

# Splenic Lymphangioma and Presentation with Frequent Urination : A Case Report

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## Abstract

Lymphangioma of the spleen is a rare disorder with a clinical manifestation ranging from incidental findings of an abdominal mass to symptomatic abdominal pain. The authors reported a case of splenic lymphangioma with the presenting symptom of frequent urination in a 9-year-old boy. A large firm mass was found at the left upper quadrant, 12 cm in diameter. The diagnosis was confirmed by ultrasonography and computerized tomographic (CT) scan. The symptoms of frequent urination disappeared after operative splenectomy. No evidence of recurrence occurred over a 3-year follow-up.

**Key word :** Splenic Lymphangioma, Lymphangioma of Spleen, Frequent Urination

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Lymphangioma is a malformation of the lymphatic vessels. This entity is a benign lesion, but may have multiorgan involvement. It most commonly involves the neck and axilla, and less commonly the mediastinum and retroperitoneum. Landing and Harber (1) classified lymphangioma into 3 groups: lymphan-

gioma simplex, cavernous lymphangioma, and cystic lymphangioma or cystic hygroma. Lymphangioma of the spleen is extremely rare with clinical manifestations ranging from insignificant incidental findings to a large symptomatic cystic mass requiring surgical intervention(2). An associated syndrome may be lym-

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phangiomatosis, in which the lymphangiomatous process involves other sites or organs such as the neck, axilla, mediastinum, lung, liver, spleen, adrenal gland, kidney, and colon(2-7). Most of the reported cases from the literature came to the hospital because of abdominal pain and abdominal masses in the left upper quadrant(2,3,8-10). The authors reported a case of splenic lymphangioma with frequent urination which has never been mentioned previously in the literature. Diagnosis was made initially by ultrasonography and confirmed by histological examination later.

### CASE REPORT

A 9-year-old boy was troubled with frequent urination for one month. Urinalysis (UA) revealed normal finding and unstable bladder was diagnosed. He was treated with imipramine for one month but the symptom was not relieved. Physical examination was reviewed by his physician and a round soft tissue mass was palpable at the left upper quadrant (LUQ). This mass was fixed, firm to hard in consistency, about 12 cm in diameter. No enlargement of cervical, axillary, and inguinal lymph nodes was noted. Ultrasonography of the abdomen revealed a large lobulated soft tissue mass in the LUQ, about 14.2 x 8.4 x 7.2 cm in size. This mass was anterior to the left kidney. The spleen was not visualized. Computerized tomographic (CT) scanning showed a large soft tissue mass similar to the ultrasonographic imaging (Fig. 1). The findings were suggestive of a splenic tumor which might be a malignant lymphoma. Other basic investigations

of abdominal tumor were noted to be normal. The patient underwent laparotomy on March 23, 2000 and the spleen containing tumor was removed. Retroperitoneal nodes were not enlarged. Pathologic examination reported that the specimen consisted of a rubbery dark red spleen, weighing 521 g, and measuring of 14 x 11.5 x 7.5 cm. The outer surface was nodular (Fig. 2). The cut surface showed a dark gray mass and partial fibrosis. The remaining splenic tissue was soft red brown. Microscopic findings showed numerous spaces containing eosinophilic proteinaceous material. They were lined with flat cells. Occasional papillary tufts lined with large endothelial cells were seen (Fig. 3). The final diagnosis was splenic lymphangioma with focal papillary tufts. His post-operative course was uneventful. The symptom of unstable bladder has disappeared since then. Over a 3-year follow-up with abdominal ultrasonography, there was no tumor recurrence. The patient did well and the symptom of frequent urination disappeared after the splenectomy was performed.

### DISCUSSION

Splenic lymphangioma is probably a hamartoma rather than a neoplastic lesion. It can be a small incidental finding, or can be large and multicentric(8, 10). The first report of a splenic lymphangioma was credited to Fink in 1885(2). Since then, approximately 100 cases have been reported(11), covering a broad spectrum of pathology, including focal lesions, large cystic lesion, and diffuse lymphangiomatosis. The

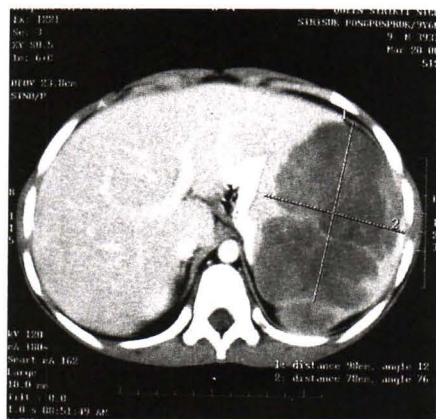


Fig. 1. A large soft tissue mass in the left upper quadrant from the abdominal CT scan.

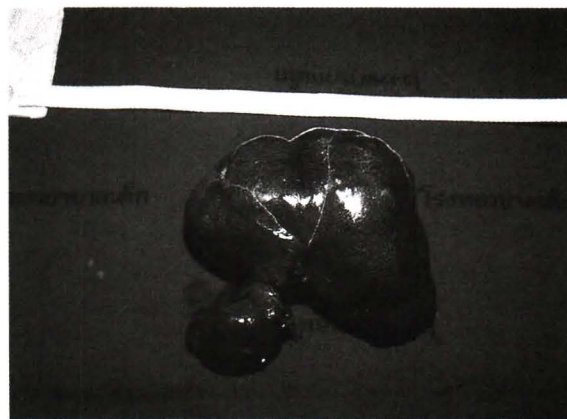
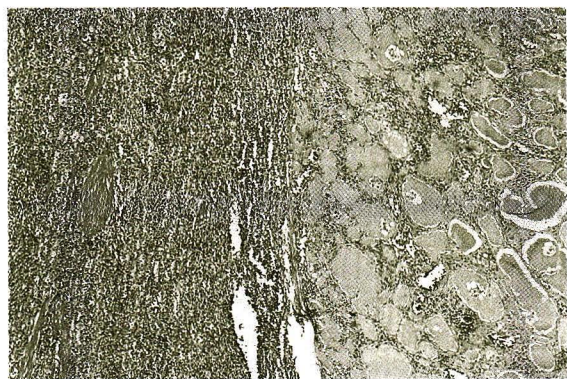


Fig. 2. Splenic tumor with nodular appearance at the outer surface.





3A



3B

**Fig. 3. Microscopic findings showed numerous spaces containing eosinophilic proteinaceous material with focal papillary tuft lined by large endothelial cells (A. from low-power fields, B. from high-power fields).**

lymphangioma of the spleen can involve the spleen alone or can be part of a syndrome of multivisceral organ involvement<sup>(2-7)</sup>. Most cases of splenic lymphangioma may also present as a large abdominal mass and abdominal pain<sup>(2,3,8-12)</sup>. On the contrary, the present case developed the symptom of frequent urination. The mass was incidentally found during meticulous physical examination on the second visit. The symptom of frequency of urination in this case might be caused by either the pressure effect from the large mass to the bladder or a coincidental finding with splenic lymphangioma. The symptom disappeared after splenectomy.

There are several indications for splenectomy. In some patients with splenic lymphangioma, splenectomy may be necessary for histological diag-

nosis. If the spleen is significantly enlarged, it can cause LUQ pain or discomfort. A large spleen is usually at risk for traumatic injury. Coagulation abnormalities including thrombocytopenia, fibrinolysis, bleeding diathesis, hypersplenism, and portal hypertension have also been reported as complications of splenic lymphangioma<sup>(8,13-15)</sup>.

Almost all of the reported cases of splenic lymphangioma were benign lesions. Only two cases were interpreted as probable malignant change in splenic lymphangioma, but neither tumor had metastasized on short-term follow-up<sup>(16,17)</sup>. The patient in the present study had a benign splenic lymphangioma according to the pathological report, and he did well with no evidence of tumor recurrence over a 3-year follow-up.

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## ความผิดปกติของการสร้างระบบทางเดินน้ำเหลืองในม้ามและมีอาการแสดงปัสสาวะบ่อย : รายงานผู้ป่วย 1 ราย

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ความผิดปกติของการสร้างระบบทางเดินน้ำเหลืองในม้ามเป็นโรคที่พบได้น้อยชนิดหนึ่ง ซึ่งมักจะมาด้วยเรื่องที่เราตรวจพบก้อนในท้องโดยบังเอิญหรืออาจมาด้วยอาการปวดท้อง คณะผู้รายงานขอนำเสนอผู้ป่วยเด็กชายอายุ 9 ปี ที่เป็นโรคความผิดปกติของการสร้างระบบทางเดินน้ำเหลืองในม้าม ซึ่งนำมาด้วยอาการถ่ายปัสสาวะบ่อย ก้อนขนาดใหญ่ที่มีเส้นผ่าศูนย์กลางประมาณ 12 ซม. ถูกตรวจพบที่ช่องท้องด้านซ้ายบน การวินิจฉัยได้รับการยืนยันจากการตรวจด้วยอัลตราซาวด์และเอกซเรย์คอมพิวเตอร์ อาการปัสสาวะบ่อยหายไปหลังจากผ่าตัดเอาม้ามออก ไม่มีอาการกลับมาเป็นซ้ำภายหลังตรวจติดตามผลมากกว่า 3 ปี

**คำสำคัญ :** ความผิดปกติของการสร้างระบบทางเดินน้ำเหลืองในม้าม, ถ่ายปัสสาวะบ่อย

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