



Case report

Calcified gliomatosis peritonei in a post-menopausal woman with mature ovarian teratoma

Nur Asma Sapiai, Wan Aireene Wan Ahmed, S Thadchaiyani Saminathan

*Department of Radiology, School of Medical Sciences, Universiti Sains Malaysia, Kelantan, Malaysia
Hospital Universiti Sains Malaysia, Health Campus, Kelantan Malaysia*

ARTICLE INFO

Article history

Received: November 6, 2020

Accepted: July 27, 2022

Available online: September 20, 2022

Keywords

Ovarian teratoma

Post-menopausal

Gliomatosis peritonei

Doi

<https://doi.org/10.29089/paom/152443>

User license

This work is licensed under a Creative Commons Attribution – NonCommercial – NoDerivatives 4.0 International License.



ABSTRACT

Introduction: Gliomatosis peritonei (GP) is characterized by the presence of benign, mature glial implants in the peritoneum or omentum.

Aim: Understanding unusual findings with clear knowledge of pathological comparisons makes diagnosis more reliable and guides future treatment.

Case study: This is a case of a 70-years-old lady, post-menopausal, who presented with abdominal mass which progressively increased in size for the past 40 years. She also had constipation requiring laxatives and occasional lower abdominal pain. Her abdomen was clinically distended with a large hard central mass.

Results and discussion: Computed tomography of the abdomen showed features of left ovarian teratoma with multiple gliomatosis peritonei. Histopathology of the mass post-surgery revealed a left ovarian mature cystic teratoma and the peritoneal deposits to be heavily calcified and fibrotic mature implants with no evidence of malignancy.

Conclusions: Gliomatosis peritonei (GP) is a rare finding associated with ovarian teratoma and rare in postmenopausal women. It is important to appreciate a variety of radiological presentations of this condition to establish an accurate diagnosis.

1. INTRODUCTION

Gliomatosis peritonei (GP) is characterized by the presence mature glial implants in the peritoneum or omentum which is of benign. It is rare with only 100 cases reported so far.¹ It is commonly associated with immature ovarian teratoma but rarely in mature ovarian teratoma.^{1,2} It predominantly occurs in women of younger age groups and rare in the post-menopausal age group.³ It occurs more frequently on the right side in unilateral cases (72.2% of cases).⁴ Some of the main challenges in diagnosing gliomatosis peritonei lies in the lack of radiological experience due to the rarity of the case and the presence of variable radiological appearances. Understanding unusual findings with clear knowledge of pathological comparisons makes diagnosis more reliable and guides future treatment.

2. CASE STUDY

Here in we present a case of a 70-years-old lady, Para 3 who had uncomplicated spontaneous vaginal deliveries in the past. Her last childbirth was 33 years ago. She attained menopause at the age of 50. She is not sexually active since the death of her husband 10 years ago. She denied being on any hormonal or traditional medication. No family history of ovarian cancer. Her eldest daughter was diagnosed with breast cancer and is currently on treatment. The patient has underlying bronchial asthma, the last attack was in 2017. She denied other medical illnesses. The patient claims to have a palpable, mobile mass of a tennis ball size at the left lower abdomen since the age of 37 years old. However, she denied being informed regarding any ovarian mass during her last two pregnancies. Her menses were regular. Since the mass was not increasing in size and she was asymptomatic, the patient did not seek any medical attention.

Recently, she developed constipation, requiring laxatives and occasional lower abdominal pain for the past 3 months. She also noticed that the left-sided abdominal mass was gradually increasing in size. She presented to the hospital and examination was carried out by the gynecology team. Abdominal examination revealed a huge mass measuring approximately 20 × 20 cm, which was solid and mobile with a smooth margin. A transvaginal scan showed a huge, echogenic, solid mass, likely left ovarian in origin. Per rectal examination revealed no abnormalities. Clinically, there were no palpable lymph nodes or breast lumps. Her vital signs were normal. Tumour markers including alpha-feto-protein, carcinoembryonic antigen (CEA), and cancer antigen 125 (Ca 125) were not raised. Other blood investigations including full blood count, renal profile, and liver function test were within normal limits. Pap smear was negative for malignancy. Colonoscopy showed no abnormalities (done because of chronic constipation).

Radiological investigations include a transabdominal ultrasound which revealed a large, echogenic, solid-cystic mass with a regular outline occupying the central abdomen.

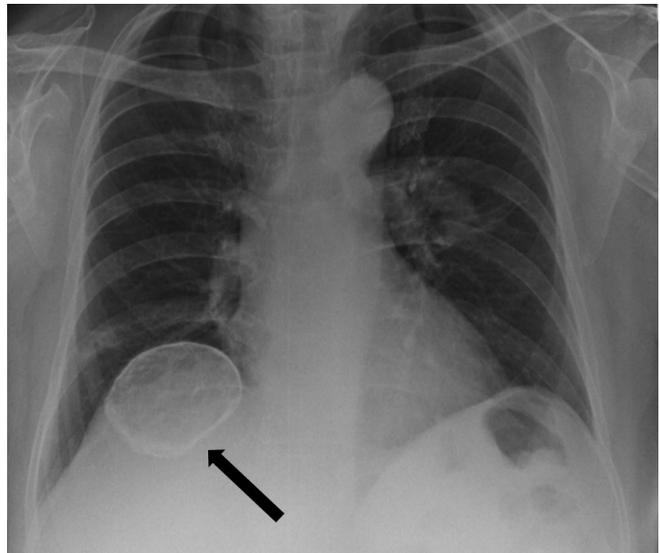


Figure 1. Plain chest radiograph in frontal view demonstrating a well-defined, oval-shaped lesion with the calcified wall (arrow) below the right diaphragm.

The uterus appeared atrophic with thin endometrium. Minimal ascites was found. Plain chest radiograph (Figure 1) demonstrating a well-defined, oval-shaped lesion with calcified wall below the right diaphragm. A plain abdominal radiograph (Figure 2) shows a large, well-defined lesion with calcified wall at left iliac fossa. Similar lesions but smaller in size are seen at the left hypochondriac and left pelvic cavity.

Contrast-enhanced computed tomography (CT) of the abdomen showed a large, well defined, oval-shaped mass measuring 12.3 × 20.9 × 19.3 cm (AP × W × CC) arising

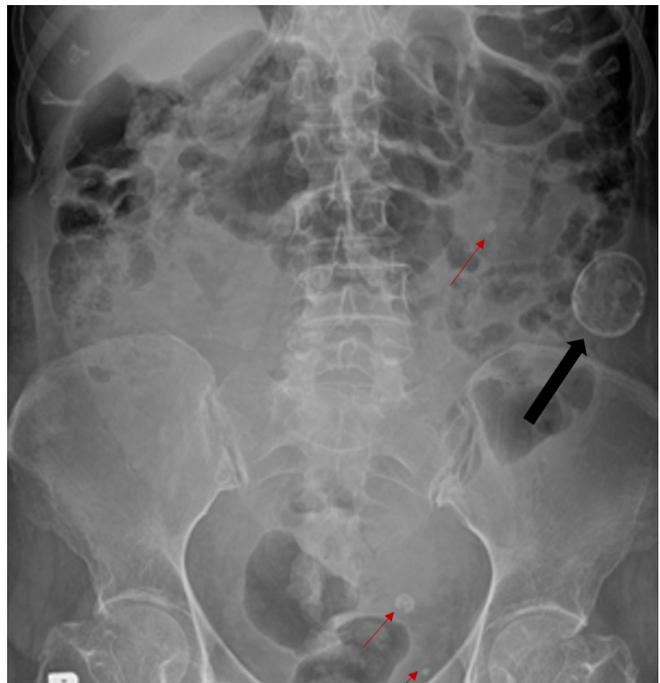


Figure 2. Plain abdominal radiograph shows a large, well-defined lesion with calcified wall at left iliac fossa (black arrow). Similar lesions but smaller in size are seen at the left hypochondriac and left pelvic cavity (red arrows).

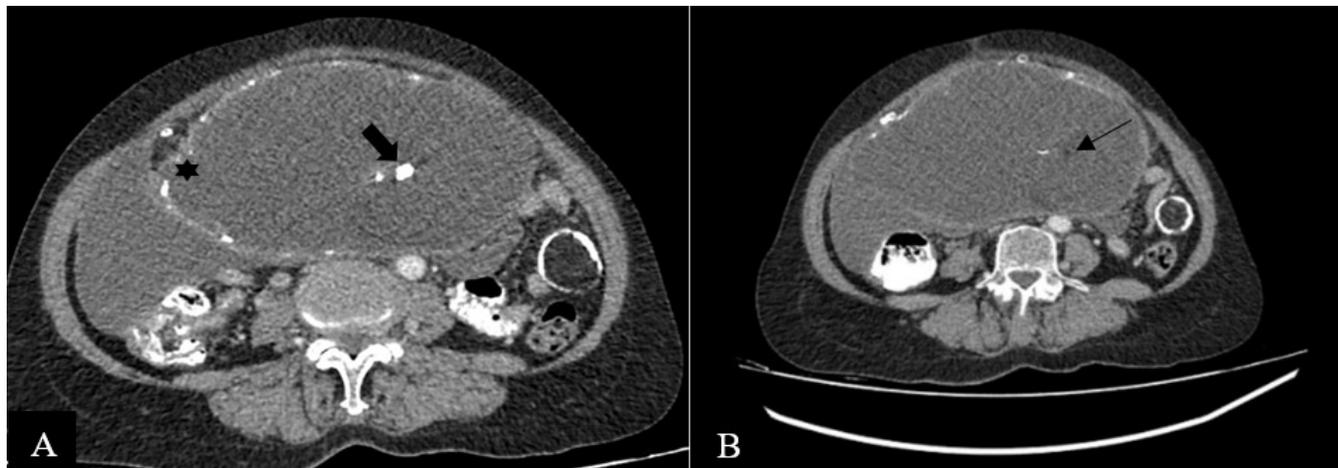


Figure 3. Contrast-enhanced computed tomography in axial view demonstrating a large, well defined, oval-shaped mass arising from the left ovary and occupying the central abdomen. This mass is mainly of cystic composition. Some calcifications (thick arrow) and fat component (thin arrow) is seen within the mass. Its wall is calcified. An area of defect is seen at its right lateral wall suggestive of a wall rupture (star).



Figure 4. Contrast-enhanced computed tomography in coronal view demonstrating multiple well defined rounded lesions with calcified walls at the left paracolic region and pelvic cavity (red arrows). The largest lesion at subdiaphragmatic space is scalloping the superior margin of the liver (black arrow). Calcified lesion within the uterus is a leiomyomata (blue arrow). A rupture at the right lateral wall of the mass is seen, resulting in a leak and formation of the adjacent collection at the right paracolic region (star).

from the left ovary. It occupied the pelvic cavity and extended superiorly till the upper abdomen (L1 level). The mass was mainly of cystic components surrounded by wall calcifications. It also contained fat and soft tissue component. A focal defect in its wall suggested a rupture complicated with adjacent collection (Figure 3). Multiple well defined, rounded lesions with wall calcifications were seen scattered in the abdominal and pelvic cavity. The largest lesion is seen in the right subphrenic region and scalloped the superior liver margin (Figure 4). The CT concluded the presence of left ovarian teratoma with an area of defect likely representing a rupture complicated with the adjacent collection. Multiple peritoneal lesions with the calcified wall are likely peritoneal implants (gliomatosis peritonei). Histopathology revealed that the left ovarian tumour to be a mature cystic teratoma with extensive fibrotic and calcified walls. Omental and peritoneal nodules revealed calcified and fibrotic mature implants with no malignant transformation. Pap smear was negative for malignancy.

Exploratory laparotomy, total abdominal hysterectomy with bilateral oophorectomy and omentectomy, peritoneal fluid cytology, and adhesiolysis were performed. Intra-operative findings include a large left ovarian tumour measuring 15 × 10 cm, twisted ×2 with an area of rupture at its right wall with solidified sebaceous material and hair strands. Multiple firm peritoneal nodules 1–2 cm in size found. The peritoneum was thickened, consistent with reactive peritoneum. Sebum collection was adjacent to tumour. Right ovary and uterus appeared atrophic. Presence of a calcified uterine fibroid. No ascites seen.

The patient was discharged well after surgery. She was scheduled for follow up in the gynecology clinic one-month post-surgery. At the time of this writing, the patient is well with no active complaints.

3. DISCUSSION

Teratoma is the ovaries most common germ cell tumor and represents about 20% of all ovarian neoplasms.⁴ Four types of teratoma in ovaries are recognized. They are types of matures, immature, mono-dermal, and fetiform.^{4,5} The most common of these groupings is mature cystic teratoma (MCT).⁴ Histologically, MCT consists of at least two well-differentiated mature layers of germ cells (ectoderm, mesoderm, endoderm).^{4,5} Mature skin and hair tissues (from the ectoderm) as well as fat and muscle tissues (from the mesoderm) typically make up MCT.⁴ MCTs are usually seen in younger (mean age 30 years) age groups.⁴ In 12% of cases, they are bilateral. MCT occurs more often on the right (72.2%) in unilateral situations.⁴ Well-known threats of MCT are torsion (16% ovarian teratomas), rupture (1%–4%), malignant transformation (1%–2%) and infection (1%).⁴ A preoperative teratoma diagnosis is made from a combination of imagery and clinical findings.¹

Benign, mature glial implants throughout the peritoneum, omentum, or pelvis is known as GP.³ GP is a rare histological entity.⁶ It is typically accompanied by ovarian teratoma which is mature or immature.⁶ In 1906, Neuhäuser coined the term ‘gliomatosis peritonei’ as a disorder of ovarian teratomas with metastatic implantation in the abdominal cavity of mature glial tissue.⁵ GP is characterized as nodular or miliary peritoneal implants composed of mature glia.⁷ Peritoneal implants can transform and disappear into a fibroblastic form, turn into malignant tissue or even survive without any morphological changes.

The pathogenesis of GP development is not well established.^{3,8} There are two hypotheses about the production of GP according to previous studies.¹ One relates to capsular defects of the primary teratoma or dissemination via angio-lymphatic channels.¹ The capsule either had a tear or adhered to the omentum or adnexal structure, as reported in 11 of the 12 cases by Robboy.¹ GP being commonly associated with immature teratoma.^{1,2} The previous study shows adverse prognosis of GP in immature teratomas.³

Radiological findings for MCT have a wide spectrum of radiological presentation. Its ranging from a purely cystic mass to a mixed complex solid cystic mass.^{4,9} Imaging studies, including CT scan, magnetic resonance imaging, and sonography, can provide the diagnosis of teratomas and GP.^{3,6} CT is imaging of choice for the diagnosis of MCT due to identification of fat.^{4,5} While for GP, in imaging, there are varying sizes from miliary spread lesions to big implants.¹ They have predilection to subdiaphragmatic surface, parietocolic gutters and omentum. The findings can be mimicking peritoneal tuberculosis and peritoneal dissemination of malignant tumors.⁶ Imaging analysis of peritoneal dissemination alone cannot aid the differentiation of benign implants from diffuse peritoneal malignant seeding.⁹ Therefore, in patients with underlying ovarian teratomas and imaging shows evidence of peritoneal implants, GP should be considered as a possible diagnosis.⁸ The patient also can has nodal gliomatosis although its rare.¹

Complete GP resection is usually challenging, especially when extensive. Fortunately, GP residual peritoneal illness can be asymptomatic and quiescent over a long period.¹ Residual peritoneal implants can therefore be ignored, and the primary ovarian teratoma should be the focus of treatment. Long-term follow-up is therefore recommended for residual disease patients.⁷ Due to proven cases of malignant transformation of the glial components, it is highly recommended long after initial surgery.^{1,2} There is, however, no consensus on the length of follow-up. CT and ultrasound are modalities of choice suggested for disease monitoring.⁵ If peritoneal implants are well sampled, and proof of mature cells is seen histologically, a favorable clinical course can be predicted. Considering prior cases, a favorable prognosis can be expected.

Good outcomes were seen in mature teratomas and GP in previous records. Therefore, we are expecting a good prognosis for our patient too. Since our patient was a postmenopausal woman, she agreed for the removal of both her uterus and adnexa. Unilateral salpingo-oophorectomy in young women who wish to maintain fertility is recommended for mature or immature teratomas.³

4. CONCLUSIONS

- (1) GP is a rare finding associated with ovarian teratoma, more commonly in immature teratoma, and rarely in mature teratoma.
- (2) It affects young women and is rare in postmenopausal women.
- (3) It is vital to appreciate a variety of radiological presentations of this condition to establish an accurate diagnosis.
- (4) Follow-up is strongly recommended long after initial surgery because of proven cases of malignant transformation as residual peritoneal illness can be asymptomatic and quiescent over a long period.

Conflict of interest

No conflict of interest has been disclosed by the authors.

Funding

This study did not received any special funding.

Acknowledgements

This manuscript was produced without any financial support from any organization. Many thanks to co-authors for contribution making this manuscript successfully produced.

References

- ¹ Wang D, Jia C-w, Feng R-e, Shi H-h, Sun JJoor. Gliomatosis peritonei: a series of eight cases and review of the literature. *J Ovarian Res.* 2016;9(1):45: 1–7. <https://doi.org/10.1186/s13048-016-0256-5>.

- ² Das CJ, Sharma R, Thulkar S, Mukhopadhyay S, Deka D, Mannan R. Mature ovarian teratoma with gliomatosis peritonei-A case report. *Indian J Cancer*. 2005;42(3):165–167. <https://doi.org/10.4103/0019-509x.17064>.
- ³ Lin C-S, Huang C, Li P-C, Hsu Y-H, Ding D-CJT-CMJ. Huge ovarian mature cystic teratoma with gliomatosis peritonei and massive ascites in a postmenopausal woman. *Tzu Chi Medical Journal*. 2019;31(4):289–291. https://doi.org/10.4103%2Ftcmj.tcmj_176_18.
- ⁴ Sahin H, Abdullazade S, Sancı MJ. Mature cystic teratoma of the ovary: A cutting edge overview on imaging features. *Insights Imaging*. 2017;8(2):227–241. <https://doi.org/10.1007/s13244-016-0539-9>.
- ⁵ Outwater EK, Siegelman ES, Hunt JL. Ovarian teratomas: tumor types and imaging characteristics. *Radiographics*. 2001;21(2):475–490. <https://doi.org/10.1148/radiograph.ics.21.2.g01mr09475>.
- ⁶ Lavoie J-M, Lacroix-Poisson F, Hoang LN, Wilson DC, Seck MJ, Tinker AV. 18F FDG positron-emission tomography findings of gliomatosis peritonei: A case report and review of the literature. *Gynecol Oncol Rep*. 2017;20:105–107. <https://doi.org/10.1016%2Fj.gore.2017.03.018>.
- ⁷ Menéndez-Sánchez P, Villarejo-Campos P, Padilla-Valverde D, Murillo-Lázaro C, Martín-Fernández J. Gliomatosis peritonei: recurrence, treatment and surveillance. *Cir Cir*. 2011;79(3):256–259.
- ⁸ Wang J, Xu J, Zhang M, Li B. Gliomatosis peritonei with bilateral ovarian teratomas: A report of two cases. *Oncology Letters*. 2016;12(3):2078–2080. <https://doi.org/10.3892/ol.2016.4866>.
- ⁹ Bajracharya A, Shrestha S, Singh M, Dhakal HP. Mature ovarian teratoma with gliomatosis peritonei: A rare case report. *Clin Case Rep*. 2021;9(9):e04879:1–5. <https://doi.org/10.1002/ccr3.4879>