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A Rare Case of Intra-Abdominal Lympho - Venous Malformation Detected in Utero with Post Natal Transabdominal Ultrasonography and Histopathological Correlation

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Abstract*—Veno lymphatic malformations are congenital malformations showing) slow flow lymphovenous anomalies with lymphatic differentiation, commonly seen in head, neck and axilla. They are congenital, often asymptomatic. Abdominal lymphatic malformation can arise from mesentery, omentum, gastrointestinal tract, and retroperitoneum and are rather rare and account for approximately 5% of all lymphatic malformation. Depending on the site, size and symptoms, treatment options vary from conservative management to surgical resection. In our present case, we discuss the intrauterine sonographic imaging appearance of abdominal lymphatic malformation incidentally detected in a later trimester scan with histopathological confirmation.*

***Keywords*—** *Veno-lymphatic malformations, congenital malformation, vascular anomalies.*

I. INTRODUCTION

Lymphatic malformations are rare congenital vascular anomalies. They are usually located in head and neck. Abdominal lymphatic malformations are rare, comprising only 5% of all lymphatic malformations[1]. Within the abdominal cavity, they occur in mesentery, retroperitoneum, solid organs (liver, spleen, pancreas) and gastrointestinal tract[2]. Usually there are no symptoms but symptoms of an acute tumor may appear and require emergency surgery[3]. Complications such as gastrointestinal obstruction, bleeding and volvulus require treatment in all cases. Surgery remains the treatment of choice. In this case we present the imaging and histopathological findings of an intra natal abdominal lymphatic malformations.

CASE: A 24-year-pregnant female presented for regular antenatal check-up at 36 weeks and 5 days of gestation. She had no complaints and no bad obstetric history. On clinical examination, she was hemodynamically stable and her haemoglobin was 9.6 g/dl which was less, as is in most of the expectant mothers in our part of the world. She was subject to a trans abdominal ultrasonography which revealed an ill-defined hyperechoic soft tissue intrabdominal lesion inferior to the liver and anterior to the abdominal aorta and majority of the bowel loops(fig-1).

There were intra lesional hypoechoic serpiginous spaces which on Color Doppler revealed no significant flow or pulsations within the lesion. The intralesional cystic spaces showed no peristalsis. After birth the patient was sent for USG evaluation of abdomen to confirm the pre - natal findings. Post natal followup (fig-2) of the patient revealed the real extent of the lesion occupying a large portion of the mesentery, with some clear ascites displacing the bowel loops with no involvement of the adjacent structures.

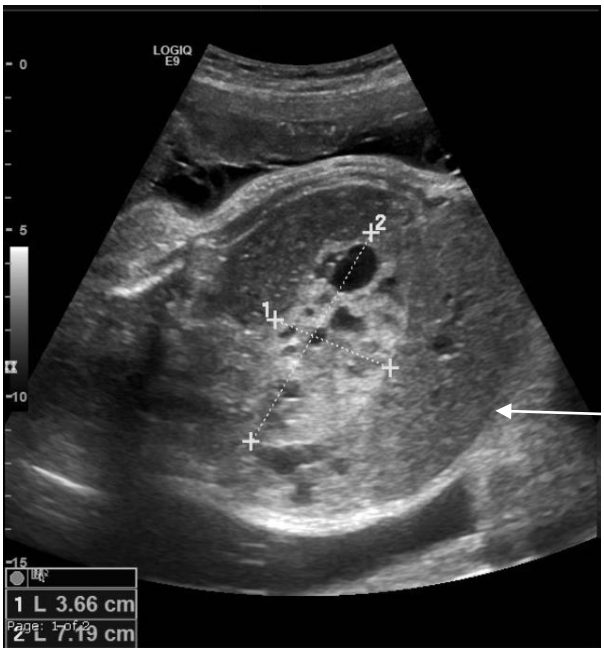

Fig. 1. Grey scale image showing echogenic lesion insinuating between bowel loops.

Post natal abdominal plain film was taken and several small well defined radio opacities were identified in the site of

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the lesion (Fig. 3), which would suggest a differential diagnosis of meconium peritonitis as it is commonly seen in it. On ultrasound the calcifications were present within the complex cystic areas suggestive of intravascular calcifications, inconsistent with meconium peritonitis. Moreover the new born had passed meconium normally and was asymptomatic.

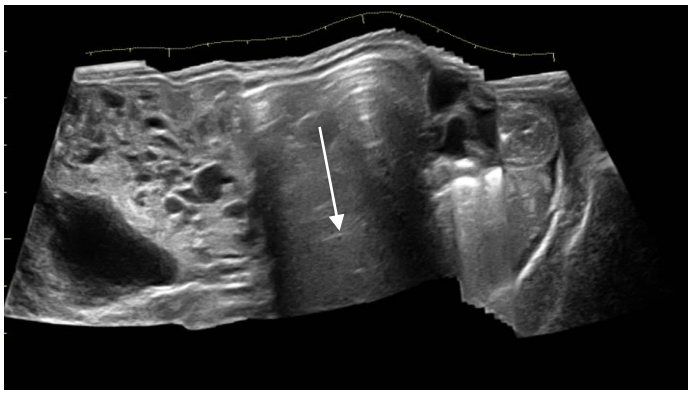
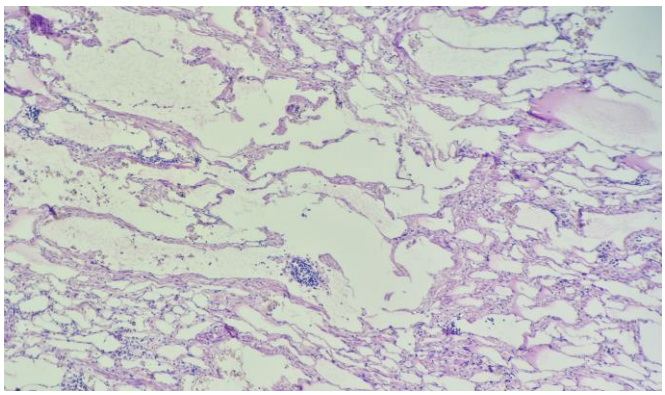


Fig. 2. Panoramic sonogram of the post-natal child showing the relationship of the lesion to the rest of bowel

Later patient presented with pain in epigastric area accompanied with low grade intermittent fever, anorexia and vomiting for 10 days. Lab investigations showed neutrophilic leucocytosis with raised CRP and ESR. Ultrasonography revealed a well defined multiloculated cystic lesion in the epigastric region suggesting an infected mesenteric cyst or a lymphangioma. Similar findings were seen on CT scan. The mass was excised through laparotomy and specimen was sent to the pathology department. Larger specimens showed multiple cystic lesions on the mesocolon.

Histopathologic examination revealed a lobular structure consisting of a mixture of small, thin-walled vessels, mostly capillaries, interspersed with numerous channels and larger spaces. The areas were filled with pink fluid, mostly lymph nodes. Numerous variably sized vessels showing branching and intercommunicating complex network of vessels were also seen. Periphery of the lesion showed presence of lipomatous tissue. The mitotic count was low (fig.4). A diagnosis of Lymphovascular malformation with hemangiomatous change was given.



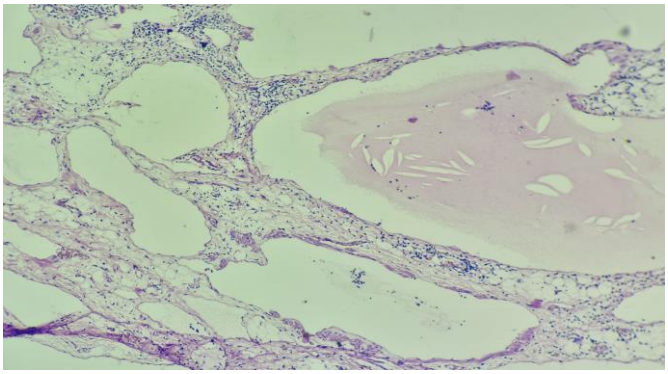


Fig. 3, 4 Photomicrograph of histopathological specimen showing admixture of capillary sized vessels, admixed with few larger venous channels and venules.

The patient was doing well at follow up.

II. DISCUSSION

Vascular malformations are structural lesions resulting from errors of vascular morphogenesis caused by dysfunction in embryogenesis and vasculogenesis[6]. A lymphangioma appears to result from a congenital malformation of the lymphatic vessels rather than a true lymphatic tumor[7]. Lymphangiomas in the jejunum or ileum are extremely rare and account for less than 1% of all lymphangiomas[8].

They are classified into two large groups, hemangiomas and vascular malformations[9], which are further classified into high flow (arterial malformation, arteriovenous fistula, AVM and complex combined vascular malformations), low flow (capillary, venous and lymphatic malformations) or mixed lesions.

TABLE 1. Classification of vascular anamolies[10]

Vascular Anomalies				
Vascular Tumors	**Vascular Malformations**			
	Simple	***Combined***	***Of Major Named Vessels***	***Associated with Other Anomalies***
Benign	Capillary malformations	defined as two or more vascular malformations found in one lesion	abnormalities in the origin/course/number of major blood vessels that have anatomical names	syndromes in which vascular malformations are complicated by symptoms other than vascular anomalies
Locally aggressive or Borderline	Lymphatic malformations Venous malformations Arteriovenous malformations *			
Malignant	Arteriovenous fistula *			

* High-flow lesions.

Venous malformation (VM) is the most common type of congenital vascular malformation (CVM) with an incidence of 1 to 2 in 10,000 and a prevalence of 1% Vascular malformations are composed of ectatic vessels commonly seen in head, neck and extremities and rarely affect the trunk. There are only few case reports of intra abdominal AVM, even more rarer so are those that are detected in utero. A vast forte of differentials exist for such lesions including duplication cysts, hydroureteronephrosis secondary to megacystitis or an inconspicuous posterior urethral valve. In our case, it was an

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incidental finding on routine ultrasound where an echogenic intra-abdominal lesion separates from the bowels caught our attention. Doppler ultrasound (US) and magnetic resonance imaging (MRI) are key imaging methods used to characterize and diagnose vascular malformations The most effective treatment for AVM is transarterial embolization. In conclusion though uncommon it is helpful to keep in mind as one of the differentials such rare entities for an intra-abdominal echogenic lesion especially in the intra uterine setting.

III. CONCLUSION

Lymphangiomas are uncommon benign lymphatic lesions that may occur at virtually any anatomic location in the abdomen[5]. Knowledge of the imaging and pathologic spectrum of abdominal lymphangiomas is necessary when evaluating paediatric and adult patients with intra-abdominal cystic masses. Early surgical intervention is suggested to avoid serious complications.

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