Bilateral thigh and calf compartment syndromes following elective surgery: Case report and review of the literature

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Bilateral thigh and calf compartment syndromes are rare in pediatric patients, especially following elective foot surgery. Such a case in a child with attention deficit hyperactivity disorder is described in the present report. This child was taking multiple medications, including Ritalin (methylphenidate hydrochloride), Paxil (paroxetine hydrochloride) and clonidine. Other factors that potentially contribute to the multiple compartment syndromes include pneumatic tourniquet use and caudal block augmentation of general anesthesia. Behaviour associated with attention deficit hyperactivity disorder complicated the detection of the patient's compartment syndromes. The development of the compartment syndromes was delayed more than 48 h on one side. The potential for this patient's pharmacotherapy to have contributed to the development of the compartment syndromes is discussed.

Key Words: *Attention deficit hyperactivity disorder; Compartment syndrome; Methylphenidate*

Syndromes du compartiment bilatéraux à la cuisse et au mollet après chirurgie élective : Rapport de cas et revue de la littérature

RÉSUMÉ : Les syndromes du compartiment bilatéraux affectant la cuisse et le mollet sont rares chez les patients pédiatriques, surtout après une chirurgie élective pour le pied. On décrit ici le cas d'un enfant atteint d'un trouble d'hyperactivité avec déficit de l'attention. Cet enfant prenait plusieurs médicaments dont Ritalin (chlorhydrate de méthylphénidate), Paxil (chlorhydrate de paroxétine) et clonidine. D'autres facteurs qui contribuent peut-être au syndrome du compartiment multiple sont l'utilisation d'un brassard pneumatique et d'une épidurale avec l'anesthésie générale. Les comportements associés aux troubles d'hyperactivité avec déficit de l'attention ont compliqué le dépistage des syndromes du compartiment chez le patient. Le développement des syndromes du compartiment a été retardé de plus de 48 heures d'un côté. On aborde ici la possibilité que la pharmacothérapie du patient ait contribué à l'installation des syndromes du compartiment.

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Compartment syndrome is a well-described complication of extremity injuries. It has been defined as increased tissue pressure in a closed fascial space that compromises the circulation to muscles and nerves (1). Compartment syndrome of the thigh is very rare and particularly uncommon following elective surgery, particularly when the surgical site is remote from the thigh. Its occurrence in children is even more uncommon. A review of the English literature revealed only nine previous cases in patients younger than 18 years of age (2-10).

The reported etiologies of compartment syndrome in the thigh include severe exercise- and sports-related injuries (11), femoral and intertrochanteric fractures (12), intramedullary nailing (13), the use of pneumatic antishock trousers (14,15), prolonged tourniquet application (9,16), gunshot wounds (17), epidural anesthesia (7), false aneurysms and bleeding (4,17), femoral artery cannulation (18), and total hip and knee replacements (19,20).

The review of the literature revealed no previously reported association between compartment syndrome and medications used for the management of attention deficit hyperactivity disorder (ADHD).

The following case presentation reports a child taking methylphenidate hydrochloride (Ritalin, Novartis Pharmaceuticals, Canada), clonidine hydrochloride and paroxetine hydrochloride (Paxil, GlaxoSmithKline, Canada) for the treatment of ADHD, who developed bilateral thigh and calf compartment syndromes after elective surgery.

CASE PRESENTATION

A seven-year-old boy was transferred to a regional pediatric hospital for treatment of bilateral lower extremity compartment syndromes with postoperative myoglobinuria, elevated creatine phosphokinase, and left thigh and leg swelling and tenderness. Four days before the transfer, the patient underwent surgery for the correction of bilateral skew feet at a local hospital, under general anesthesia and with caudal augmentation. A pneumatic tourniquet was used. Sequential corrective foot surgery was completed under pneumatic tourniquet control. The duration of tourniquet use was 120 min for the left side and 129 min for the right side, at a controlled pressure of 250 mmHg. The surgical site was limited to the foot.

Approximately 10 h after surgery, the patient was agitated and complained of severe right leg pain; unfortunately, these symptoms were initially attributed to his hyperactivity. A clinical examination 36 h later revealed a swollen, hard and tender right thigh and leg. At that time, the left thigh and leg were normal and the casts were not tight. A diagnosis of compartment syndrome was made and an immediate fasciotomy of the right thigh and leg was performed. Forty hours after right-sided fasciotomies were performed, the same symptoms of pain, agitation and screaming resumed. A suspected, delayed left-sided compartment syndrome initiated the patient's transfer to our institution. Upon admission, the left thigh and leg were tense and swollen. The patient was unable to move the toes of his left foot. There was good capillary refill and pulses were present. However, sensory examination was unreliable because of the child's behaviour associated with ADHD. Therefore, the intracompartmental pressures were measured using a Stryker (Stryker Corporation, USA) pressure testing system. The anterior compartment of the thigh measured 43 mmHg, and the anterior and anterolateral compartments of the leg measured 53 mmHg and 48 mmHg, respectively. The patient's systemic diastolic pressure was 70 mmHg. The patient was taken to the operating room for immediate fasciotomies. The anterior and posterior compartments of the thigh were decompressed through a lateral incision. The leg was also decompressed through medial and lateral incisions. Muscles of the anterior and posterior compartments of the thigh, and anterior and lateral compartments of the leg were pale and did not contract when stimulated with electrocautery. No muscle was excised during the first procedure. All wounds were dressed openly. Clinically, his pain was markedly reduced; however, a peroneal nerve deficit persisted. When the swelling had significantly decreased three days later, the patient returned to the operating room where necrotic muscles, namely the tibialis anterior, extensor hallucis longus and the lateral head of gastrocnemius were partially debrided from the left leg, and the wounds were closed. The patient had an uneventful postoperative course following delayed primary closure.

The patient underwent extensive physiotherapy postoperatively to mobilize his ankle and desensitize his leg and foot. Initially, a drop-foot orthotic was utilized. By four months, there was evidence of partial recovery of the peroneal nerve function, but his right foot dorsiflexor was weak (MRC grade 4/5 power). He could walk without the aid of any orthotics. His ADHD medical management was reviewed thoroughly by the pediatric staff and his Ritalin, clonidine and Paxil use was discontinued. His ADHD is managed satisfactorily with behavioural therapy.

DISCUSSION

Bilateral thigh compartment syndrome is uncommon and has been reported primarily in adults following exercise (21-24). We found no previously reported cases of bilateral thigh and leg compartment syndromes in children. In addition, the compartment syndrome on the left side developed almost 48 h postoperatively, and this delay raised the possibility of a systemic contribution to the development of the patient's compartment syndromes.

Although causes of thigh compartment syndrome are varied, we present this case to illustrate two points: It is difficult to establish the diagnosis due to the presence of behaviours associated with ADHD; and there is potential for medications taken for ADHD to contribute to the development of thigh compartment syndrome. The hyperactivity of the child contributed to the delayed diagnosis of the compartment syndrome on the right side. Other factors known to contribute to the development of compartment syndrome, such as tourniquet use, may have contributed, but are unlikely to have been the primary problem.

Ritalin is a piperidine-derivative stimulant. The pharmacological actions of Ritalin include central nervous system and respiratory stimulation and weak sympathomimetic activity. The safety, efficacy and long term effects of Ritalin in children younger than six years of age have not been established (25,26). The described adverse vascular effects of Ritalin include, but are not limited to, cerebral arteritis and/or occlusion, increase or decrease in blood pressure and heart rate, cardiac arrhythmia, angina pectoris, peripheral vasodilation, and erythema multiform with necrotizing vasculitis (25,26). Ritalin has been shown to inhibit liver microsomal enzymes (27); therefore, the effect of any drug undergoing elimination or metabolism in the liver may be prolonged significantly. Approximately 50% of the absorbed dose of clonidine is metabolized in the liver (26).

Clonidine, an imidazoline derivative, is a central alpha-adrenergic stimulant that inhibits sympathetic cardioaccelerator and vasoconstrictor centres, which ultimately results in decreased peripheral vascular resistance. As with Ritalin, the safety, efficacy and long term effects of clonidine are not established in children (25,26). We propose that the adverse effects of Ritalin and clonidine on the peripheral vascular system may have contributed to the development of the compartment syndrome – either by enhancing the effect of the tourniquet or by their synergistic effects on the vascular system.

A review of the literature by Rire et al (28) revealed limited data on the mechanism of action of methylphenidate and its interaction with other drugs, and in their case report, they raised the possibility of interaction between methylphenidate and anesthetic agents. Malony and Schwam (29) reported three cases of sudden death in children taking a combination of methylphenidate and clonidine. One other case of a severe compartment syndrome following a fractured tibia in a pediatric patient taking Ritalin was identified (personal communication, RA Ogilvie). We think that it is prudent to consider the possibility that the prolonged use (2.5 years) of these two medications may have resulted in permanent or temporary peripheral vascular changes that became evident by the stress of surgery and/or the application of the tourniquet, and may have contributed to the development of the compartment syndrome.

The tourniquet is unlikely to be the primary cause of the development of these multiple bilateral extensive compartment syndromes because excessive tourniquet pressure or duration causes more damage to the muscles lying beneath it than those distal to it (30,31), whereas, in our case, distal muscles of both legs were more damaged than were proximal muscles. Bilateral thigh and leg compartment syndromes are rare in children, and the multiple sites of involvement suggest a systematic contribution to its development. Also, elective orthopedic procedures are common in children and postoperative compartment syndromes are rare. The surgical technique and the tourniquet time, pressure and type in this case were similar to those that were used for other children with similar deformities.

We believe that the use of methylphenidate alone or in combination with other drugs may result in an increased risk of compartment syndrome in children with traumatic injury or surgery under tourniquet control. We recommend a higher level of suspicion and suggest that further research may deliniate the pathophysiological mechanism responsible for this observation.

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