


Incidental finding of intra-abdominal hemangioma in an infant presenting with intestinal obstruction: a case report

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Hemangiomas are the most common benign vascular tumours of the pediatric age group. They originate from embryonic remnants of unipotent angioblastic cells. The liver is the most common site for hemangioma in the abdomen.¹ The prevalence of hepatic hemangioma (HH) varies from 1% to 20% in the general population, and it is two to five times more common in women. HH is usually detected incidentally and shows no symptoms; therefore, most of the time it does not require any treatment.² However, hemangioma may sometimes occur at the atypical location and present with unusual clinical manifestations within the abdominal cavity. Due to these reasons, it leads to diagnostic confusion, resulting in their inadvertent surgical excision.^{1,2} We present here a case of intra-abdominal hemangioma in an infant who presented with the signs and symptoms of intestinal obstruction.

A 4-month-old male infant presented to the pediatric surgery emergency department with excessive crying and bilious vomiting for 2 days. There was a history of associated low-grade fever, which responded to local medications (syrup paracetamol). At presentation, he had a pulse rate of 120/min and a respiratory rate of 28/min. Skin turgor was maintained with no dehydration. Abdominal examination revealed a soft abdomen with mild upper abdomen fullness without any tenderness or lump formation. Ultrasound of the abdomen showed an ill-defined lump in the right lumbar region measuring about 2.4×1.6 cm, and suspicion of intussusception was raised. There was no free fluid in the peritoneal cavity. X-ray of the abdomen showed air-fluid levels with dilated bowel loops (*figure 1A*). Laboratory investigations showed hemoglobin 141 g/L (95–141 g/L), total leukocyte counts of 14.2 × 10⁹/L (6–17.5 × 10⁹/L), and C reactive protein of 18 mg/L (2–5 mg/L). The rest of the parameters were within normal limits. There was no significant personal and family history. For the

initial 24 hours, the patient was kept nil per oral, given intravenous antibiotics and maintenance fluids. However, the patient had similar findings on repeat imaging. Exploratory laparotomy was planned owing to suspicion of intussusception. Laparotomy revealed normal-looking bowel without any intussusception or malrotation. There were few congenital/adhesive bands, one of which was encircling proximal ileum, causing partial obstruction. The bands were converging to a yellowish color structure, which was adhered to the liver margin (*figure 1B*). These bands, along with yellowish structure, were cut using harmonic in flush with Glisson's capsule at the liver margin and were sent for histopathological examination (*figure 2A*). On gross examination, the yellowish structure measured 1.5×1.3×0.5 cm. The microscopic examination showed a moderately cellular lesion composed mainly of capillaries with variable-sized and dilated vascular channels. The vascular channels were lined by flattened endothelial cells, highlighted on CD34 immunostaining (*figure 2B*). Findings were consistent with involuting hemangioma. The child was asymptomatic in the postoperative period and was doing well in the follow-up period at 2 years.

Hemangiomas are benign vascular tumours, the most common benign soft-tissue neoplasm of the pediatric age group, that grow by endothelial cell proliferation. Surgeons and pathologists must differentiate hemangiomas from vascular malformations, which are structural anomalies of vessels.^{3,4} The prevalence of hemangioma is around 12% in less than 1 year of age. Hemangiomas are more commonly seen with premature infants, increased maternal age, female sex, white race, and twins.^{4,5} Based on the size of vessels on histopathology, hemangioma is divided into capillary, cavernous, and mixed type.⁵

HH shows growth and involution patterns similar to cutaneous hemangioma. The



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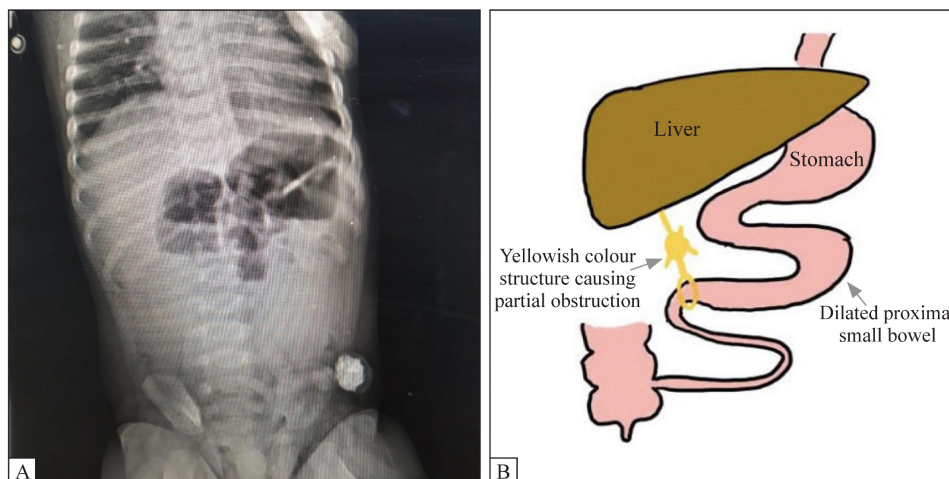


Figure 1 (A) X-ray of the abdomen showing proximal bowel dilated loops with air-fluid levels. (B) Schematic diagram to show intraoperative findings (illustrated by Parveen Kumar and drawn by Arti Khatri).

mainstay of hemangioma treatment is to wait and watch as the majority follow a natural course of proliferation, phase of involution, and finally regress without any active management. HH commonly shows no symptoms and is often incidentally detected.⁶

The most common symptom of abdominal hemangioma (when symptomatic) is abdominal pain and very rarely it can cause bleeding, rupture, and pressure symptoms due to compression of adjacent organs. Also, very few patients have complications secondary to HH, such as high-volume shunting, intra-abdominal bleeding, pressure symptoms on other organs, congestive heart failure, and Kasabach-Merritt syndrome. Kasabach-Merritt syndrome is characterized by hemolytic anemia, thrombocytopenia, prolonged prothrombin time, and hypofibrinogenemia. Thus, above are the findings to go for active management of hemangioma. However, the majority of these lesions do not require any surgical excision.^{2,7,8}

Compared with vascular malformations on microscopy, hemangiomas show plump endothelial cells, endothelial mitoses, and the absence of intralésional nerve bundles. However, hemangiomas can show flattened endothelial lining and variable-sized dilated blood vessels in the

involution phase.⁹ The role of immunohistochemistry comes in the capillary-predominant vascular lesion. Similar to the vascular malformation, the large draining blood vessels can be seen in involuting hemangioma and in congenital hemangioma. CD34 shows immunoreactivity for vascular endothelial cells. The other immunostains, such as glucose transporter protein isoform-1, are useful sensitive and specific immunostaining in delineating the endothelial cells of infantile hemangioma and in delineating the intralésional nerve bundles of vascular malformations. WT-1 is a tumour suppressor gene, which encodes for a transcription factor that shows cytoplasmic immunoreaction in hemangiomas, whereas vascular malformation is WT-1 negative.^{8,9}

The treatment options are many; however, no standard algorithmic approach has been determined to date. Many asymptomatic hemangiomas undergo spontaneous regression in the first year of life. In the management of symptomatic hemangiomas, pharmacotherapy in the form of beta-blockers, steroids, and interferons is preferred over surgery and laser ablation nowadays. The latter ones are used in complex cases.^{6,9}

In the current case, although the preoperative and intraoperative diagnosis of the hemangioma was not made, surgical excision was done for the diagnostic purpose. The initial and later ultrasounds did not pick up hemangioma, as it may have been already in the involution phase. No abnormality was detected in the liver on scans. MRI is a diagnostic aid to diagnose hemangiomas. However, we believe that it would not have been helpful in the index case, given the already involuting nature of the lesion. In addition, MRI does not detect adhesive/fibrous bands often in such cases.

This report documents our experience with a case of incidental involuting hemangioma on the surface of the liver with adhesive bands in a 4-month-old infant with signs and symptoms of intestinal obstruction and with a provisional clinical diagnosis of intussusception. Knowledge of atypical location and presentation of

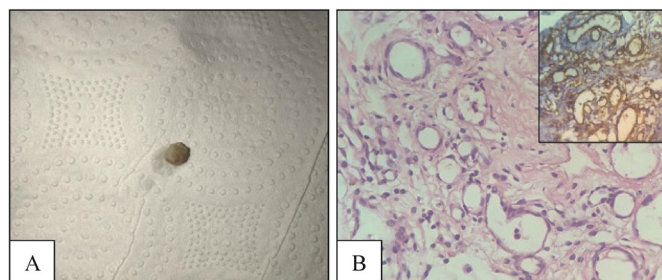


Figure 2 (A) Gross photomicrograph of yellowish colored structure. (B) Microscopy examination shows variable-sized vascular channels (predominantly capillaries) lined with flat endothelial cells (H&E, 400x). Inset showing endothelial cells showing CD34 positivity (immunohistochemistry, 400x).

hemangioma as an atypically localized small mass in the abdomen during an examination, imaging and intraoperatively can avoid inadvertent diagnostic errors in pediatric patients. There are patients' perspectives: First of all, I want to thank doctors for treating my child. When we visited the hospital with our four months baby, he had abdominal distension. After examination and various tests, the surgeon told us that they have to perform emergency surgery. After surgery, the doctor informed us that our child had a benign tumour which they have already removed. After two years post-surgery, our child is doing fine.

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REFERENCES

- 1 Ojili V, Tirumani SH, Gunabushanam G, *et al*. Abdominal hemangiomas: a pictorial review of unusual, atypical, and rare types. *Can Assoc Radiol J* 2013;64:18–27.
- 2 Vilgrain V, Boulous L, Vullierme M-P, *et al*. Imaging of atypical hemangiomas of the liver with pathologic correlation. *Radiographics* 2000;20:379–97.
- 3 Mansour S, Abofoul S, Bahouth H. Parietal peritoneal hemangioma mimicking acute appendicitis: a case report. *Gen Surg Rep* 2018;2:4.
- 4 George A, Mani V, Noufal A. Update on the classification of hemangioma. *J Oral Maxillofac Pathol* 2014;18:117–20.
- 5 Regula J, Wronska E, Pachlewski J. Vascular lesions of the gastrointestinal tract. *Best Pract Res Clin Gastroenterol* 2008;22:313–28.
- 6 Ernst L, Grabhorn E, Brinkert F, *et al*. Infantile hepatic hemangioma: avoiding unnecessary invasive procedures. *Pediatr Gastroenterol Hepatol Nutr* 2020;23:72–8.
- 7 Ilhan M, Oner G, Gök AFK, *et al*. Treatment of atypically-localized cavernous hemangioma in abdomen with atypical pain. *Int J Surg Case Rep* 2016;25:24–7.
- 8 Khatri A, Mahajan N, Khan NA, *et al*. Kaposiform haemangioendothelioma of duodenum in a neonate. *BMJ Case Rep* 2021;14:e239527.
- 9 Rastogi K, Singh L, Khan NA, *et al*. Benign vascular anomalies: a transition from morphological to etiological classification. *Ann Diagn Pathol* 2020;46:151506.