

Pouch of Douglas Liposarcoma—A Rarity

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Abstract

Introduction: This case report is one of a fairly common tumour in an extremely uncommon anatomic location. Statistically liposarcoma is the commonest type of soft issue malignancy, but publications of such a tumour arising from the pouch of Douglas (POD) to involve the uterus, are very few and far between. **Case details:** A 52-year-old woman presented with a mass in the lower abdomen, post-menopausal vaginal bleeding, and lower abdominal discomfort. Investigations revealed a large pelvic tumour that was attached to the posterior wall of a bulky uterus. There was no evidence of dissemination of the tumour to distant sites, and a laparotomy was performed. A massive soft tissue tumour occupied the POD. The tumour was dissected out from the surrounding structures, and the uterus and its appendages were removed in toto. The histopathological examination revealed a liposarcoma of the pleomorphic type which was arising from the pouch of Douglas (POD), and was attached to the posterior wall of the uterus. Extensive leiomyomatous changes were seen in the uterus. Immunohistochemistry confirmed the liposarcoma to be of pleomorphic type. **Conclusion:** This case report is being published for its rarity and to illuminate the specific issues in the treatment of this ubiquitous tumour in an unusual site. The involvement of a Multidisciplinary Team (MDT) helps to choose the optimal combination of cytoreductive surgery, chemotherapy, and radiation for a given case with a POD malignancy.

Keywords

Pouch of Douglas, Liposarcoma, Multidisciplinary Approach, Cytoreductive

1. Case Report

A 52-year-old woman presented with complaints of severe lower abdominal discomfort, increasing swelling of the lower abdomen, and two episodes of vaginal

bleeding in the prior two months. The patient was thinly built, and abdominal and vaginal examination revealed an obvious swelling in the lower abdomen that arose from the pelvis. Routine reports were normal and an ultrasonogram showed a large pelvic tumour, possibly of ovarian origin. A Contrast Enhanced CT scan (**Figure 1**) showed a large pelvic mass with the consistency of fat which was attached to the posterior wall of the uterus. The ovaries were imaged separately, and found to be normal. There was no regional lymphadenopathy, omental involvement or hepatic secondaries. Tumor markers for ovarian malignancies were normal. As MDT concluded on an exploratory laparotomy.

Through a vertical midline incision, the abdomen and pelvis were thoroughly explored. There were no peritoneal or liver metastases. A large solid mass occupied most of the pelvis and had infiltrated and displaced an enlarged fibroid uterus upwards against the anterior abdominal wall. (**Figure 2**) The ovaries were normal. The operative strategy was to free the uterus from its attachments and to address the uterus and the tumour en bloc. (**Figure 3**)

A classical total abdominal hysterectomy with bilateral salpingo-oophorectomy was performed, and the enormous tumour was removed in toto, in a macroscopic R0 resection. The vaginal vault was closed 2 cm lower than usual.

1.1. Histopathology Findings

Histopathological examination revealed a large leiomyomatous uterus with one area of ulceration. In this area, the liposarcoma from the pouch of Douglas (POD) was infiltrating the uterus. The ovaries, fallopian tubes, and the rest of the uterus were free of disease. Immunohistochemistry confirmed the tumour to be a pleomorphic type of liposarcoma. The tumour showed a combination of lipogenic and non-lipogenic areas. (**Figure 4**) There were distinct regions in the tumour that showed epitheloid features, and in two of the sectioned areas a pericytic pattern was seen. (**Figure 5**) Tumour necrosis and vascular invasion were seen. The epithelial membrane antigen was positive, and CD 68 and CD 34 were also positive. Polycyclic chemotherapy was instituted as per the MDT recommendations and upon review 24 months' post procedure, she remains free of disease and is asymptomatic (**Figure 5**).

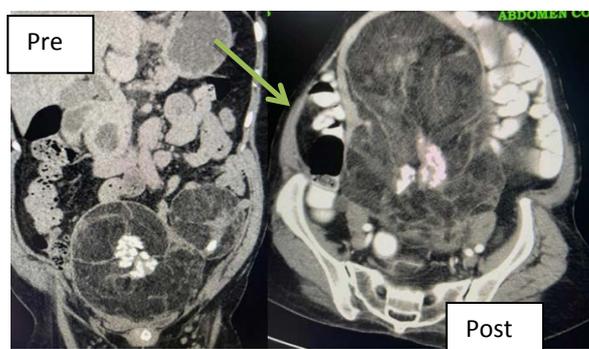


Figure 1. Pre and Post Contrast enhanced CT showing the large pelvic tumor.



Figure 2. Tumor occupying the pelvis completely 1.1. Operative procedure.



Figure 3. Dissection of the tumor along with the uterus.

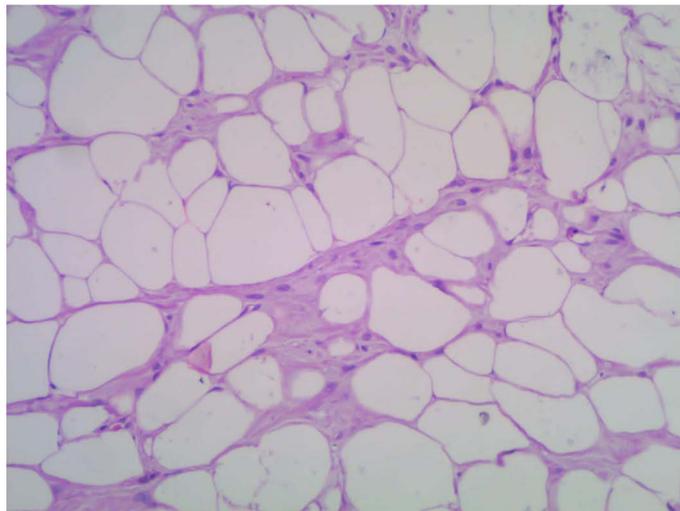


Figure 4. HPE slide at 40 magnification showing lipogenic and non-lipogenic areas.

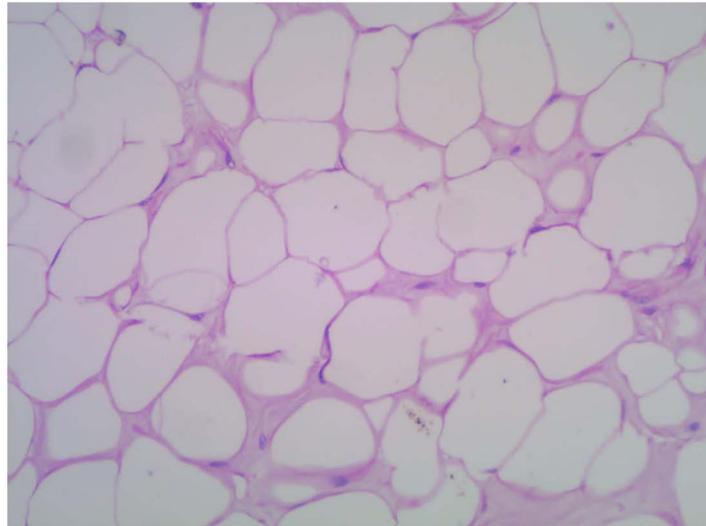


Figure 5. HPE slide at 40 magnification showing epithelioid features and pericytic pattern.

1.2. Follow-up

24 months' follow-up:

- 10 days post-op;
- 2 months post-op;
- 6 months post-op;
- 12 months post-op (Contrast Enhanced CT Abdomen and pelvis);
- 24 months post-op.

2. Discussion

James Douglas, the Scottish anatomist has lent his name to this space.

The Pouch of Douglas (POD) is the posterior cul-de-sac bordered posteriorly by the rectosigmoid, and anteriorly by the uterus. Its peritoneal lining originates from remnants of the Mullerian system. As the embryology is common, both benign and malignant tumours that mimic Mullerian end organs can develop here. Another well-defined mechanism for primary POD malignancies, is the malignant transformation of endometriosis. [1] [2]

Liposarcoma is the commonest soft tissue tumour in the human body. However, it is extremely rare in the pouch of Douglas. Only a few cases of carcinosarcoma in the Pouch of Douglas have been reported so far. [3] [4] Primary malignancies of the POD are exceedingly rare. [5] On extensive search of current English literature by the authors, we identified around 40 cases of primary malignancies of the Pouch of Douglas. The first case was reported by Dockerty *et al.* (1954).

Papillary serous carcinoma, clear cell adenocarcinoma, adenosarcoma and carcinosarcoma are some of the Mullerian types of POD malignancies. Placenta site trophoblastic tumor, extragastrointestinal stromal tumour and malignant mesothelioma are some other tumor types that have been reported. [1] [6] Wong *et al.*, concluded that there was no consensus on the optimal treatment for POD

malignancies.

These lesions are often diagnosed as uterine or ovarian, especially if large, as they occupy most of the pelvis. Even imaging studies often fail to identify them separately. [2] [7]

The presenting symptoms almost always include abdominal pain and distention, and abnormal uterine bleeding. Hence, these patients are often first seen by the gynecologist.

MRI or CECT helps considerably in diagnosing these lesions, as pelvic ultrasound, often done as the first modality, usually only suggests a large pelvic mass [2].

Despite the CT done preoperatively, this was also thought to be a uterine tumour that was occupying the pouch of Douglas. Due to the classic Hounsfield units of the fat content, the liposarcomatous nature of the neoplasm was suggested even on the pre-operative CT scan. However, it was thought to be arising from the uterus.

Histopathology and immuno histochemistry were crucial in clinching the exact diagnosis.

The tumour was well circumscribed but not encapsulated, with infiltrative borders. Clumps of pleomorphic cells were found in more than 70% of the cut surface. There were distinct areas of MFH like, or spindle cell areas, as well as round cells, without a vascular network. The tumour was of a high grade, with enlarged nuclei, with tumour necrosis and more than 25 mitotic figures seen per 10 HPF. There were focal areas of pericytic involvement, and lipoblasts present as well. IHC was positive for vimentin, S 100, CD 34, and smooth-muscle actin, and this tumour was negative for t (12; 16) (q13 p11)—T L S CHOP (more commonly seen in the myxoid variety). Estrogen receptor was negative.

Pleomorphic liposarcoma of the uterus has also been reported in a patient on tamoxifen, but the tumour cells in that particular case were IHC positive for estrogen receptors apart from S 100 [8].

The pathological report suggested, that there was only a tiny area of secondary involvement of the uterus, pointing towards a primary POD malignancy that had secondarily infiltrated the uterus. There was no concurrent endometriosis seen on the histology, a well-known source of a pouch of Douglas neoplasm. In most of the cases, patients undergo cytoreductive surgery, which was considered by the authors to be optimal in this case. In view of the size of the tumour, the MDT recommended concurrent radiation as well as polycyclic chemotherapy and these were instituted.

With an overall 39% survival rate for pleomorphic liposarcoma, and with the grade of this tumour being higher in this case, we are guarded in the prognosis for this patient. However, at the time of publication, about two years post-surgery, she seems to be free of disease, clinically and radiologically.

3. Conclusion

Although liposarcomas are found everywhere, pouch of Douglas liposarcomas

are a rarity. A pleomorphic type of liposarcoma, confirmed by immunohistochemistry, arising from the Pouch of Douglas, has its specific set of challenges, in diagnosis as well as treatment. The involvement of an MDT helps to choose the optimal combination of cytoreductive surgery, chemotherapy, and radiation for a given case with a POD malignancy. This case is being published for its rarity.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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