Giant hemolymphangioma of the abdominal wall. A case report

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Abstract: We present a 61-year-old man with Child B liver cirrhosis that was evaluated because of abdominal pain accompanied by increased volume in the umbilical region. Huge acquired hemolymphangioma of the abdominal wall was diagnosed and confirmed by histopathological examination. The involvement of other organs was ruled out.

Key words: Hemolymphangioma.

Case Report

GENERAL SURGERY

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Introduction

Hemolymphangioma is a rare benign tumor characterized by malformed venous and lymphatic components.[1] They might be local or diffused and preferentially are situated in the head and neck. [2] Symptoms depend on the site of occurrence. Ultrasonography (US), computed tomography (CT), and magnetic resonance imaging (MRI) are useful for the diagnosis. Complete excision provides the best results with a lower recurrence rate. [3,4]

Case report

A 61-year-old man with Child B alcoholic liver cirrhosis for 2 years treated with spironolactone, furosemide, and propranolol. Without a family history of malignant tumor, abdominal trauma, or surgery. He had increased volume in the umbilical region 6 months ago and recently was progressive in his size, firmness, and tenderness. The main complaint was abdominal pain in the umbilical region 6 hours before admission to the emergency department. Physical examination showed a swollen abdomen secondary to ascites with erythema and fluid leakage through skin ulceration at the umbilical region under that, a tumor of 7 cm in diameter was palpable with irregular margins and fixed to deep layers with tenderness but von Blumberg sign negative. (Figure 1-A)

Biochemical workup showed: WBC 6.01x103 /µL, hemoglobin 13.30 g/dL, platelets 80x103µL, DHL 312 UI/L, creatinine 1.48 mg/dL, INR 1. Doppler US demonstrated a subcutaneous septated mass with increased central and peripheral vascularity. Also, free fluid in the pelvic cavity, parietocolic gutters, and subdiaphragmatic space (Figure 1-B). Non-contrast CT showed a heterogeneous, multiseptate, subcutaneous mass with 91.5 x 72.7 x 63.8 mm of diameter and 214 ml of volume (Figures 1-C and 1-D).

Surgical management was made with complete excision of the lesion with negative margins; the intraoperative findings were: A septate and multicystic tumor of 10 x 10 cm, with citrine fluid inside that, involved the fascia of the rectus muscle. The defect of the abdominal wall was closed with smead jones technique with polypropylene 0 and onlay polypropylene mesh.

The patient was discharged without complications on postoperative day two. Histopathological examination exhibited a hemolymphangioma with negative margins in the hematoxylin and eosin (HE) staining (Figure 2-A). Immunohistochemical staining was positive for Factor VIII, CD31, and CD34 (Figures 2-B, 2-C, and 2-D). Follow-up with contrasted CT was performed 1 year after surgery without recurrence (Figures 3 A-B).

Discussion

Hemolymphangioma´s incidence varies from 1.2 to 2.8 per 1000 newborns and rarely found in the adult population.[5] Acquired hemolymphangioma occurs due to inadequate lymphatic drainage and damage to the lymphatic vessels as a result of surgery or trauma.[3] In our case, the tumor developed in an adult man and was confined to the abdominal wall, without previous surgery or trauma. The involvement of other organs was ruled out. The symptoms depend on the size and place of occurrence, even can mimic malignancy. The main complaint in our patient was abdominal discomfort secondary to the tumor size. It was complicated by ulceration of the skin and

From the Department of Surgery at North Central Hospital PEMEX. Mexico City, Mexico. Received on June 23, 2021. Accepted on July 3, 2021. Published on July 19, 2021.

DOI: 10.17605/OSF.IO/8ZQG7

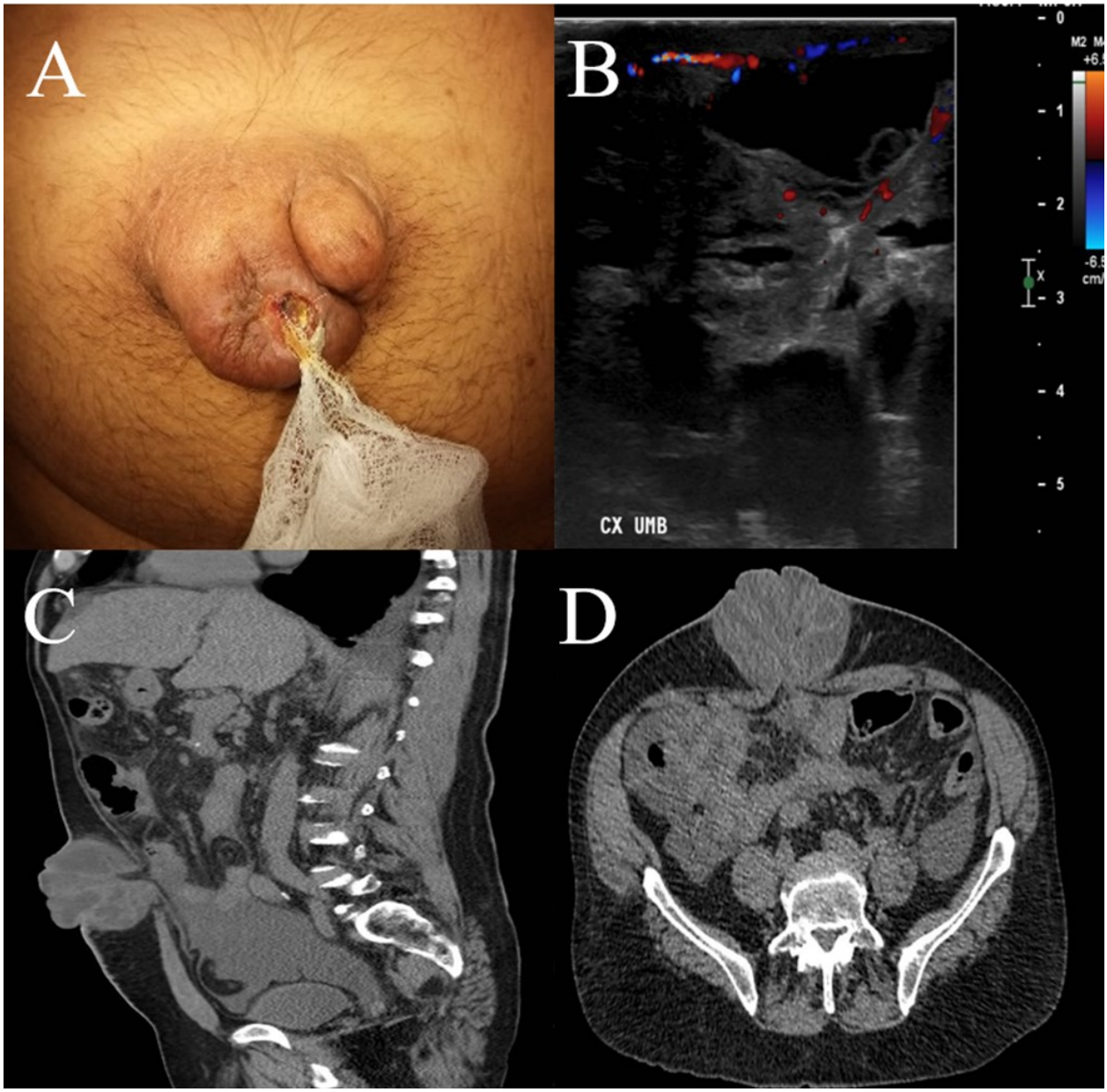

Figure 1. (A) Tumor with erythema and fluid leakage through skin ulceration at the umbilical region. **(B)** Doppler US demonstrated a subcutaneous septated mass with increased central and peripheral vascularity. **(C-D)** Non-contrast CT showed a heterogeneous,

lymphorrhea. Other complications are traumatic rupture, torsion, perforation, and hemorrhage.[3,6] Accurate diagnosis before surgery is difficult due to the low specificity of symptoms and signs. On imaging studies, they range from well-defined simple cystic lesions to ill-defined aggressive lesions, it depends on the composition of the blood vessels and any accompanying infection or bleeding. US, CT, and MRI are useful for differentiation of the tissue composition. [4,7] In our case US and CT images showed a cystic-solid tumor. Non-contrast CT was performed due to acute kidney lesion and RMN was not performed because this resource is not available in our hospital. Definitive diagnosis was based on histological evidence after complete excision of the lesion with negative margins. Histopathological findings include cystic or cavernous lesions that consist of dilated veins and lymphatic vessels interspersed with normal stromal tissue and vasculature and immunohistochemical analysis positive for CD31, CD34, and Factor VIII. [2,3] Complete excision provides the best results with a recurrence rate of 10%-27%, while 50%-100% of partially resected tumors can recur, and careful monitoring with CT or ultrasound is necessary. [8] Nonsurgical treatments with cryotherapy, laser therapy, radiotherapy, and local injection of sclerotic agents, are not superior to surgical treatments.[3]

Conclusion

Hemolymphangioma is a rare benign tumor that lacks typical clinical symptoms and specific radiological findings. Involvement only the abdominal wall there has not been described before. Complete tumor resection with negative margins is recommended to avoid recurrence and to confirm the diagnosis.

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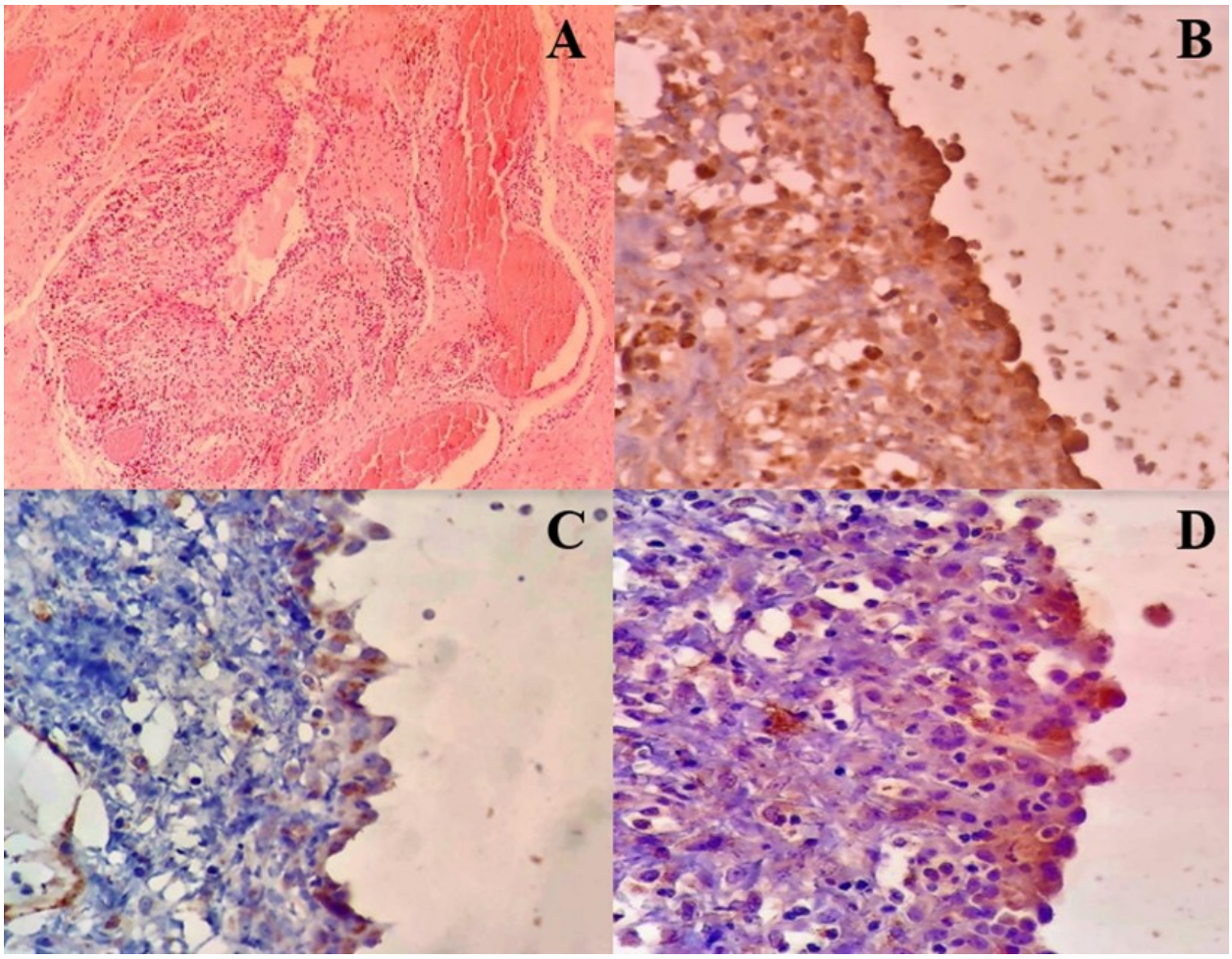

Figure 2. (A) Pathological examination of hemolymphangioma (hematoxylin and eosin staining). Immunohistochemical staining positive for **(B)** Factor VIII, **(C)** CD31 and **(D)** CD34.

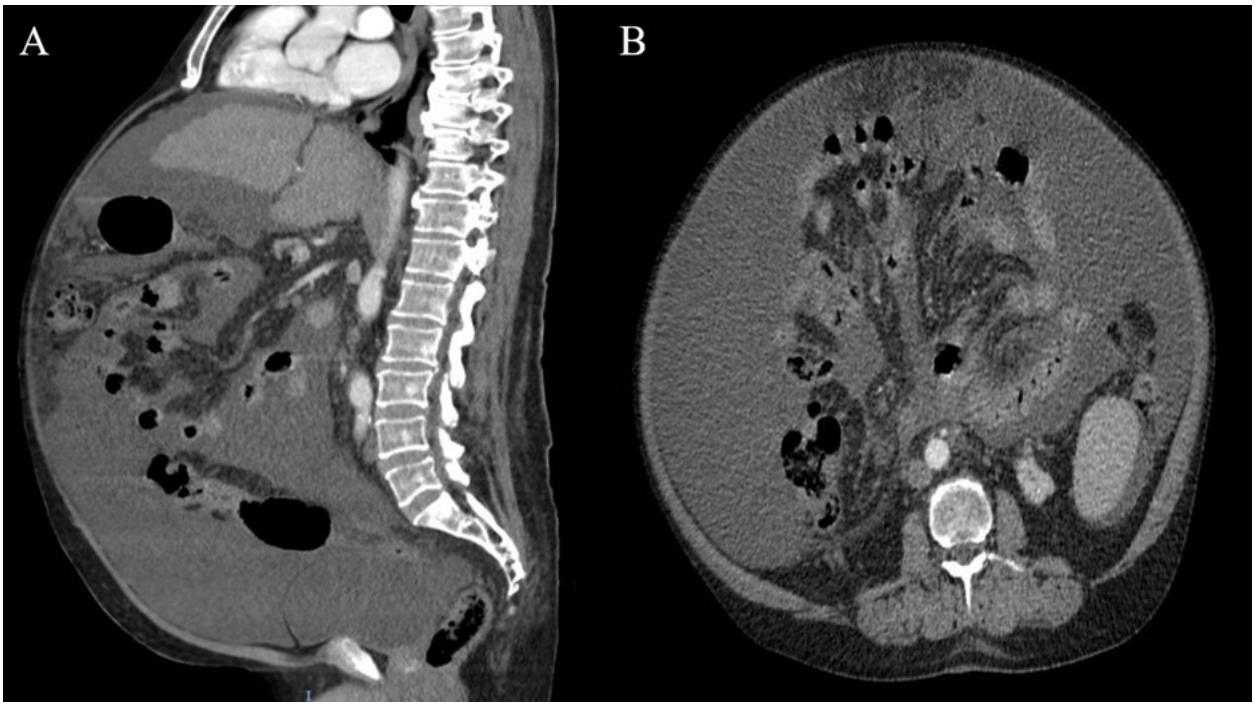

Figure 3 A-B. Contrast CT showed ascites, post-surgical changes at the umbilical region, without recurrence.

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Conflicts of interests

There was no conflict of interest during the study, and it was not funded by any organization.

Acknowledgements

We thank the Pemex Health Services Sub-Directorate for financial support in carrying out the surgery.

References

[1] Teng Y, Wang J, Xi Q. Jejunal hemolymphangioma. Medicine. 2020; https://doi.org/10.1097/MD.0000000000018863
[2] Hou G, Jiang Y, Jian S. Hemolymphangioma involving bones and bladder detected on 68Ga-NEB PET/CT A rare case report. Medicine. 2019; https://doi.org/10.1097/MD.0000000000015213
[3] Ohsawa M, Kohashi T, Hihara J, et al. A rare case of retroperitoneal hemolymphangioma. International Journal of Surgery Case Reports. 2018; https://doi.org/10.1016/j.ijscr.2018.08.030
[4] Mao C, Jin Y, Yang Q, et al. Radiographic findings of hemolymphangioma in four patients: a case report. Oncol Lett. 2018; https://doi.org/10.3892/ol.2017.7268
[5] Yong Mei, Ci-Peng J, Chen L, Li X, et al. Hemolymphangioma of the spleen: A report of a rare case. World J Gastroenterol.2015; https://doi.org/10.3748/wjg.v21.i17.5442
[6] Pandey S, Fan M, Chang D, et al. Hemolymphangioma of Greater Omentum. Medicine. 2016; https://doi.org/10.1097/MD.0000000000003508
[7] Ballard D, Mazaheri P, Oppenheimer D, et al. Imaging of Abdominal Wall Masses, Masslike Lesions, and Diffuse Processes. Gastrointestinal Imaging.2020; https://doi.org/10.1148/rg.2020190170
[8] Pan L, Jian-bo G, Javier P. CT Findings and Clinical Features of Pancreatic Hemolymphangioma. Medicine. 2015; https://doi.org/10.1097/MD.0000000000000437

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 DOI: 10.17605/OSF.IO/8ZQG7