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CASE REPORT



## Recurrent Metachronous Acute Compartment Syndrome in the Legs of a Child

Récidive du syndrome des loges aigu métachrones dans les jambes d'un enfant

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#### ABSTRACT

BACKGROUND: Compartment syndromes in children are rare, rarer still is the occurrence of metachronous compartment syndrome.

**OBJECTIVE:** To report an unusual and rare presentation of metachronous acute compartment syndrome.

METHODS: A seven-year-old boy presented with four separate episodes of acute compartment syndrome of the legs within three years. The recurrent nature of the illness prompted detail review of the records of the child.

RESULTS: There was involvement of each leg twice between the ages of three and seven years following trivial trauma. At first presentation of each leg, fasciotomy was carried out in one or two compartments. There was no recurrence after fasciotomy of the four compartments during the second presentation of each leg.

CONCLUSIONS: This case shows that a child could develop metachronous compartment syndrome in the legs following trivial injury and high index of suspicion is needed to recognize it. It also illustrates the importance of carrying out fasciotomy of all the four compartments to avoid recurrence of the pathology. WAJM 2010; 29(3): 190–192.

Keywords: Recurrent, Metachronous, Acute compartment syndrome, trivial injury.

### RÉSUMÉ

**CONTEXTE:** Compartiment syndromes chez les enfants sont rares, plus rares encore l'apparition du syndrome des loges métachrones.

**OBJECTIF:** Rapporter une présentation inhabituelle et rare du syndrome des loges aigu métachrones.

**MÉTHODES:** Un garçon de sept ans s'est présenté avec quatre épisodes de syndrome des loges aigu des jambes dans les trois ans. Le caractère récurrent de la maladie invité Détails de l'évaluation des dossiers de l'enfant.

**RÉSULTATS:** Il y avait la participation de chaque jambe deux fois entre les âges de trois et sept ans après un traumatisme trivial. À la première présentation de chaque jambe, aponévrotomie a été réalisée en un ou deux compartiments. Il n'y avait pas de récidive après aponévrotomie des quatre compartiments au cours de la deuxième présentation de chaque jambe.

**CONCLUSIONS:** Ce cas montre que l'enfant pourrait développer le syndrome des loges métachrones dans les jambes après une blessure trivial et de l'indice élevé de suspicion est nécessaire de le reconnaître. Il illustre également l'importance de mener à bien aponévrotomie de tous les quatre compartiments pour éviter la récurrence de la pathologie. WAJM 2010; 29 (3): 190–192.

*Mots-clés: Récidive, métachrones, syndrome des loges aigu, des blessures Trivial.* 

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#### **INTRODUCTION**

Compartment syndrome in children is rare but should be considered in a child presenting with persistent pain in a limb even when there is a history of trivial history. Rarer still is presentation of the syndrome that is bilateral and metachronous.<sup>1–3</sup> The diagnosis could be misleading and thus a cause of misdiagnosis. These informed our presentation of this case.

#### **Case report**

A seven-year-old boy presented with four separate episodes of acute compartment syndrome of the legs metachronously within a period of three years. He presented at the emergency room of our hospital and was subsequently referred to surgical or orthopaedic unit on-call. There was involvement of different legs at four different times, one leg after the other and each leg twice between the ages of three and seven years. History of trauma could not be acknowledged in one presentation but there was history of trivial injury between one to two days before presenting in the Emergency room in the other three occasions. There was tachycardia at each presentation but other vital signs were essentially normal. Past medical history was not significant to the development of the presentations and the child had normal developmental milestones. The white blood count, the haemoglobin level and the erythrocyte sedimentation rate on each of the four different presentations were normal. The recurrent nature of this pathology and the metachronous involvement of the legs stimulated the detail review of the patient's medical record and extraction of the information on the previous presentations and managements in this seven year old boy. The last presentation was three and a half years ago. The metachronous presentations of this pathology following trivial injuries, the clinical presentations, investigation, definitive treatment, surgical findings and the outcome of this uncommon presentation are summarised in Table 1.

Fasciotomy wounds were closed by secondary intention and recovery of the limbs was uneventful in all the presentations with normal gait.

# Table 1: Presentation, Management and the Outcome of Recurrent Metachronous Acute Compartment Syndrome in the Legs of a Child

Ago post	3	4	(	7
Presentation (Yrs. and Date	.) 04/04/03	24/03/04	07/03/06	21/04/06
Complaints	Two days history of painful R leg swelling	A day history limping gait and swelling of the L leg.	A day history of pain in the R leg.	A day history of sudden sharp pain in the L calf
History of Tauma	Fell while playing at home	Injury to the L leg while playing football	Fall in the bathroom	The mother and the child could not remember any injury
Clinical Findings	Nrmal vital signs. Tense and Right calt tenderness	Normal vital signs. Tense and swelling of the upper half calf of the L leg. Tenderness on dorsiflexion	Limping gait. Tense and tender R calf	L calf swelling which was very tense and tender on plantar and dorsiflexion
Investigations	X-ray of the R leg show soft tissue swelling USS-solid lesion of 5.5x3x2.5cm in the soft tissue of the calf CBC-Normal	x-ray showed soft tissue swelling Colour Doppler showed no abnormality in the blood vessels CBC-Normal	X-ray showed soft tissue swelling USS revealed haematoma between calf muscle plane $82 \times 56$ mm $\Delta P$ in DPICP = 20m mHg CBC-Normal	X-ray showed soft tissue swelling PT=15.4s (C=12.7s) PTT=30.6s (C=31s) INR grossly normal CBC-Normal
Treatment	Fasciotomy of the two posterior compartments	Fasciotomy of the anterior compartment	Fasciotomy of all the compart- ments	Fasciotomy of the compart- ments of the leg
Surgical Findings	4cc of clotted blood evacuated in the calf muscle plane	Not documented	Tense muscles in the anterior and deep posterior compartments	Posterior group of muscles under severe tension

**Key**: ΔP, Differential Pressure between the Diastolic and Iintracompartment Pressure; DPICP, Deep posterior intracompartment pressure; CBC, Cell Blood Count; PT, Prothrombin Time; PTT, Partial Thromboplastin Time; INR, International Normalized Ratio.

#### DISCUSSION

The metachronous and recurrent nature of the presentation calls for concern and the possible causes of the syndrome in this growing child. It is worth mentioning that English literature review did not reveal this type of recurrent acute compartmental syndrome in the lower limbs. The closest to this condition found in the English literature review was bilateral lower extremity compartment syndromes secondary to intraosseous fluid resuscitation reported by Galpin *et al* who reported severe compartment syndromes of both lower extremities complicating the use of intraosseous fluid resuscitation.<sup>1</sup> Also acute bilateral forearm compartment syndrome of unknown aetiology was reported by O'leary *et al.*<sup>2</sup> It should be observed that both reported cases were synchronous not metachronous presentations as seen in this case. It is even more unique because of the bilateral nature and presentation after trivial trauma. It was noted that presentation at the Emergency Room was after one or two days after trivial injury and this long duration of hours before presentation could be explained by the fact that the injuries were

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caused by low energy. The aetiology of this unusual presentation could only be associated with trivial trauma of the soft tissue because the X-ray of legs revealed soft tissue swelling and no fracture. The Prothrombin Time (PT); Partial Thromboplastin Time (PTT) and International Normalized Ratio (INR) were done only in one of the presentations but were normal thereby ruling out a bleeding disorder. It is thus very important to consider the diagnosis of compartment syndrome in a child who is presenting with persistent pain to the leg even when the child has sustained trivial injury. The causes of compartment syndrome in children varies widely from the fractures of the long bones<sup>3</sup> to iatrogenic causes like the application of skin traction in three-year-old child who developed compartment syndrome in the nonfractured limb after application of the skin traction.<sup>4</sup> To this list trivial soft tissue injury should be included.

The diagnosis could be very challenging and could be misdiagnosed as soft tissue injury; thus a very high index of suspicion is required.

classic symptoms The of compartment syndrome include the "8 P's": Pain at rest, Pain with passive stretch; Pressure; Parasthesia; Paresis; Pulseslessness; Pallor and Poikilothermia. Although these are considered the classic signs of the condition, they are not always present in all cases. Pain (at rest and at passive stretch) and parasthesias are the most common of these symptoms and should alter the physician of the pathology. When paralysis, pulseslesness, palor and poikilothermia are noted the diagnosis is obviously late. Other important findings on physical examination may include increased size of the affected extremity (compared to the unaffected side), or increased firmness, shinny and presence of blisters in the absence of thermal injury<sup>5</sup> on the affected limb these should raise suspicion of compartment syndrome. In this patient the legs were tense and patient was in pain either at rest or passive stretching. Bae et al reported that pain, pallor, paresthesia, paralysis, and pulselessness were relatively unreliable signs and symptoms of compartment syndrome in children.<sup>6</sup>

They also mentioned that an increasing analgesia requirement in a child in combination with other clinical signs, however, was a more sensitive indicator of compartment syndrome.<sup>6</sup> Invasive intracompartment pressure measurement might not be quite useful in the children as could be in the adults, this make it very important to make clinical diagnosis of this uncommon pathology in the children. A simple hand-held device to measure tissue hardness as a method of diagnosing compartment syndrome noninvasively in children is being developed.<sup>7</sup> This will aid the diagnosis of this pathology where the clinical symptoms and signs are not very classical. Other investigations that could help in making the diagnosis where they are available include Magnetic resonance imaging scan, Near infrared spectroscopy which are non-invasive and the minimally invasive Scintigraphy.

The treatment of compartment syndrome is mainly surgical and this patient had three fasciotomies. It is recommended that all the four compartments should be decompressed to avoid hyperemia precipitating increase in pressure in the adjacent compartment or missing established acute compartment syndrome in adjacent compartments.<sup>8, 9</sup> It was noted that not all the compartments were decompressed during the first two presentations and that must have accounted for recurrence in these legs. In the last two presentations all the compartments were decompressed and the child has not presented with compartment syndrome in the past three years of follow up. This shows the importance of decompressing all the compartments to avoid recurrence of the pathology in intact compartment. Compartment syndrome could be limb and life threatening and it could be difficult to diagnose in a child, delay in diagnosis leads to disastrous outcomes. The inability to make a prompt and accurate diagnosis can lead to significant extremity morbidity and possibly mortality.10

Our patient has been followed up for three years since the last presentation and there has been no complication of the pathology in any of the legs. These complications could be permanent nerve damage, Volkmann ischaemic contracture, infection, loss of limb, death,cosmetic deformity from fasciotomy.

#### **Conclusion:**

This case shows that a child could develop metachronous compartment syndrome in the legs following trivial injuries and that high index of suspicion is needed to recognize and manage this pathology to avoid limb and life threatening complications associated with the syndrome. It also illustrates the importance of carrying out fasciotomy of all the four compartments to avoid recurrence of the pathology.

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