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# METASTATIC OVARIAN CYSTOSARCOMA PHYLLOIDES OF BREAST

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## ABSTRACT

**Background:** Cystosarcoma Phylloides is a rare breast neoplasm constituting  $\leq 1\%$  of all breast neoplasms. These are mostly benign and recur. Malignant cystosarcoma Phylloides can recur and metastasize to lung, bone and abdominal viscera. Metastasis to Ovary is not reported in literature.

**Case:** A 46 year old multiparous lady was diagnosed with a recurrent cystosarcoma of right breast and a large Ovarian mass which was causing her dyspnoea. The mass was of 30 weeks size and was firm and tender. CECT showed a large solid abdominopelvic mass with irregular enhancing septate extending from pelvis to infracolic area with minimal free fluid. Uterus and Ovaries could not be delineated. FNAC from the mass was reported as low-grade malignant mesenchymal tumour. CA 125 was within normal range. Laparotomy revealed a large fleshy mass with jelly like material which was adherent to intestines and pelvic and parietal peritoneum. Right ovary is not visualized. Left ovary partially visualised and incorporated into the mass. Excision of the mass with TAH and BSO was carried out. There was diffuse ooze from the pelvic and peritoneal cavity which was managed by packing, blood product transfusion and tranexamic acid. She received massive transfusion and survived. Later she developed haemoptysis and underwent tracheostomy and feeding ileostomy and was managed in ICU for 8 weeks. Palliative mastectomy and adjuvant Radiotherapy and chemotherapy were differed by Oncologists and hence she was discharged after 4 months of admission.

**Conclusion:** Managing ovarian metastasis from cystosarcoma phylloids can be challenging and the quality of life is poor when the primary disease is not managed adequately.

**Key Words:** Palliative mastectomy, Phylloid tumours, Ovarian tumour

## INTRODUCTION

Phylloid tumours account for less than 1% of breast masses and they occur rarely. They affect women at young and middle age unlike adenocarcinomas which occur at a later age. They most often recur locally and 20 % to 40% develop distant metastasis. The most common sites of distant metastases are lung, bone and abdominal viscera<sup>1</sup>. The abdominal visceral metastasis is reported in duodenum and Pancreas.<sup>2</sup> Metastasis to ovaries is not found in literature. Hence this rare case is reported.

## CASE

A 46 year old para2 live 2 whose child birth was 14 years back underwent simple mastectomy for a right sided breast mass one month ago at a private hospital, the histopathology was consistent with cystosarcoma phylloids. The breast mass re-appeared at the same site with in one month of surgery.

FNAC from the recurrent breast mass revealed cystosarcoma phylloids with stromal elements. She also developed sudden onset of pain abdomen and distension of abdomen of one month duration and decreased urine output, difficulty in breathing and constipation of 12 days duration. CT scan of abdomen and pelvis reported a large solid mass in abdomen and pelvis with minimal free fluid in pelvis. The mass is not separately seen from uterus and ovaries. She was referred to our Institute with a diagnosis of recurrent cystosarcoma phylloids with Ovarian tumour.

Her Obstetric and gynecological history revealed that she had 2 normal deliveries and underwent tubectomy 14 years back and there was no family history of malignancies. She had polymenorrhoea. She gave history of loss of weight and loss of appetite for the past 6 months. She also had urinary retention and hence she was on continuous bladder drainage for one week. She complained of fever with chills of 10 days duration.

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On examination she was emaciated, tachypnoeic, febrile, pulse was 108/min regular, BP 100/60 mm Hg. No significant lymphadenopathy. There was a hard mass of 10x6x3 cm in size on the lateral aspect of right breast with local rise of temperature and tenderness. Left breast was normal. Respiratory system was normal. Cardiovascular system was normal except for tachycardia. Abdomen was grossly distended in sub-umbilical and umbilical region. There was a hard immobile tender mass of 30 weeks size arising from pelvis. Bowel sounds were normal. General surgical opinion was recurrent or residual cystosarcoma phylloides (as the resected margins were reported positive) with Ovarian tumour and advised to manage the ovarian tumour first.

Her abdominal USG was reported as a large abdominopelvic complex mass with minimal ascites displacing the bowel loops laterally suggestive of Ovarian mass. Her CECT at our Institute after one week of admission reported as a large abdominopelvic dense mass with irregular enhancing septate extending from POD to infra colic areas. Ovaries are not visualized separately. FNAC from the abdominal mass was reported as a low grade malignant mesenchymal tumour. She was taken up for laparotomy 2 weeks after admission after surgical oncologist opinion. On laparotomy there was hemorrhagic ascites of more than 500ml. There was a large fleshy mass with jelly like material with a breach on the surface suggesting tumour rupture. The mass was of 28 week size occupied pelvis and lower abdomen and extended up to the root of the mesentery and covered by small bowels which were closely adherent. There was lot of mucoid material within the mass appearing like myxoid degeneration (Fig 1a). Both ovaries were incorporated in to the mass and right ovary could not be recognised and left ovary was only partially recognized (Fig 1b). Both fallopian tubes were free. Uterus was 10 weeks size and anterior to the mass which was adherent to POD, lateral pelvic walls and rectosigmoid. The mass was separated from the intestines and excised with the help of surgical oncologist. Total abdominal hysterectomy with bilateral salphingo ophorectomy was carried out. There was lot of oozing from the intestinal surfaces and peritoneal surfaces of POD. Multiple haemostatic sutures were taken and bilateral internal iliac arteries were ligated. As the oozing persisted from the peritoneal surfaces, pelvis and abdomen was packed. Blood loss was 1000 ml and she received 5 units of FFP, 4 packed cells. In view of poor general condition she was ventilated and was kept on SIMV mode. Abdominal pack was removed after 48 hours under general anaesthesia. She was monitored in post-operative ward and received 22 units of FFPs, 4 units of platelets, 3 units of Packed cells, 12 units of cryoprecipitate over a period of 8 days. She also received intravenous tranexamic acid during surgery and for 48 hours following surgery. Mastectomy was deferred at the time of laparotomy though it was planned to do earlier by the surgical oncologist.

She could be started on oral fluids after a week and was shifted to ward after 10 days of surgery. On 14 th postoperative day she developed sudden dyspnoea and was managed conservatively with oxygen and sedation. Her fever persisted despite of 3 broad spectrum intravenous antibiotics. She had superficial wound gaping and developed dyspnoea again with decreased saturation. She was kept on mechanical ventilation. X Ray chest P/A revealed minimal left sided effusion with basal atelecasis. She underwent tracheostomy after 4 days as prolonged ventilation was required. She produced thick sputum which required mucolytic and frequent suctioning. After 10 days she was weaned off ventilator and was maintaining 100% saturation. The histopathological report was metastatic malignant cystosarcoma Phylloids to ovary and parametrium. Fallopian tubes and uterus and cervix were free of tumour. Tumour showed high cellularity and moderate nuclear atypia and high mitosis 16/10 high power field with extensive myxoid change. (Fig 2.a,b,c). This was consistent with previously diagnosed and treated malignant phylloids. Her haematological parameters were within normal limits. She was given one course of Ifosphamide and MESNA which she tolerated well. She was given total parenteral nutrition for almost one month. After seven days of receiving chemotherapy she became dyspnoeic and right sided air entry decreased and she was shifted to RICU (Respiratory Intensive care Unit) under care of the anaesthetists. Her abdominal wound healed by secondary intention. She was given ICU care and underwent feeding jejunostomy. She was decanulated after 8 weeks of tracheostomy. The breast mass increased in size 20x10 cms, infected and displaced to right lateral side of chest. Surgical Oncologists deferred in doing any kind of palliative surgery and medical and radiation oncologists in giving chemotherapy and radiation therapy. She was in RICU for 6 weeks and received antibiotics as per the sensitivity of the organisms from wound swab, tracheal swab, infected breast mass swab etc.,. The organisms were Acinetobacter, klebsiella, pseudomonas. The abdomen was scaphoid and there was evidence of fluid or mass. She was asked to take over by Gynaecologists. As there was no gynaecological treatment necessary and the surgical Oncologists deferred in performing palliative surgery for the breast mass, she was explained the inability to give further supportive treatment and discharged home.

## DISCUSSION

The options for treatment of Cystosarcoma phylloids include local excision, wide local excision and mastectomy. In wide local excision the resected margin should be free of tumour for 1 cm. Local recurrence is expected in 15 % of cases even with this modality of treatment<sup>3</sup>.

Cystosarcoma Phylloids are diagnosed to be benign, border-

line and malignant based on histopathological characteristics and a clinical diagnosis of malignant variety is not made as recurrence and even metastasis can occur in benign tumours. A study correlating histopathological features with clinical presentation in 187 cases, found local recurrence in 27%, 32% and 26% of benign, borderline and malignant tumours respectively. Metastasis was present in two borderline and six malignant tumors out of 100 (8%). There were no specific histological features that correlated with local recurrence and metastasis but cytological atypia of stromal cells, stromal overgrowth and mitotic figures of >15 per 50 high power fields were present in those who showed metastasis.<sup>4</sup> Flow cytometric analysis of s fractions greater than 0.05 was found to be a useful predictor of clinical outcome along with histological features of stromal overgrowth and infiltrating margins<sup>5</sup>. A literature review in 1999 to find out the predictors of recurrence after conservative surgery of cystosarcoma phylloides concluded that wide local excision is also a suboptimal modality of treatment for borderline and malignant phylloid tumours because the recurrence rate is high (29% for borderline and 36% for malignant).<sup>6</sup>

The most common site of metastasis is lung<sup>7</sup> and other sites reported are spine, brain, parotid gland, nasal cavity, forearm and mandible. The metastatic sites in the abdomen reported are pancreas, duodenum, jejunum and liver. Metastasis to genital organs is very rare and only one case report of metastasis to vulva<sup>8</sup> and another to Brenner tumour of ovary is found in literature<sup>9</sup>. The metastasis to vulva occurred along with pulmonary metastasis a year after the management of primary by surgery and local radiation. PET-CT as the metastatic nodule was only 2x2cm and the diagnosis was confirmed by fine needle aspiration<sup>8</sup>. The metastasis to ovary could not be diagnosed prior to laparotomy by fine needle aspiration in the present case. This is because FNAC has very high false negative rates in diagnosing cystosarcoma phylloides.<sup>10</sup>

Adjuvant therapy for management of metastatic Phylloides includes chemotherapy and Radiotherapy. Response to chemotherapy was observed in lung metastasis and abdominal metastasis but not in bone metastasis. Single agent and combined regimens have been used and response is long lasting with increase in progression free survival when Ifosfamide is used<sup>11</sup>. Radiotherapy has a role for loco regional control in recurrent benign as well as malignant Phylloides. However the control of primary tumour is important when metastasis had been taken care of to improve the survival rates as well as the quality of life. The present case though she survived after laparotomy and complete removal of the large ovarian metastasis, she continued to suffer as the primary was not taken care of. Palliative surgery, chemotherapy and radiotherapy were deferred in the present case even after repeated discussions saying there is no role for surgery

of primary in metastatic disease. But in this case the ovarian metastasis was taken care of almost completely by surgery. Aggressive palliative surgery in metastatic Phylloides is reported to improve survival as well as quality of life. The physical and mental well being was improved after radical surgery after for repeated recurrence that occurred twice and this improved the nutritional status and immunity to undergo further treatment with chemotherapy<sup>7</sup>. In the present case, she was unable to get up from the bed because of the weight of the tumour mass (breast) that progressed to large size and also because of poor nutritional status. Palliative breast surgery and palliative Radiotherapy under high risk consent may have improved her quality of life. A recent study which assessed the predictive factors for the local recurrence and distant metastasis of phylloides tumours of the breast in 192 cases concluded that histopathological type and margin status were independent predictors of distant metastasis-free survival and overall survival and it is essential to reduce the local recurrence to prevent distant metastasis.<sup>12</sup>

## CONCLUSION

Managing ovarian metastasis from cystosarcoma phylloides can be challenging and the quality of life is poor when the primary disease is not managed adequately. When an abdominal mass and a breast mass co-exist, metastasis from the breast mass to be considered as the first etiology rather than an association of different pathology until proved otherwise.

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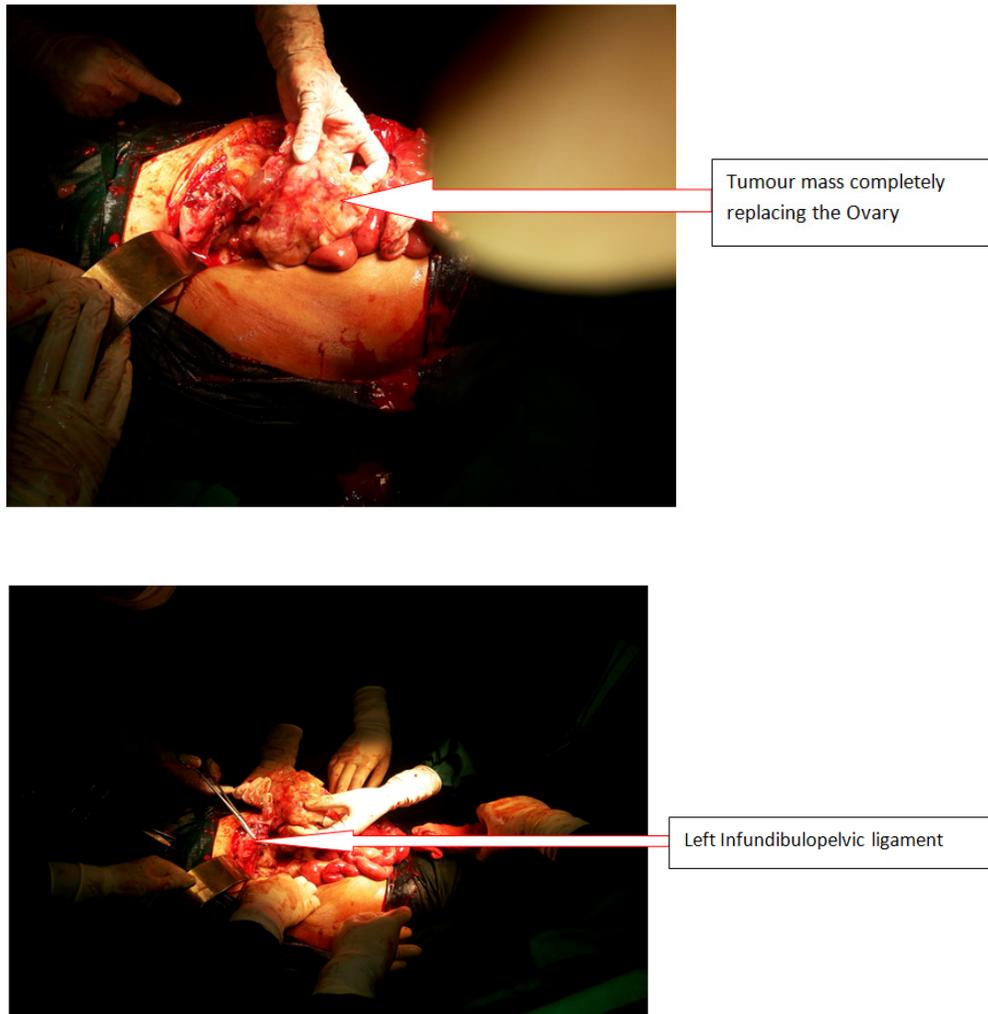
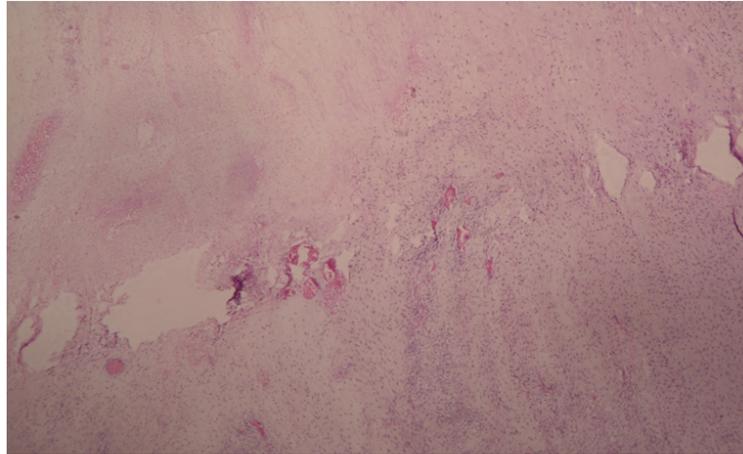
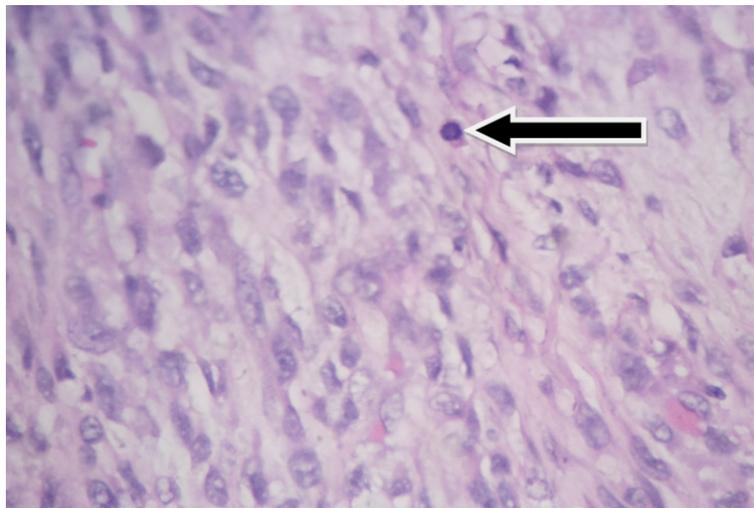


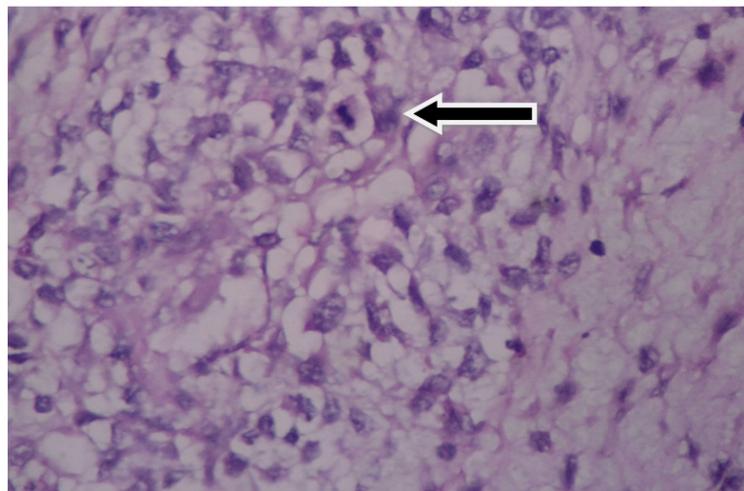
Figure 1 (A, B): Laparotomy findings



A. Hematoxylin & eosin stained sections (100X magnification): Viable tumor cells arranged in fascicles (right) Tumor with large areas of necrosis (left)



B. Hematoxylin & eosin stained sections (400X magnification): Pleomorphic spindle cells with ring mitosis (arrow)



C. Hematoxylin & eosin stained sections (400X magnification): Tumor cells in the background of myxoid material and bipolar mitotic figure (arrow)

**Figure 2 (A–C):** Histopathological Features- Metastatic Cystosarcoma Phylloides