less than 30 mmHg from mean arterial pressure is considered to be diagnostic of compartment syndrome [8,10,11]. In paediatric forearm fractures, no long term complications have been noted under the threshold of 40 mmHg [12].

In the paediatric population, despite delays in diagnosis and treatment, good outcomes have been reported [6,8]. It has been noted that forearm compartment syndrome typically has a longer delay in diagnosis, but such a delay has not been associated with a worsening outcome [3]. 74 % of patients who underwent decompressive fasciotomy had a long-term outcome rated as 'excellent' and an additional 22 % rated as 'fair' on the premise of motor function restoration, lack of stiffness and sensation range [3]. Therefore, it is reasonable to consider decompression/fasciotomy despite delay in diagnosis [3,13,14]. Our case report above supports a good functional outcome following a decompression despite delays in presentation and diagnosis.

The subtlety and variability of these presentations in children require clinicians to maintain a high level of suspicion, especially in cases of trauma or after surgical interventions, to ensure timely diagnosis and treatment. Lack of awareness can be a contributing factor as there is a lower index for suspicion of compartment syndrome in children among acute healthcare providers. This is partly due to its rarity in children, resulting in less experience with the condition and potentially leading to delayed recognition, treatment and management.

Conclusion

Current literature supports the variability of clinical presentations, making diagnosis challenging. In addition to the cardinal signs of compartment syndrome, a triad of 3A's should raise a high index of suspicion for compartment syndrome. Needle nanometry is a useful diagnostic adjunct and an absolute compartment pressure of 40 mmHg in forearm fractures should prompt consideration for decompression. Despite delays in diagnosis, forearm compartment syndrome in the paediatric population leads to good functional outcomes.

List of abbreviations

CS	Compartment syndrome
II	Image intensifier
NM	needle manometry
NCS	Near infrared spectroscopy

CRediT authorship contribution statement

P.A. Saxena: Conceptualization, Data curation, Formal analysis, Validation, Writing – original draft, Writing – review & editing. **M. Veylamuthen:** Conceptualization, Writing – original draft. **L. Kitsi:** Writing – original draft. **A. Crawford:** Writing – original draft. **A.P. Dekker:** Formal analysis, Writing – review & editing, Supervision. **N. Ashwood:** Conceptualization, Formal analysis, Supervision, Writing – review & editing.

Declaration of competing interest

The authors declare that they have no competing interests.

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