Bowel obstruction from intramural hematoma in two children treated with low molecular weight heparin: Case report and review of the literature

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Abstract
Low molecular weight heparin (LMWH) is frequently employed in children to prevent extension of intravascular thrombosis. However, this therapy can result in pathologic bleeding, including spontaneous intramural hematoma of the intestinal wall. In this report, we describe two cases of intestinal obstruction resulting from intramural hematomas during therapeutic LMWH therapy in children. The diagnostic studies and management of spontaneous intramural hematoma in children are discussed.

Low molecular weight heparin (LMWH) is an anticoagulant often preferred for the prophylaxis and treatment of deep venous thrombosis (DVT) because of its predictable pharmokinetics and comparatively low rates of complications as compared to unfractionated heparin or warfarin [1]. Intramural hematoma is a rare complication of anticoagulation therapy that may result in small intestinal obstruction [2]. To date, this complication has been reported for adult patients during therapy with warfarin [3–7] or unfractionated heparin [8]. However, only one prior case report has described this complication of anticoagulant therapy in children [1]. In this report we describe two pediatric patients who developed intramural hematoma associated with bowel obstruction concurrent with LMWH therapy.

1. Case reports
1.1. Case 1

Patient One was a nine month-old boy with a history of infantile spasms (West Syndrome). His medical history included multiple hospitalizations and difficult peripheral venous access, necessitating placement of a right femoral central venous catheter. A subsequent ultrasound of the right leg for unilateral swelling identified a deep venous thrombosis (DVT) of the femoral vein. The patient was treated with LMWH, with a planned course of three months. A hypercoagulability workup conducted at that time revealed no abnormalities in serum prothrombin, Factor V, or homocysteine levels.

The patient was subsequently admitted to the hospital for elective placement of a gastrostomy tube. LMWH was held for over 12 h prior to his operation. His immediate post-operative course was uncomplicated, and LMWH therapy was restarted on post-operative day 5. On post-operative day 14, he developed bilious emesis and distension. He was found to be markedly anemic with a hemoglobin level of 4.5 g/dl, requiring blood transfusion. His obstructive symptoms initially resolved after 2 days of nasogastric...
decompression, but recurred 6 days later, with copious bilious nasogastric output and abdominal distension. Computed tomography (CT) of the abdomen and pelvis demonstrated dilated proximal small intestine, with a transition point in the small intestine. Apparently intramural densities projecting into the lumen of the small bowel were also noted (Fig. 1). He was brought emergently to the operating room for exploration.

Initial laparoscopic inspection of the abdomen demonstrated massively dilated loops of bowel with multiple adhesions. A laparotomy was performed, and further exploration revealed a 10 cm distended loop of devitalized and perforated jejunum at the site of a large subserosal hematoma (Fig. 2). This segment was resected. In the more distal jejunum, a smaller intramural hematoma was found in a viable, non-obstructed segment of small bowel. This hematoma was drained with repair of the serosal defect. The bowel appeared viable and was thus not resected.

Histologic examination of the resected segment demonstrated areas of necrosis and an intraluminal clot with acute inflammation, as well as intramural hematoma (Fig. 3).

The patient recovered uneventfully after the procedure and was discharged to a rehabilitation facility after his feeding regimen had been established.

1.2. Case 2

Patient Two was a 4 year-old boy with a complicated medical and surgical history, including a seizure disorder, failure to thrive, epileptic focus resection, and fundoplication. He was admitted to the hospital for increasing frequency of his seizures and abdominal distention. He was noted to have a thrombus in the left iliac vein associated with a femoral central line. LMWH therapy was initiated.

Five days after LMWH therapy was begun, he developed retching and was found to have a palpable mass in the left lower quadrant. He was taken to the operating room, and was determined to have a closed loop intestinal obstruction caused by intramural hemorrhage with an associated volvulus. The affected segment was resected. Pathologic evaluation of the resected intestine demonstrated intramural hematoma with ischemia.

The patient recovered uneventfully and was discharged after resuming gastrostomy tube feedings to a rehabilitation facility. He subsequently succumbed to unrelated respiratory complications eight months later.

2. Discussion

Spontaneous intramural hematoma as a result of systemic anticoagulation has been described in the adult literature [2]. The most common cause of this pathology is treatment with vitamin K antagonist therapy (warfarin, phenprocoumon, and acenocoumarol) [9]. Unfractionated heparin has also been associated with intramural hematoma [8]. The incidence of intramural hematoma as a result of anticoagulation in adults treated with anticoagulants has been estimated at 1 in 2500, although due to the infrequent
Clinical presentation of this condition, reports are largely anecdotal [2]. The common manifestations of clinically apparent intramural hematoma are generally those of intestinal obstruction, and include abdominal distention, nausea, vomiting, and pain [8].

There are few prior reports of spontaneous intramural hematoma in anticoagulated children. Intramural hematomas have been observed in children with genetic disorders of coagulation, such as hemophilia [10] and von Willebrand disease [11]. In contrast, a solitary prior case report describes intestinal hematoma associated with LMWH therapy in a child [1]. Our report presents two cases of intramural hematoma, both in children who also had seizure disorders treated with anticonvulsants and a ketogenic diet. It is possible that the relative infrequency of anticoagulation in children compared to adults contributes to the clinical rarity of this phenomenon. Nonetheless, because of the potentially devastating consequences, intramural hematoma should be considered in the differential diagnosis of children with intestinal obstruction in the setting of altered coagulation.

Diagnostic imaging can be helpful in the diagnosis of intramural hematoma. Many authors suggest a noncontrast CT scan be performed in adult patients suspected of this condition, although no definitive studies or guidelines have been published [12]. Ultrasound has also been shown to be useful in the recognition of small bowel hematoma in adults [13]. Ultrasound may be a preferable initial modality for the management of children, as it does not entail ionizing radiation and may be highly informative.

The management of intramural hematoma in adults generally includes non-operative therapy consisting of intravenous fluids and nasogastric suction, combined with treatments to normalize clotting parameters. Surgical intervention is generally reserved for patients who develop signs of intestinal obstruction or perforation [6]. Our patients both developed unequivocal obstructive and peritoneal signs requiring surgery.

It is possible that patients with multifactorial alterations in coagulation may be at risk for spontaneous bleeding during anticoagulation therapy. Both of the patients in this report had seizure disorders, and were treated with a ketogenic diet in addition to medications. It has been reported that a ketogenic diet increases the likelihood of bruising and minor bleeding in a subset of individuals, in part through diet-induced decrease in platelet responsiveness [14]. In addition, it has been suggested that children with seizure disorders may be at increased risk for bleeding as a side effect of anticonvulsant medications, such as valproic acid [15].

3. Conclusion

Although spontaneous intramural hematoma is a rare event, it is a potentially fatal condition which should be considered in children who develop intestinal obstruction during anticoagulation therapy. Modern imaging can be informative both in diagnosing and localizing such intramural lesions. Patients with additional propensity for altered coagulation, a group that may include children treated with ketogenic diets or anticonvulsants, may be at increased risk for this complication.

Conflict of interests statement

The authors of this paper have no conflicts of interest to disclose.

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