Radiology Case Reports 19 (2024) 3970–3974

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Case Report

Large ovarian mature teratoma with gliomatosis peritonei in a young female: A case report ☆

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ARTICLE INFO

Article history:
Received 27 March 2024
Revised 1 June 2024
Accepted 4 June 2024

Keywords:
Immature teratoma
Dermoid
Enhancing peritoneal nodules
Glial tissues

ABSTRACT

Gliomatosis peritonei is an extremely rare condition usually associated with either immature teratoma or, less commonly, mature teratoma. We present a case of a young female with long-standing progressive abdominal distension, who was diagnosed with mature ovarian teratoma with gliomatosis peritonei and gross ascites. The final diagnosis in this case was determined through the correlation of imaging, operative, and histopathological findings. The presence of enhancing peritoneal nodules usually leads to a suspicion of peritoneal carcinomatosis or abdominal tuberculosis, especially in endemic regions; however, gliomatosis peritonei should always be considered in the differential diagnosis, particularly when associated with teratomas. Radiological findings combined with histopathological reports are valuable in reaching the final diagnosis in these cases.

Introduction

Gliomatosis peritonei (GP) is an extremely rare condition, with only a limited number of cases documented in medical literature [1]. GP represents the deposition of mature glial elements into the peritoneum and is usually associated with either immature teratoma or, less commonly, mature teratoma [2,3]. However, there are certain other conditions associated with GP described in the literature, some notable ones being gastric teratoma, hepatic teratoma, and endometrial adenocarcinoma [4–6]. GP primarily affects young females, likely because of its strong association with ovarian teratomas [1]. Cases of GP have been reported from different parts of the world without obvious geographical predilection.

Here we present a case of a young teenage girl with long-standing progressive abdominal distension, who was diagnosed with mature ovarian teratoma with GP and gross

☆ Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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https://doi.org/10.1016/j.radcr.2024.06.018

ascites. Long-standing progressive ascites without significant abnormalities in the lab parameters should lead to the suspicion of some underlying tumoral etiology or some chronic infection like tuberculosis, especially in endemic zones. Imaging in such cases is vital to rule out the cause of ascites.

Case report

A 19-year-old female presented with progressive abdominal distension over the last 6 months. Living in a rural area, she had visited the primary health center only a few times, none of which included detailed investigations. During this period, she took some pain medications and proton pump inhibitors for occasional abdominal discomfort. Her family history was unremarkable. As her symptoms progressed further, she was referred to our center for further evaluation. On examination, her abdomen was distended with a dull note on percussion. Ascitic fluid analysis revealed an elevated total leucocyte count and was negative for malignancy or tuberculosis. Serum tumor marker profile showed elevated cancer antigen 125 (CA-125) and cancer antigen 19-9 (CA 19-9) levels, with a normal alpha-fetoprotein (AFP) level.

An ultrasound of the abdomen and pelvis revealed ascites with few loculations and a heteroechoic solid-cystic abdominopelvic mass with internal calcifications (Figs. 1A and B). An abdomen and pelvic contrast-enhanced computed tomography (CE-CT) scan showed a large, well-defined abdominopelvic solid cystic mass measuring 21 × 15 × 13 cm with internal calcifications and fat components. Some enhancing solid components and septa were noted within the lesion. Gross ascites with few enhancing septations were observed. Enhancing irregular nodular peritoneal and omental deposits were noted (Figs. 2A–D). Based on these imaging findings, a provisional diagnosis of immature ovarian teratoma with gliomatosis peritonei was made.

Our case was managed with surgery (staging laparotomy with right salpingo-oophorectomy and omentectomy), and the operative findings were consistent with the radiological findings. There was a large cyst with fat and calcific components, consistent with teratoma. Multiple nodularities were noted in the peritoneum and omentum (Fig. 3). The excised surgical specimen was sent for histopathology. Multiple sections taken from the cyst stroma revealed mature glial tissue, cerebellar tissue, bony tissue, pilosebaceous units, and mature adipocytes (Figs. 5A–C). Immature neural elements were not seen. Sections examined from the omentum and peritoneum showed fibrofatty tissues with nodules of mature glial tissues (Fig. 4). After correlating with the histopathological report, a final diagnosis of mature ovarian teratoma with gliomatosis peritonei was made. Our patient is doing well 6 months post-surgery.

Discussion

The precise incidence of GP remains unknown because of its rarity [1]. In a study by Wang et al. in 2016, involving 8 patients with GP, the median age at diagnosis was 20 years (range 15–25 years) [7]. In Liang et al.'s study of 21 patients with GP in 2015, ages ranged from 5 to 42 years (median 19 years) [1]. In both studies, the most frequently associated primary ovarian tumor was immature teratoma, followed by mature teratoma, with the former study revealing a mean tumor size of around 20.4 cm (range 11-30 cm) [1,7].

The exact mechanism regarding the origin of these glial deposits is not well understood. Some authors consider it to have an autonomous origin from the Müllerian stem cells, and another possible mechanism could be due to the rupture of the capsule of the ovarian teratoma or angiolymphatic spread of the tumor [8,9]. Patients with GP generally exhibit symptoms associated with the underlying ovarian teratoma, including abdominal pain, bloating, or a noticeable mass. The glial im-

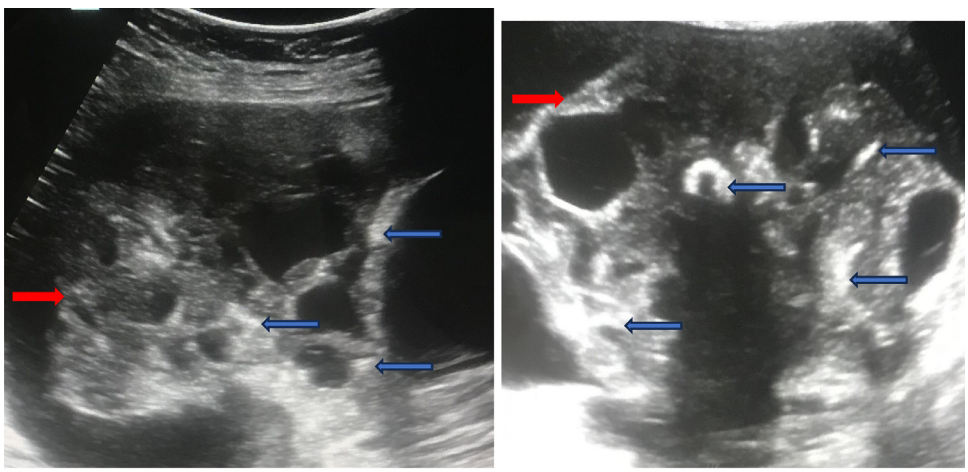

Fig. 1 – (A and B) Ultrasound of abdomen and pelvis showing ascites and heteroechoic solid-cystic abdominopelvic mass (red arrow) with internal echogenic foci and calcifications (blue arrow).

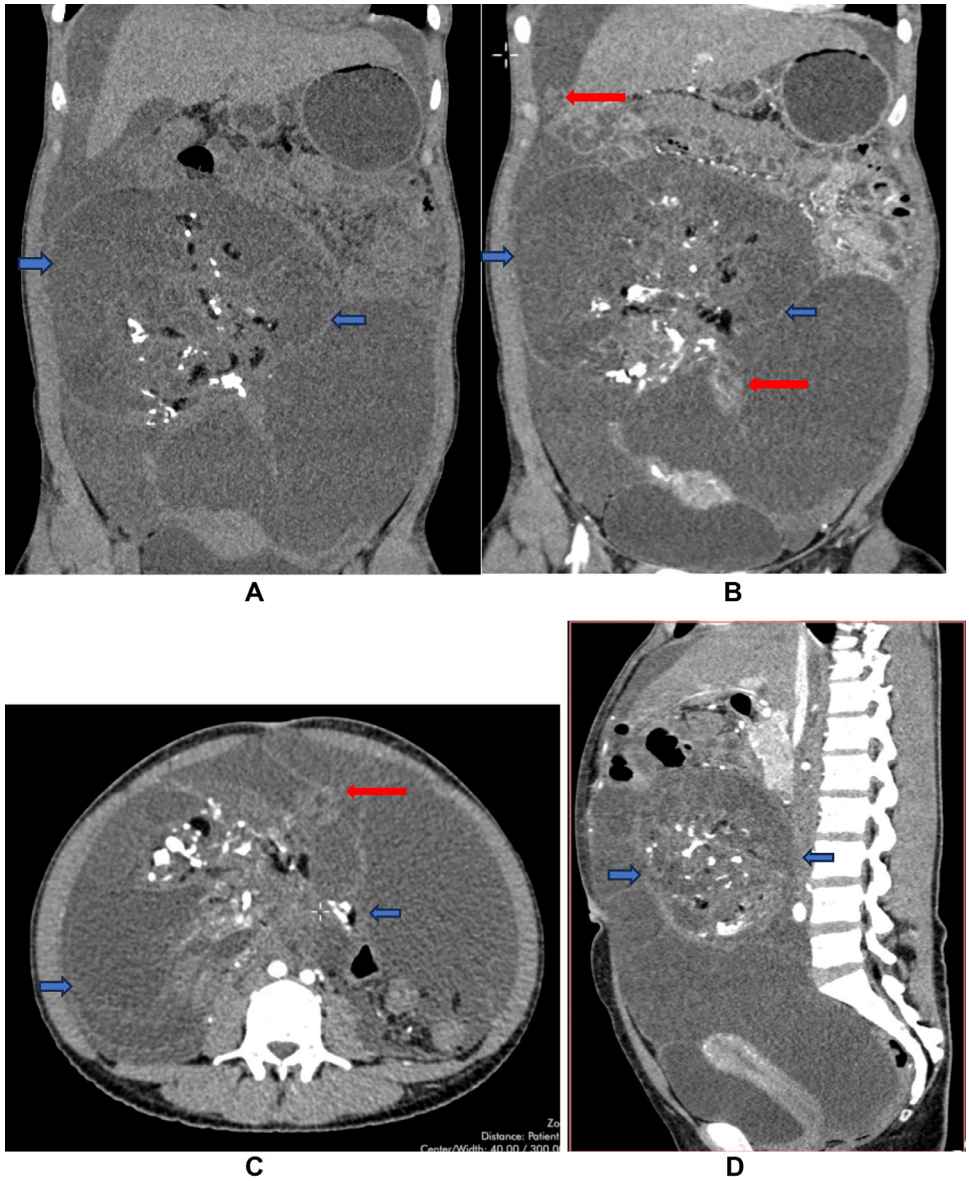

Fig. 2 – (A) Abdomen and pelvic non contrast computed tomography (CT) scan coronal view showing large well defined pelviabdominal solid cystic lesion (blue arrow) predominantly towards the right side with calcifications and fat components. Note the loculated gross ascites. (B) Abdomen and pelvic CE-CT scan coronal view showing the large well defined solid cystic lesion (blue arrow) in right side of abdomen and pelvis. The solid components and septa show enhancement. Note the irregular nodular peritoneal enhancement (gliomatosis peritonei, red arrow) and ascites. (C) Abdomen and pelvic CE-CT scan axial view showing the teratoma (blue arrow) with adjacent enhancing peritoneal nodules (red arrow) and ascites. (D) Abdomen and pelvic CE-CT scan sagittal view showing the teratoma (blue arrow) and loculated gross ascites.

plants themselves are usually asymptomatic and discovered incidentally during surgery or imaging [10].

Computed tomography (CT) scan is the modality of choice to evaluate the dermoid cyst as well as gliomatosis peritonei. Typical imaging findings in mature ovarian dermoid include cystic lesions with fat and calcific components, mural nodule (Rokitansky nodule), fat-fluid levels, and tuft of hair [11]. Larger size (>10 cm) and the presence of enhancing solid components lead to the suspicion of immature teratoma, as in our case [11]. However, mature teratoma can also have some of these enhancing solid components [12]. Magnetic resonance imaging (MRI) is usually performed for difficult cases as it is more fat-sensitive.

GP appears as multiple enhancing nodules in the peritoneum and omentum. The size of the nodules ranges from 0.3 to 1.2 cm in diameter [13,14]. Surgery is the usual modality of treatment for large teratomas with gliomatosis peritonei [15,16]. Adjunctive chemotherapy is given for the immature

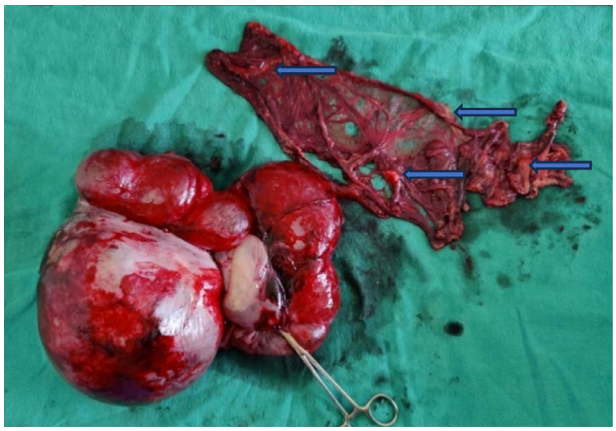

Fig. 3 – Postsurgical specimen of excised right ovary with teratoma, fallopian tube, omentum, and adjacent peritoneum. Note the nodularities within the peritoneum and omentum (gliomatosis peritonei, blue arrow).

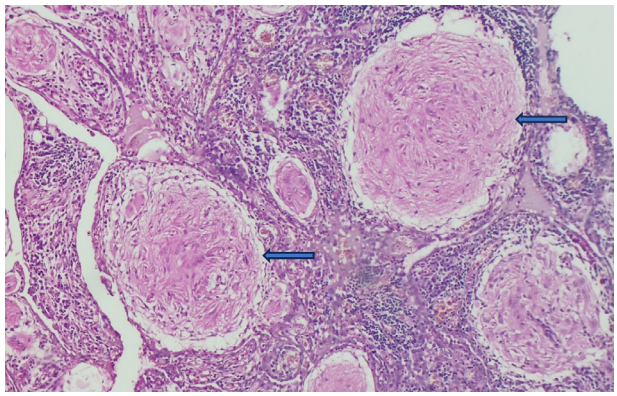

Fig. 4 – Microscopic examination (Hematoxylin and Eosin [H&E] stain, 400 times magnification) of the section examined from the peritoneum shows nodules of mature glial tissue (consistent with gliomatosis peritonei, blue arrow).

teratoma [15,16]. The prognosis for GP is good when associated with mature teratoma. However, long-term follow-up is indicated as GP may progress to malignant transformation on rare occasions [17].

Few similar cases are reported in the literature. Lin et al. described a 54-year-old female with a large ovarian mass, peritoneal seeding, ascites, and enlarged para-aortic lymph nodes [13]. Similarly, Bajracharya et al. reported a 21-year-old female with a large abdominopelvic heterogeneous complex cystic solid mass and nodular omental thickening [18]. In both cases, histopathological examination revealed a mature teratoma with peritoneal nodules displaying glial tissue consistent with GP.

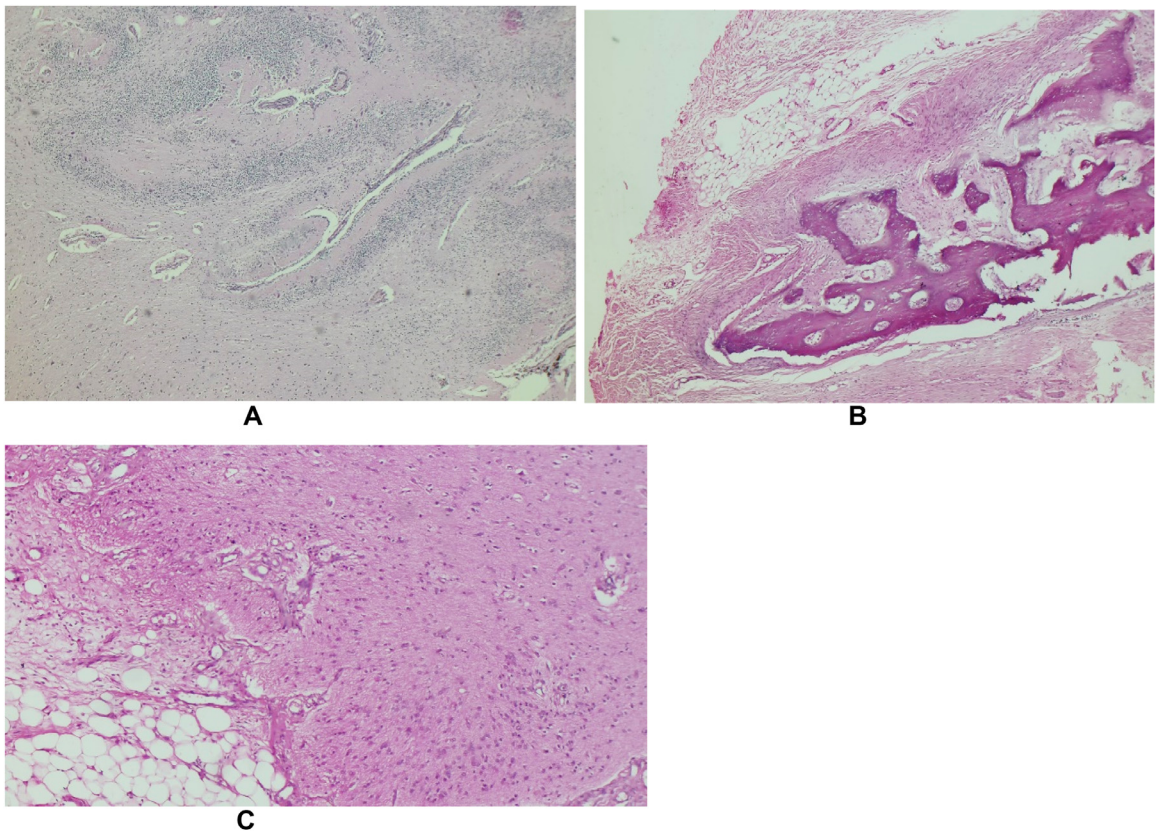

Fig. 5 – (A-C) Microscopic examination (H&E stain) of the sections examined from the intervening stroma of cysts showing mature cerebellar tissue (A), mature bony tissue (B) and mature glial and adipose tissue (C).

Conclusion

This was a rare case of mature teratoma with GP in a young female. Irregular nodular peritoneal enhancement is not always indicative of peritoneal carcinomatosis, and one should always include GP in the provisional diagnosis, especially when associated with teratoma. Enhancing peritoneal nodularities with gross ascites may sometimes indicate tubercular pathology, particularly in endemic regions. However, a proper radiological study combined with histopathological examination in suspicious cases is critical in reaching the appropriate diagnosis.

Patient consent

Written informed consent was obtained from the patients for publication of this case report and any accompanying images.

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