Cystic lymphangioma presenting as an ovarian mass in an adolescent: A case report and literature review

RUI SUN and CHUN-RUN YANG

Department of Obstetrics and Gynecology, Shandong Provincial Hospital Affiliated to Shandong First Medical University, Jinan, Shandong 250021, P.R. China

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Abstract. Cystic lymphangiomas of the abdominal cavity are rare benign lymphatic malformations that can be difficult to distinguish from other cystic abdominal tumors on preoperative imaging. The present study reported a rare case of a giant abdominopelvic cystic lymphangioma in a 16-year-old female who presented with abdominal pain following exercise and was initially suspected to have a benign ovarian tumor. Despite comprehensive imaging evaluation, however, a definitive diagnosis could not be established preoperatively. The final diagnosis of abdominal cystic lymphangioma was confirmed by histopathological examination following surgical resection. The present case has highlighted the rarity and diagnostic challenge of differentiating abdominal cystic lymphangiomas from large benign ovarian tumors in adolescents. It underscores the importance of maintaining a broad differential diagnosis when evaluating abdominopelvic cystic masses in young females. Surgical resection remains the cornerstone of treatment and is associated with favorable outcomes, and regular postoperative follow-up is essential to detect recurrence.

Introduction

Lymphangiomas are rare benign tumors of the lymphatic system that can occur anywhere in the skin or mucous membranes (1). They are typically classified as deep or superficial lesions and they have been shown to be either congenital or acquired in origin (2). Congenital lymphangiomas result from lymphatic obstruction during fetal development, although their exact cause has yet to be fully elucidated (3). They are usually diagnosed at birth or within the first 5 years of life, and have been associated with genetic conditions such as trisomy 13 (or Patau syndrome), trisomy 18 (or Edwards syndrome), trisomy 21 (or Down syndrome) and Turner and Noonan syndromes (4). By contrast, acquired lymphangiomas are often secondary to trauma, surgery or inflammatory lymphatic obstruction, and typically occur in adults (5,6). Abdominal lymphangiomas are particularly uncommon, accounting for ~1% of all cases of lymphangioma (7). The condition has rarely been reported in adolescent patients, and cases in girls and adult women are also uncommon, most often involving mesenteric or retroperitoneal locations with variable clinical presentations. Although numerous patients remain asymptomatic, progressive enlargement of the lymphangioma may result in nonspecific symptoms, including nausea, vomiting, pain or intestinal obstruction (8,9). Importantly, in adolescent females, large abdominal cystic lymphangiomas may closely mimic benign ovarian tumors (BOTs) on imaging, which frequently leads to diagnostic uncertainty and delayed recognition.

Previous reports in patients have largely described smaller lesions or cases with an established preoperative diagnosis (10,11); however, cases of giant abdominopelvic cystic lymphangiomas presenting with acute symptoms and masquerading as ovarian neoplasms remain poorly documented. At present, the optimal diagnostic approach and surgical management in such scenarios are therefore not well defined. In this case report, a rare case of a giant abdominal cystic lymphangioma in a 16-year-old female presenting with acute abdominal pain was described. The lesion extensively involved the abdominopelvic cavity and was initially presumed to be of ovarian origin based on imaging findings. Through detailing the diagnostic challenges, intraoperative findings and surgical management, this case has added to the limited literature that exists on abdominal lymphangiomas in adolescents, thereby underscoring the importance of including this rare entity in the differential diagnosis of large cystic abdominal masses in young females.

Case report

A 16-year-old female presented with abdominal pain for 5 days following abdominal exercise in November 2023 at Shandong Provincial Hospital Affiliated to Shandong First Medical University (Jinan, China). The patient had taken self-administered ibuprofen without relief and the pain progressively worsened, making it impossible for the patient to lie flat.

Correspondence to: Dr Chun-Run Yang, Department of Obstetrics and Gynecology, Shandong Provincial Hospital Affiliated to Shandong First Medical University, 324 Jingwuweiqi Road, Jinan, Shandong 250021, P.R. China
E-mail: yangchunrun@sdfmu.edu.cn

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Additional symptoms included fatigue, dyspnea, dizziness and headache, without abdominal distension or diarrhea. The patient's medical and menstrual history were unremarkable.

Ultrasound (US) revealed the presence of a large multilocular cystic mass in the abdominopelvic cavity, extending up to the xiphoid process and bilaterally to the anterior axillary lines. The mass exhibited clear borders, poor internal echogenicity, multiple septations and irregular cyst wall thickness and was closely associated with the left ovary. The US diagnosis suggested a mucinous cystadenoma of the ovary with intracystic hemorrhage. A computed tomography (CT) scan indicated a right adnexal tumor origin with no enlarged lymph nodes (Fig. 1). Magnetic resonance imaging (MRI) analysis revealed the presence of a large cystic lesion, measuring ~28.1x16.7x8.1 cm, with clear borders, heterogeneous internal signals, uniform cyst wall and septal thickness and pronounced enhancement (Fig. 2). The lesion's lower margin was closely associated with the left adnexa, raising suspicion of an adnexal cystadenoma. Serum tumor markers (including carcinoembryonic antigen, alpha-fetoprotein and cancer antigen 125) and sex hormone levels were found to be normal.

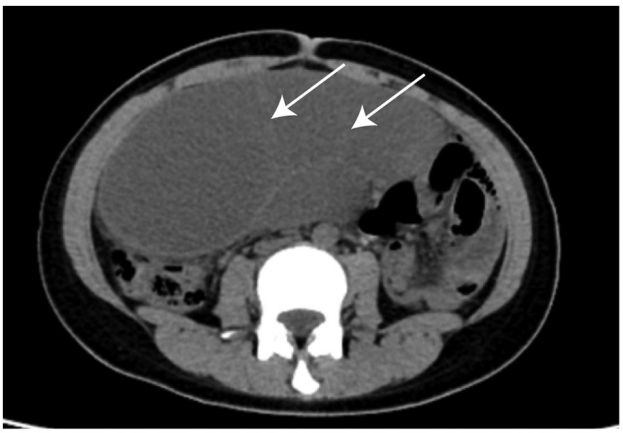

Figure 1. CT scan demonstrating a giant multilocular cystic mass occupying the entire abdominopelvic cavity. Multiple internal septations are visible (arrows), with well-defined margins and no solid enhancing components. The lesion displaces adjacent organs, including the bowel loops and adnexal structures.

Based on the presumptive diagnosis of a benign ovarian cystic tumor, and given the giant size of the lesion, a minimally invasive approach was initially planned. Single-incision laparoscopic surgery was used to optimize the cosmetic outcome and to reduce surgical trauma in this adolescent patient. To facilitate safe manipulation and improve visualization, controlled cyst aspiration was performed intraoperatively using a suction device after having carefully isolated the cyst wall. A total of ~2,000 ml pale-yellow, clear, watery fluid was aspirated and the cyst wall was sutured to reduce leakage during mobilization. Laparoscopic exploration revealed that the mass was not of adnexal origin, but was densely adherent to the inferior liver edge and greater curvature of the stomach, transverse colon and mesentery. The uterus and adnexa were unremarkable. Intraoperative consultation with the gastrointestinal surgery team was sought, and it was determined that the cystic cavity did not communicate with the gastrointestinal lumen. Given the large lesion size, multiloculated cystic architecture, indistinct tissue planes and high risk of cyst rupture, continuation of a purely laparoscopic resection was considered unsafe. These factors limited intraoperative visualization and increased the risk of incomplete excision, as well as potential injury to adjacent organs. Therefore, conversion to open surgery was undertaken to achieve better exposure and to facilitate complete resection, which is widely regarded as the most effective strategy for diagnostic confirmation, curative treatment and reduction of recurrence (11). Following discussion with the patient's family, complete excision of the cyst was performed. The pathological specimen after resection is shown in Fig. 3.

Intraoperative frozen section pathology revealed a benign cystic lesion, mostly lacking epithelial lining, with focal fibrous thickening of the cyst wall. Immunohistochemical analysis was subsequently performed to support the histopathological diagnosis. The endothelial lining cells were found to be positive for lymphatic markers including D2-40, whereas epithelial markers, including cytokeratin, were negative. The final pathological diagnosis was abdominal cystic lymphangioma (Fig. 4; method detailed in the supplementary Data). Postoperative recovery was normal (the patient passed flatus and resumed ambulation on postoperative day 2, returned to a regular diet on day 3 and had the sutures removed on day 9) and the patient was closely monitored for common complications associated with lymphangioma resection, including hemorrhage, lymphatic leakage, infection and bowel dysfunction. Particular attention was paid to signs of chylous ascites or persistent drainage, given the lymphatic origin of the tumor. The patient recovered uneventfully and resumed oral intake (beginning with clear liquids and progressing to a regular diet) gradually, with no evidence of postoperative lymphatic leakage or organ dysfunction. Finally, the patient was discharged on postoperative day 10 without complications. Follow-up examinations at 1 and 6 months post-surgery were both normal, and the follow-up interval was subsequently extended to once a year. At the 1.5-year follow-up, the patient remained asymptomatic without radiological evidence of recurrence (Fig. 5).

Discussion

Lymphangioma was first discovered and defined in 1913 as a benign tumor (12). In excess of 80% of cases are diagnosed in early childhood and are typically attributed to congenital lymphatic malformations (13). The remaining cases are typically identified around the age of 40 years and are often secondary to trauma, having received prior surgery or inflammatory obstruction of the lymphatic vessels (14). Pathologically, lymphangiomas are primarily classified into three types: Simple capillary, cavernous and cystic lymphangiomas (15).

Abdominal lymphangiomas are especially rare, accounting for ~1% of all cases; furthermore, their presentation in adolescents is particularly rare (5,16). These lesions most commonly arise in areas surrounded by loose connective tissue, such as the mesentery and retroperitoneum, and are predominantly of the cystic type (15). As a result, abdominal cystic lymphangiomas in this age group are frequently underrecognized and may pose significant diagnostic challenges. Several previously published studies (11,17-19) have described abdominal or omental cystic lymphangiomas that closely mimicked ovarian tumors, leading

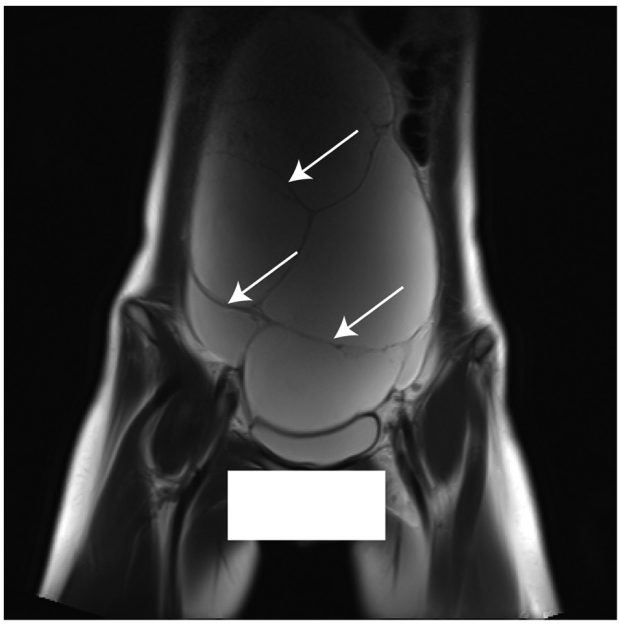

Figure 2. MRI scan (T2-weighted image) of the abdomen and pelvis showing a large multilocular cystic lesion with heterogeneous signal intensity and internal septations (arrows). The lesion appears closely related to the adnexa, suggesting a possible ovarian origin on imaging. This case illustrates the limitation of MRI in accurately determining the true anatomical origin of giant cystic lesions.

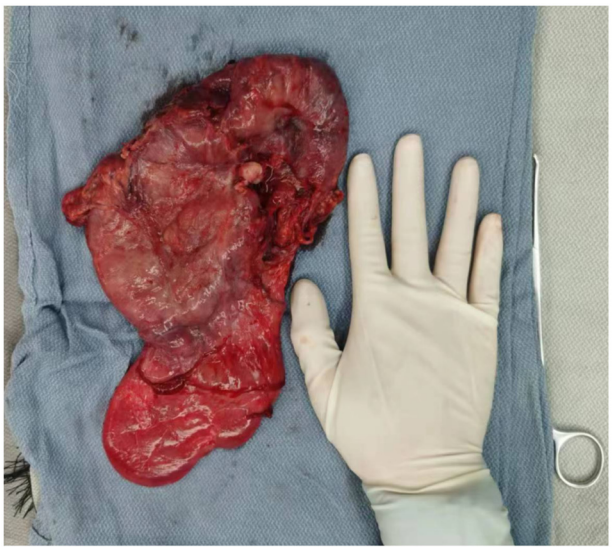

Figure 3. Gross specimen of the resected lesion presenting as a large multilocular cystic mass with thin walls and multiple internal septa. The cyst contained a clear yellow serous fluid. No solid nodules were identified. The macroscopic appearance supports a benign cystic process and is consistent with cystic lymphangioma.

to diagnostic uncertainty and unexpected intraoperative findings. In these cases, large multilocular cystic masses located in the pelvis or lower abdomen were initially interpreted as ovarian cystadenomas or adnexal tumors on imaging, largely due to organ displacement rather than true ovarian involvement. Similarly to the present case, a definitive diagnosis in the majority of the published reports was only achieved after surgical exploration and histopathological examination, with the ovaries found to be normal and separable from the lesion at surgery. These observations underscore a consistent imaging pitfall in which the apparent proximity of a giant cystic mass to the ovary may obscure its true origin. From a surgical perspective, prior cases (20) have emphasized the importance of flexible operative planning, as extensive adhesions to the omentum, mesentery or gastrointestinal structures are frequently encountered, and may necessitate multidisciplinary collaboration. The present case study has contributed to the existing literature by highlighting that, even with advanced imaging modalities, an adnexal origin cannot be reliably assumed in giant abdominopelvic cystic lesions, and careful intraoperative assessment is essential to avoid unnecessary adnexal resection.

Clinically, abdominal lymphangiomas are often asymptomatic until they enlarge sufficiently to cause compressive or acute symptoms, including abdominal pain, nausea, vomiting or intestinal obstruction (21-23). Acute presentations due to rupture, hemorrhage or torsion have also been reported (24). The severity and nature of symptoms depend on the size and location of the lesion, as well as on its proximity to adjacent vital structures (25). In the present case, the patient presented with exercise-associated abdominal pain and no prior medical history, which further complicated the diagnostic process and broadened the differential diagnosis.

Imaging has a central role in the evaluation of abdominal cystic masses. According to US, CT and MRI analyses (24,26-28), these lesions typically appear as well-circumscribed unilocular or multilocular cystic masses with variable internal septations (24). However, these imaging features have been shown to be nonspecific (29). In females, large abdominopelvic cystic lymphangiomas located adjacent to the adnexa may closely mimic BOTs, as reported in several previous cases (11,30). In the present case, the mass occupied the entire abdominopelvic cavity and appeared to be closely associated with the ovary on preoperative imaging, leading to an initial diagnosis of a large BOT. This highlights an important imaging pitfall and underscores the limitations of preoperative radiologic assessment in distinguishing lymphangiomas from adnexal tumors.

Definitive diagnosis often relies on surgical exploration and histopathological examination. In the present case, intraoperatively, the identification of extensive adhesions to the stomach, colon and mesentery, along with the preservation of normal ovarian anatomy, suggested an omental origin, rather than a gynecological one. Histopathological findings of dilated lymphatic spaces lined by endothelial cells (31) have been used to confirm the diagnosis of cystic lymphangioma. The use of lymphatic-specific immunohistochemical markers, particularly D2-40 and cytokeratin, is critical for differentiating cystic lymphangioma from other cystic lesions of ovarian, mesothelial or gastrointestinal origin, especially in cases with atypical location or misleading imaging findings. These intraoperative and pathological findings emphasize the importance of careful surgical assessment to avoid unnecessary resection of reproductive organs in young patients.

The management of abdominal cystic lymphangiomas largely depends on symptomatology and lesion size (32). Although spontaneous regression has been reported in a minority of asymptomatic cases, surgical excision remains

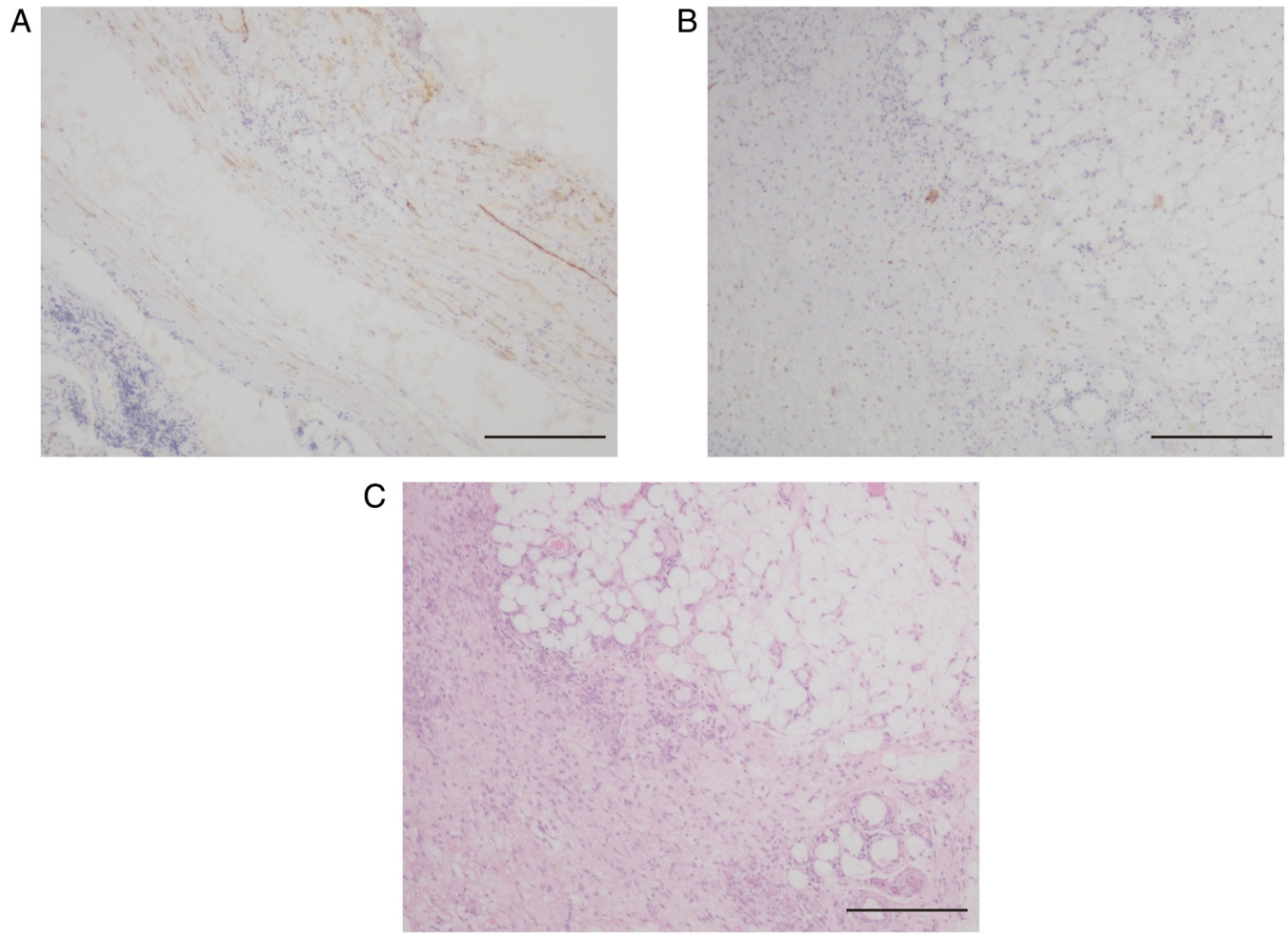

Figure 4. Histopathological and immunohistochemical findings of the lesion confirming the diagnosis of cystic lymphangioma. (A) D2-40 staining shows strong positivity in the endothelial lining cells, indicating a lymphatic origin. (B) Cytokeratin staining is negative, excluding an epithelial cystic lesion. (C) Hematoxylin and eosin staining demonstrates dilated cystic spaces lined by flattened endothelial cells without cytologic atypia (scale bars, 200 μm).

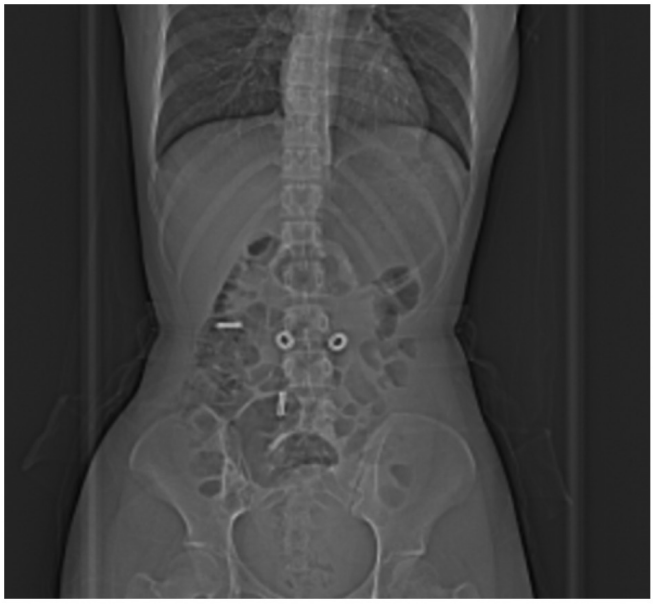

Figure 5. Follow-up CT scan obtained 1.5 years after surgery showing no evidence of recurrence or residual lesion. The abdominal and pelvic organs are in a normal anatomical position. This image demonstrates the favorable outcome following complete surgical excision.

the treatment of choice for symptomatic or large lesions. Notably, even when preoperative differentiation from BOTs is not possible, surgical resection is often indicated and awareness of alternative diagnoses may assist surgeons in tailoring intraoperative decision-making and surgical planning.

Complete excision is associated with a significantly lower recurrence rate compared with incomplete resection (a rate of 35-64% compared with 17-24%), highlighting the need for a meticulous surgical technique (13,30,33,34). Nonsurgical treatments, including localized radiotherapy, chemotherapy or sclerotherapy, have been described; however, these approaches are associated with relatively high rates of complication and recurrence (34). Sclerotherapy is generally more suitable for well-circumscribed, unilocular lymphatic malformations that lack diagnostic uncertainty. In addition to achieving complete resection, postoperative management has a critical role in optimizing outcomes. Given the risk of recurrence, long-term postoperative follow-up is essential. Regular post-operative surveillance, including periodic clinical evaluation and imaging follow-up, is essential for the early detection of recurrence, particularly in cases involving large or complex lesions. Furthermore, careful intraoperative assessment to ensure complete removal of the lesion and adjacent lymphatic

channels, when necessary, may help reduce the risk of recurrence.

In summary, the present case has contributed to the limited number of studies that have been published on adolescent abdominal cystic lymphangiomas masquerading as ovarian tumors. It reinforces key clinical teaching points, including the diagnostic limitations of imaging, the importance of maintaining a broad differential diagnosis for large abdominopelvic cystic masses in adolescent females and the value of careful intraoperative assessment to guide appropriate surgical management and organ preservation. Based on the diagnostic challenges encountered in this case and previous reports, a schematic diagnostic approach for large abdominopelvic cystic masses in adolescent females may be proposed (Fig. S1).

In conclusion, giant abdominal cystic lymphangiomas are rare and may closely mimic BOTs, particularly in adolescent female patients, thereby posing a significant diagnostic challenge. The present case has underscored the importance for gynecologists to maintain a broad differential diagnosis when evaluating large abdominopelvic cystic masses and to involve multidisciplinary teams early in management. For radiologists, an awareness both of the imaging features and of limitations in distinguishing lymphangiomas from ovarian or gastrointestinal cystic lesions is critical to guide appropriate preoperative planning. For pediatric and general surgeons, this case highlights the necessity for individualized surgical strategies, intraoperative flexibility and the priority of complete excision to minimize recurrence. Comprehensive postoperative surveillance remains essential and multidisciplinary collaboration is key to improving diagnostic accuracy, optimizing surgical outcomes and ensuring long-term disease control in this uncommon, but clinically significant condition.

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Availability of data and materials

The data generated in the present study are included in the figures and/or tables of this article.

Authors' contributions

CY was responsible for the design of the study, performed the surgery and revised the manuscript. RS was responsible for the acquisition of data, performed the literature search and writing the draft of the manuscript. Both authors checked and confirmed the authenticity of the raw data, and have read and approved the final manuscript.

Ethics approval and consent to participate

The study was conducted according to the principles of the World Medical Association's Declaration of Helsinki.

Patient consent for publication

Written informed consent was obtained from the patient and the patient's legal guardian for publication (including clinical data, imaging and pathological and images).

Competing interests

The authors declare that they have no competing interests.

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