DOI: 10.7860/JCDR/2022/56624.16462

Case Series

Pathology Section

# Uncommon Pathological Presentations of Mature Cystic Teratoma: A Case Series

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## ABSTRACT

Mature cystic teratomas are benign tumours of the ovary but rarely certain unusual gross and microscopic findings might complicate their diagnosis or may have an impact on their clinical outcome. Here, we are presenting a case series of five unusual pathological presentations of mature cystic teratomas of ovary. Out of five cases, three cases were of collision tumours, in which combination of mature cystic teratoma is seen with mucinous cystadenoma in two cases, and with serous cystadenoma in one case. True collision tumours are defined as histologically distinct neoplasms in the same tissue or organ without any histologic admixture or intermediate cell population zone between two components. These tumours are located in various organs but ovarian location is rare. Mature cystic teratoma is the most common component of collision combinations in the ovary. All classes of ovarian tumours, benign, borderline and malignant may collide and therefore, clinical outcomes in collision tumours depend on individual tumour characteristics. The fourth case was of a mature cystic teratoma associated with a distinct haemangiomatous component. Mature cystic teratoma associated with prominent haemangiomatous component is a very rare finding with only few case reports in the literature and it should be differentiated from true ovarian haemangioma, lymphangioma, and angiosarcoma. The fifth case was of bilateral mature cystic teratomas associated with a well-defined lipomatous lesion in right sided mature cystic teratoma mimicking lipoma. Lipomatous ovarian lesions are very rare and mature cystic teratoma with prominent adipocytic proliferation is also an exceedingly uncommon finding and it should be differentiated from true ovarian lipoma and other atypical lipomatous proliferations. Most of these tumours remain clinically and radiologically unrecognised, therefore their histological recognition is essential for adequate patient management.


**Keywords:** Collision, Cystadenoma, Dermoid cyst, Haemangiomatous, Lipomatous

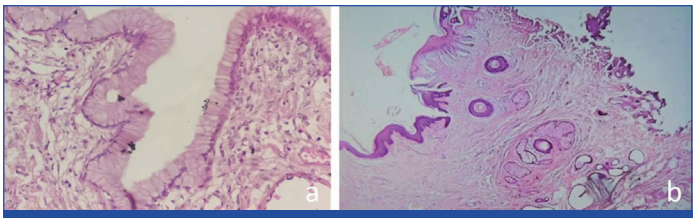
## INTRODUCTION

Mature cystic teratomas are the most common ovarian germ cell neoplasms and constitute 10-20% of all ovarian neoplasms [1]. These neoplasms can rarely have unusual gross and microscopic presentations which interferes with their clinical and radiological interpretation and histopathological examination becomes crucial for their correct diagnosis. The cases included in this study presented in the Department of Obstetrics and Gynaecology and were reported in the Department of Pathology, Government Medical College, Haldwani, Uttarakhand, India. Collision tumours are rare neoplasms which are reported in various organs but ovarian location is rare. Mature cystic teratoma is the most common component of collision combinations in the ovary. Three cases of collision tumours are included in this series and all these cases had mature cystic teratoma as one of the colliding components. Another case was of a mature cystic teratoma with florid vascular proliferation, which is an exceedingly rare entity with only a few reported cases in the literature. Lipomatous ovarian lesions are very rare and mature cystic teratoma with prominent adipocytic proliferation is also an exceedingly uncommon finding and it should be differentiated from true ovarian lipoma and other atypical lipomatous proliferations. One case of an adipocyte rich teratoma is included in this series to highlight this rare presentation.

## CASE SERIES

### Case 1

First case was of a 24-year-old female, unmarried, who presented with complaints of abdominal pain since five months. Physical examination revealed right sided adnexal mass. Her medical history was non contributory. The Ultrasonography (USG) findings revealed a cystic multilocular mass originating from the right ovary. Laparoscopic cyst removal was done with a provisional diagnosis of right-sided mucinous cystadenoma. Gross examination showed cystectomy specimen measuring 6.0×5.0×2.0 cm. Wall thickness was 0.1-0.2 cm. External surface was smooth. Cut surface showed multiloculated cyst, filled with mucinous material. Another area in the cyst showed pultaceous material and tufts of hair. Differential diagnosis considered were mature cystic teratoma, collision tumour and composite tumour, based on gross examination. On microscopy, sections from multiloculated cyst revealed fibrocollagenous cyst wall lined by gastric foveolar type of epithelium. Sections from another area depicted keratinised stratified squamous epithelium with underlying pilosebaceous units and adipose tissue. These two components were discrete with no histological admixture, therefore a final diagnosis of collision tumour comprising of mature cystic teratoma and mucinous cystadenoma was made [Table/Fig-1a,b].

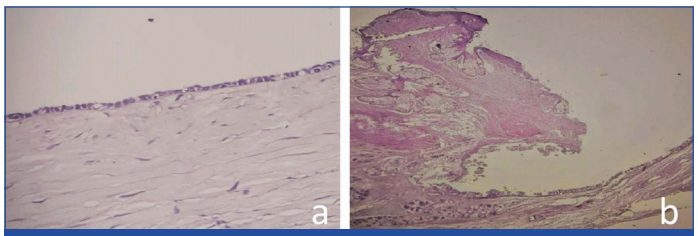



**[Table/Fig-1]:** a) H&E stain; (400X) Mucinous cystadenoma component of a collision tumour lined by gastric foveolar type of epithelium; b) H&E (40X) Teratomatous component of the same tumour depicting epidermis and pilosebaceous units.

### Case 2

Second case was of a 43-year-old female patient, who presented with an abdominal lump and menstrual irregularity for the past 8 months. She was Para 2, Living 2 (P2L2) and her past medical history was unremarkable. Physical examination revealed left side adnexal mass. Magnetic Resonance Imaging (MRI) findings suggested left-sided cystic ovarian mass. Her Cancer Antigen (CA) 125 level was 33 IU/mL. Provisional diagnosis of left sided dermoid cyst was made. Laparotomy was done with left-sided salpingo-oophorectomy. Gross examination showed salpingo-oophorectomy specimen measuring 18.0×15.0×13.0 cm with attached fallopian tube measuring 4.0×0.8 cm. External surface was smooth. On cut,

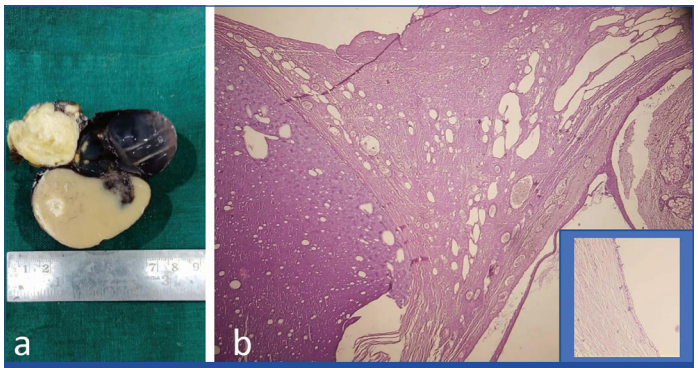
a loculated cyst was identified, filled with the clear serous fluid. A smaller cyst was present within cystic cavity measuring 2 cm in maximum dimension and filled with pultaceous material and hair. Representative sections were taken. On microscopy, sections from the larger cyst revealed fibrocollagenous cyst wall lined by low cuboidal ciliated epithelium. Sections from the smaller cyst showed keratinised stratified squamous epithelium with underlying tissue displaying sebaceous glands, respiratory type epithelium and collections of seromucinous glands. These two components were discrete with no histological admixture. Based on these histological findings, a diagnosis of collision tumour comprising of dermoid cyst and serous cystadenoma was established [Table/Fig-2a,b].




**[Table/Fig-2]:** a) Serous cystadenoma component of a collision tumour shows cyst wall lined by cuboidal to low columnar ciliated epithelium (H&E stain; 400X); b) Teratomatous component of collision tumour showing epidermis, sebaceous glands, seromucinous glands and respiratory type epithelium (H&E stain; 40X).

## Case 3

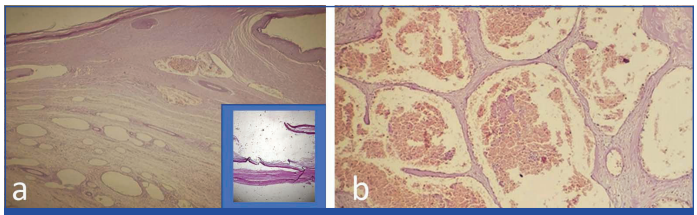
A 41-year-old female, presented with complaints of lower abdominal pain, heaviness and menstrual irregularity since 9 months. She was P1L1 and her past medical history was non contributory. Radiological investigation had suggested the diagnosis of fibroid uterus with a cystic, multilocular, and non vascularised formation in the right ovary. Total Abdominal Hysterectomy (TAH) with bilateral salpingo-oophorectomy was done. Right sided ovarian cyst measured 9.0×7.0×3.0 cm. External surface was lobulated. Cut surface revealed loculated cyst filled with mucinous material. One cystic cavity was filled with pultaceous material and hair. Multiple sections taken from cystic component depicted features of benign mucinous cystadenoma, whereas sections from solid pultaceous component exhibited features of mature cystic teratoma. Thus, establishing the diagnosis of collision tumour, comprising of mucinous cystadenoma and mature cystic teratoma [Table/Fig-3a,b].




**[Table/Fig-3]:** a) Gross photograph showing a multiloculated cyst with pultaceous material in one of the cysts; b) Photomicrograph of the same lesion as in Fig 3a. One of the cystic cavity is showing dermoid elements, adjacent cyst depicting luminal mucinous material and third cystic cavity is lined by low columnar to flattened non ciliated epithelium. Inset showing lining epithelium of cystadenoma component (H&E stain; 40x). Inset (b): (400X).

## Case 4

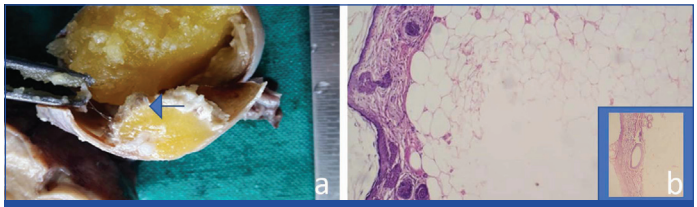
Fourth case was of a 37-year-old female patient, who presented with off and on lower abdominal pain since one year. Her past medical history and laboratory investigations were unremarkable. Physical examination revealed a right sided adnexal mass. The USG revealed right sided ovarian semicystic mass measuring 9.6×6.0×3.0 cms. There was no sign of ascites on USG examination. Her CA-125 level was 30 IU/L. Laparotomy was done with right sided ovarian cystectomy. Gross examination showed cystectomy specimen measuring 9.0×5.0×2.5 cm. External surface was smooth. Cut surface was partly solid and partly cystic. On microscopy, sections studied from cystic area revealed cyst wall lined by stratified squamous epithelium with luminal keratinous flakes. Sections from solid area showed a relatively circumscribed lesion depicting florid proliferation of dilated and congested vascular channels, lined by flattened endothelium, embedded in a fibrocollagenous stromal tissue. On extensive sampling from solid area, focal areas lined by stratified squamous epithelium were also identified. On the basis of these findings a diagnosis of mature cystic teratoma with prominent haemangiomatous component was made [Table/Fig-4a,b].




**[Table/Fig-4]:** a) Stratified squamous epithelium lined area at one focus in a solid haemangiomatous component of a mature cystic teratoma of ovary. Inset showing cystic teratomatous component of the lesion with luminal keratinous flakes (H&E stain; 40x) ; b) Haemangiomatous component of a mature cystic teratoma of ovary, showing closely packed thin walled and dilated vascular channels from same lesion as in Fig. 4a mimicking true ovarian cavernous haemangioma (H&E stain; 100x).

## Case 5

Fifth case was of a 41-year-old female, who presented with abdominal lump, abdominal pain and weakness for the past four months. Her medical history was unremarkable with regular menstrual cycle and no signs of hyperandrogenic state were seen. Physical examination revealed bilateral adnexal mass. The USG revealed solid cystic bilateral adnexal mass. Gross examination showed right sided salpingo-oophorectomy specimen and left sided cystectomy specimen. Right sided ovarian cyst was 5.0×4.0×3.0 cm with attached fallopian tube measuring 4.8×0.7 cm. External surface was smooth. Cut surface showed cystic cavity filled with pultaceous material and hair. At one place a well-defined solid area was identified measuring 2.0×1.8 cm. On cut, it was homogeneous and yellowish in colour. Left cystectomy specimen was 7.4×6.0×4.1 cm. On cut, it was filled with pultaceous material and hair. Residual ovary was identified at one focus. Sections taken from representative areas. On microscopy, sections from bilateral cysts showed features of mature cystic teratoma. Sections taken from the well-defined solid area in right-sided cyst revealed a well circumscribed lesion comprising of lobules of mature adipose tissue, separated by thin fibrous septa and few benign eccrine ducts were seen at the periphery of the lesion. Based on gross and microscopic evaluation a diagnosis of bilateral mature cystic teratoma with a distinct right sided adipocyte rich teratoma was made [Table/Fig-5a,b].




**[Table/Fig-5]:** a) Gross photograph showing a distinct lipomatous lesion in the cyst wall of mature cystic teratoma; b) A distinct well defined lipomatous lesion was seen microscopically from the area pointed in Fig 5a. Inset showing benign eccrine ducts at one focus in the periphery of lipomatous lesion (H&E stain; 100x). Inset b- (H&E stain; 40x).

# DISCUSSION

True collision tumours are defined as histologically distinct neoplasms in the same tissue or organ without any histologic admixture or intermediate cell population zone between two components, whereas there is histological intermixing of different components in composite tumours [2]. These tumours are located in various organs but ovarian location is rare. The biological behaviour of each component in a collision tumour depends on individual tumour characteristics, therefore their recognition is important for further management.

Various combinations have been reported in collision tumours of the ovary such as mature cystic teratoma and mucinous cystadenoma [3], serous cystadenocarcinoma and teratoma [4], carcinosarcoma and dermoid [5], mature cystic teratoma and fibroma [6].

Many theories have been proposed explaining the origin of collision tumour. According to the first proposed theory, coexistence of two primary tumours in the same tissue or organ is due to a "chance accidental meeting" [7]. The second theory states that the first tumour present at the primary site leads to some changes in the microenvironment causing the development of second primary tumour or facilitates the seeding of metastatic tumour cells. The third hypothesis considers the possibility of origin of different components from a common stem cell [8]. Few other hypotheses regarding origin of collision tumour include surface metaplasia and teratomatous origin theory. Ultrastructural and mucin histochemical studies support the surface metaplasia theory [9,10]. Whereas, frequent co-existence of mature cystic teratoma with mucinous cystadenoma forms the basis for teratomatous origin theory. As mucinous element in mature cystic teratoma is more commonly of intestinal type rather than Mullerian, this again supports the teratomatous origin of mucinous tumours. Fuji K et al., had also demonstrated similar genetic pattern in both mucinous and teratomatous components, further attesting to the teratomatous origin theory [11].

Three cases in this series are diagnosed as collision tumours; one case had a combination of mature cystic teratoma with serous cystadenoma and two cases had mature cystic teratoma with co-existing mucinous cystadenoma. The cystadenoma component was colliding with mature cystic teratoma in all the three cases, and no admixture of two components or any intermediate cell zone was identified on extensive sampling, thus excluding the possibility of composite tumour and establishing the diagnosis of collision tumour [Table/Fig-6].

Vascular lesions of ovary is a highly debatable topic and some authors had earlier considered pure ovarian haemangioma as monodermal teratoma. However, this concept was challenged by Prus D et al., when they established on the basis of molecular genetic tests that ovarian haemangioma associated with mixed germ cell tumour in their case of Turner syndrome was of somatic origin and not germ cell derived [15]. In the present case, cystic teratomatous area was present separately from the greyish brown solid haemangiomatous component raising the possibility of mature cystic teratoma associated with cavernous haemangioma. However, on further sampling of solid component, focal areas lined by stratified squamous epithelium were identified, thus establishing the diagnosis of mature cystic teratoma with a prominent haemangiomatous component. Therefore, in a case of teratomas with prominent vascular proliferations, extensive sampling is required to unearth other teratomatous components, as their presence would rule out the possibility of true ovarian haemangioma.

Adipose tissue is not native to the ovary and lipomatous ovarian lesions are exceedingly rare. Various theories explaining the histogenesis of lipomatous lesions in the ovary are:

- Misplaced embryonic fat cells
- Adipose metaplasia of ovarian mesenchymal cells
- Lipomatous lesions originating from fat cells of teratoma [16]

In the present case, possibilities of ovarian lipoma, adipocyte rich teratoma and atypical lipomatous tumour were considered [Table/Fig-6]. However, this lipomatous lesion was associated with adjacent mature cystic teratoma and also few eccrine ducts were present at the periphery of the lesion [Table/Fig-5b], thus excluding the possibility of pure ovarian lipoma. No atypical hyperchromatic nuclei or lipoblasts were seen even on extensive sectioning, which ruled out the possibility of atypical lipomatous tumour, and thus establishing the diagnosis of a separate adipocyte rich teratoma.

| Case No. | Age/ Sex | Chief complaints | Radiological findings | Provisional diagnosis | Differential diagnosis | Final diagnosis | Follow-up |
|---|---|---|---|---|---|---|---|
| Case 1 | 24 years/ Female | Abdominal pain | Benign cystic multilocular mass-right ovary | Mucinous cystadenoma | Mature cystic teratoma, collision tumour, composite tumour | Collision tumour-right ovary | Uneventful postoperative recovery period |
| Case 2 | 43 years/ Female | Abdominal lump and menstrual irregularity | Cystic ovarian mass-left ovary | Benign cystic lesion (dermoid cyst) | Mature cystic teratoma, collision tumour, composite tumour | Collision tumour-left ovary | Uneventful postoperative recovery period |
| Case 3 | 41 years/ Female | Abdominal pain, heaviness and menstrual irregularity | Cystic, multilocular, non vascularised formation-right ovary | Benign cystic lesion | Mature cystic teratoma, collision tumour, composite tumour | Collision tumour-right ovary | Uneventful postoperative recovery period |
| Case 4 | 37 years/ Female | On and off abdominal pain | Semicystic mass-right sided ovary | Benign cystic lesion | Mature cystic teratoma associated with vascular mass lesion (possibility of haemangioma, lymphangioma and angiosarcoma were considered) | Mature cystic teratoma associated with florid vascular proliferation-right ovary | Uneventful postoperative recovery period. No recurrence after two years follow-up |
| Case 5 | 41 years/ Female | Abdominal pain, abdominal lump and weakness | Solid-cystic bilateral adnexal mass | Benign cystic lesion (dermoid cyst) | Mature cystic teratoma with ovarian lipoma, mature cystic teratoma with adipocyte rich teratoma, mature cystic teratoma with atypical lipomatous tumour | Bilateral mature cystic teratoma with distinct right sided adipocyte rich teratoma | Uneventful postoperative recovery period. No recurrence after two years follow-up |

**[Table/Fig-6]:** Summary of cases.

Mature cystic teratomas associated with prominent haemangiomatous component is a very rare finding with only few case reports in the literature [12,13]. The pathological differentials for this phenomenon are true ovarian haemangioma, prominent haemangiomatous component of a teratoma, lymphangioma and angiosarcoma [Table/Fig-6]. However, in the present case, no nuclear pleomorphism, mitosis or necrosis was seen excluding the diagnosis of angiosarcoma and also there was absence of pale eosinophilic secretions in vascular lumens which ruled out the possibility of lymphangioma. Other entities which should be excluded are proliferating blood vessels in hilar region and closely arranged medullary blood vessels in a post-menopausal female, though these physiological changes in ovary do not form a mass lesion and are usually admixed with nerve fibres and lymphatics [14].

Gardella C et al., [17], had reported a case of ovarian lipomatous tumour. However, in their case the presence of benign sweat ducts at the periphery of ovarian lipoma, was indicative of teratomatous origin of lipomatous tumour. Mpatsoulis D et al., had reported prominent adipocytic differentiation in mature cystic teratoma associated with Leydig cell hyperplasia and clinical manifestations of hyperandrogenism. The authors contended that adipose tissue is a complex endocrine organ and might be at least partially responsible for androgenic manifestations [18]. However, no evidence of hyperandrogenic state or Leydig cell hyperplasia was associated with lipomatous proliferation in the present case. Therefore, we conclude that this discrete lipomatous lesion in our case is a separate non functioning adipocyte rich teratoma.

The factors which affect management and prognosis of these tumours include types of colliding or associated components, most aggressive component present, and stage of malignant tumour, if malignant component is present [19]. However, in all these cases mature cystic teratomas were associated with benign lesions, thus surgical excision of affected ovary was considered curative.

## CONCLUSION(S)

The cases included in this series had very uncommon pathological presentations of mature cystic teratomas, and they were diagnosed incidentally after histopathological evaluation only. Therefore, extensive tissue sampling of cystic ovarian masses is highly recommended to avoid a possible misdiagnosis and to unearth the buried components, as this might impact patient management and outcome.

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**PLAGIARISM CHECKING METHODS:** [Jain H et al.]
- Plagiarism X-checker: Mar 28, 2022
- Manual Googling: May 12, 2022
- iThenticate Software: May 16, 2022 (4%)

**ETYMOLOGY:** Author Origin

**AUTHOR DECLARATION:**
- Financial or Other Competing Interests: None
- Was informed consent obtained from the subjects involved in the study? Yes
- For any images presented appropriate consent has been obtained from the subjects. Yes

Date of Submission: **Mar 24, 2022**
Date of Peer Review: **Apr 07, 2022**
Date of Acceptance: **May 13, 2022**
Date of Publishing: **Jun 01, 2022**