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Case Series

Seromucinous tumors of ovary: A case series

Divya Dinesh¹, Vinutha Gali¹, Ajitha Rajalingam¹, J Thanka^{1*}

¹Dept. of Pathology, Sree Balaji Medical College and Hospital, Chennai, Tamil Nadu, India



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ABSTRACT

Seromucinous neoplasms are new category of ovarian epithelial tumor in the 2014 revised World Health Organization classification. Surface epithelial neoplasms are the most common ovarian tumors. Based on the degree of stratification and the presence or absence of invasion, these epithelial proliferations are further divided into benign, borderline, and malignant categories. Benign tumors are divided into three subtypes: cystadenoma (cystic areas), cystadenofibroma (cystic and fibrous areas), and adenofibroma (predominantly fibrous sections). Serous adenofibromas are rare variant of serous surface epithelial tumors. They affect females between the ages of 15 and 65 and make up 1.7% of all benign ovarian tumors. Serous tumor with a solid, fibrous component is designated as serous cystadenofibroma in which both serous and mucinous components are closely intermixed. Seromucinous adenofibroma is a benign cystic neoplasm composed of glands and cysts lined by an admixture of bland mullerian type epithelium which includes ciliated, endocervical type mucinous, and endometrioid. They have a characteristic prominent fibromatous hypocellular stroma which helps differentiate it from the commoner cystadenoma. Seromucinous borderline tumors is 10% -20% of all epithelial ovarian tumors. Develops in younger age, usually bilateral in upto 40%, associated with endometriosis. We discuss about four different cases of seromucinous tumor reported in our institute.

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1. Introduction

The most prevalent ovarian neoplasms, surface epithelial stromal tumors, are classified into five subtypes: serous, mucinous, endometrioid, clear cell, and transitional cell, as well as combinations of these types.¹ When seromucinous ovarian tumors were included as a new category to the 2014 WHO classification, the name "seromucinous" attracted a lot of attention. These cancers included seromucinous borderline tumor/atypical proliferative seromucinous tumor, seromucinous carcinoma, and seromucinous cystadenoma/cystadenofibroma. They ranged in nature from benign to malignant. Nevertheless, seromucinous carcinomas were reclassified as endometrioid

carcinomas with mucinous differentiation in the 2020 WHO Classification of Female Genital Tumours, eliminating them as a separate entity.² The clinical and pathological characteristics of seromucinous tumors are described in this case series.

2. Case History

First case is a 32-year-old female with low back ache. Per abdomen examination showed an 18-week size mass occupying suprapubic region and left iliac fossa. CA-125 value was 27.20u/ml. MRI Pelvis showed a bulky right ovary and a well-defined abdominopelvic cystic lesion measuring 12.2x11x7.9cm. A cyst with attached ovary and tube sent for histopathological examination showed a uniformly thickened cyst wall. Cut surface of ovary

* Corresponding author.

E-mail address: sauparnika.dd@gmail.com (J. Thanka).

appeared grey brown. Microscopy showed cyst wall lined by cuboidal epithelium and focal tall columnar epithelium with apical mucin (Figure 1). Ovarian parenchyma showed follicular cyst with areas of hemorrhage. A diagnosis of seromucinous cystadenoma of right ovary was made.

Second case is a 63-year old postmenopausal woman attended gynecology out patient department with complaints of intermittent lower abdominal pain for one year. On per vaginal examination, a non tender cystic mass of 10cm was felt in right fornix. Ultrasound abdomen showed a right ovarian cyst measuring 10x8.4x6.1cm with a volume of 270ml in right adnexa extending till middle, suggestive of right adnexal complex cyst. Left ovary was bulky measuring 5x2.9cm with a cyst measuring 3x1.9cm, suggestive of left ovarian simple cyst. Computed tomography of abdomen with pelvis showed a cystic lesion in right adnexa measuring 64x60mm with septation. Another satellite component and calcification suggestive of serous cystadenoma in right ovary. CA-125 value was 21.3units/ML. Uterus with cervix and bilateral ovary and fallopian tubes was received in the central laboratory of Pathology department. Right ovary was measuring 5x3x2cm with an attached right adnexal cyst measuring 11x10 cm. External surface and cut surface of right ovarian parenchyma with attached adnexal cyst and right fallopian tube appeared brownish black in color (Figure 2). Cut surface of right ovarian parenchyma showed cystic areas. Wall thickness measured 0.3cm with presence of papillary excrescence. Cut surface of right adnexal cyst was uniloculated with serous fluid within. Left ovary surface showed nodular bossilated areas and two cysts, cut surface also showed the same. Nodular area was gritty to cut. Two cysts seen, largest was multiloculated measuring 2cm in diameter (Figure 3). On Histopathological examination, right ovary showed features of near completely infarcted benign adenofibroma. Left ovary showed features of benign seromucinous adenofibroma (Figure 4) with areas of calcification. There was no evidence of malignancy in the examined sections. Hence, a final impression of near completely infarcted right ovarian adenofibroma and benign seromucinous adenofibroma of left ovary was made.

Third case is a 28year unmarried female who had heavy menstrual bleed and lower abdominal pain of 20 days duration. On ultrasound abdomen a cyst of size 6.5x5.8cm and volume of 316cc showing fish net appearance with internal echoes was replacing right ovary. Helical CT study of abdomen showed a large midline pelvic multiloculated cystic lesion of varying attenuation abutting the bilateral ovaries. CA125 value was 170.40u/ml. Postoperatively peritoneal wash and right ovarian cyst was received in histopathology lab measuring 12x8x7cm. External surface was smooth and globulated with areas of congestion. Cut surface showed multiloculated thin walled cyst filled with mucin which also showed multiple

papillary excrescence and one foci of solid area (Figure 5). Histopathological examination showed serous component (90%) and mucinous component(10%), micropapillary variant with microinvasion (Figure 6). As tumor was limited to one ovary with intact capsule and there was no malignant cells in peritoneal washing, the case diagnosed as PT1A/FIGO IA seromucinous borderline tumor with 90% serous component, 10% mucinous component, micropapillary variant with microinvasion according to CAP protocol for examination of specimen from patients with primary tumor of ovary.

Fourth case is a 72year old female presented with mass descending per vagina. On evaluation MRI abdomen showed multiple ovoid foci in endometrium. A heterogenous lesion measuring 4.1x6.5x4.6cm noted in the right adnexa with edematous changes pushing the uterus toward left lateral aspect suspicious of endometriotic cyst. Staging laparotomy with TAH with BSO was performed and the specimen was sent for histopathological evaluation. On gross examination multiple endometrial polyp and fibroids were noted in the uterus. Cut surface of right ovary showed multiloculated solid and cystic lesion with mucinous area and necrosis. Left ovary showed grey white solid areas with multiple tiny cysts (Figure 7). Histopathological examination of right ovary showed seromucinous borderline tumor without ovarian surface involvement (Figure 8). Benign seromucinous adenofibroma of left ovary (Figure 9). Peritoneal fluid did not show any malignant cell.

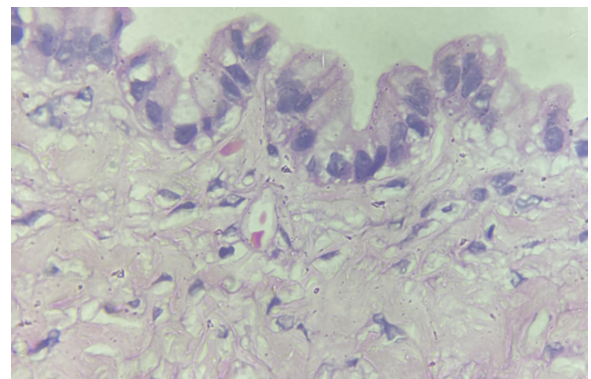


Figure 1: Cyst wall lined by tall columnar epithelium with apical mucin, 100x

3. Discussion

Surface epithelial stromal tumors of the ovary, encompass five main distinct subtypes, namely, serous, mucinous, endometrioid, clear cell and transitional cell along with combination of these types.^{1,3} Seromucinous neoplasms are new category of ovarian epithelial tumor in the 2014 revised World Health Organization classification.⁴ Seromucinous tumors are rare neoplasm of mullerian origin,



Figure 2: Specimen showing infarcted right ovary, adnexal mass, right fallopian tube and multiple cysts in left ovary



Figure 3: Left ovary showing solid area and multiple cystic areas

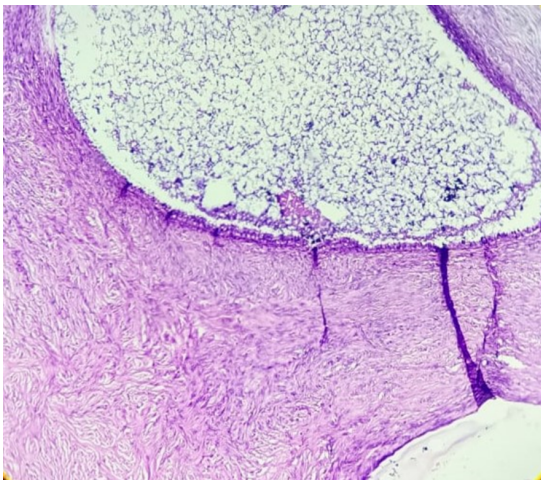


Figure 4: Benign seromucinous adenofibroma, 40x



Figure 5: Multiloculated thin walled cyst filled with mucin which also showed multiple papillary excrescence

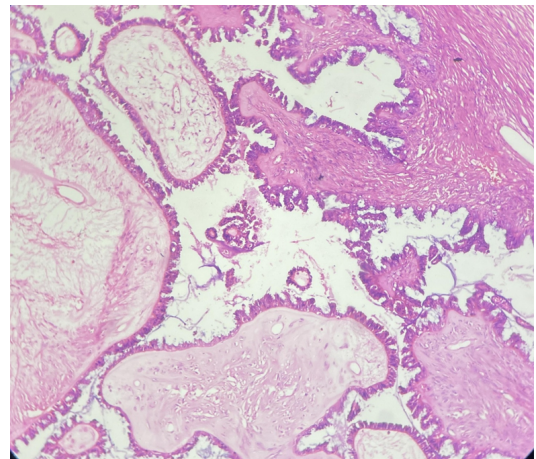


Figure 6: Section show micropapillary variant, 100x

they are benign, borderline and malignant categories and are characterized by an admixture of various cell types including endocervical-type mucinous, endometrioid and squamous type epithelium and have a close relationship with endometriosis. Every category of seromucinous tumor shares three similar characteristics: (i) frequent involvement of both ovaries (20–40%); (ii) relationship with endometriosis (30–70%); and (iii) absence of gastrointestinal differentiation, as shown by Paneth cells and/or goblet cells mixed together. The diagnosis of seromucinous tumor is skeptical with the evidence of gastrointestinal differentiation.⁵

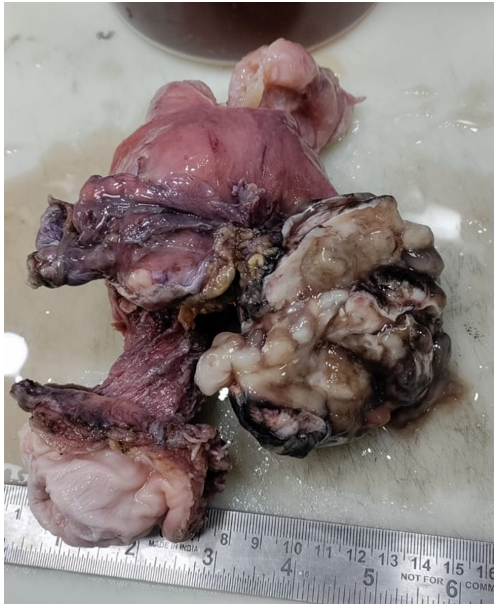


Figure 7: Ovary showing grey white solid areas with multiple tiny cysts

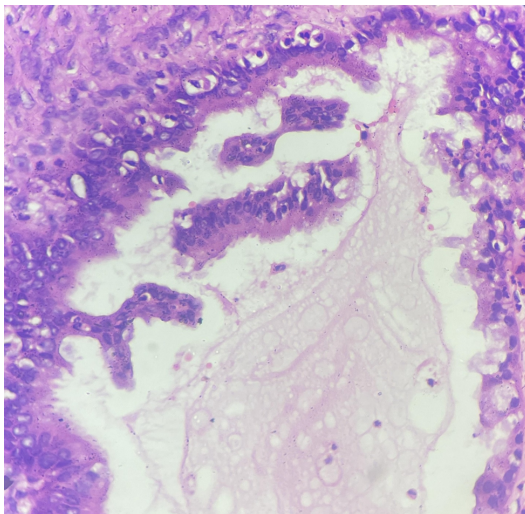


Figure 8: Right ovary showing seromucinous borderline tumor, 100x

3.1. Serous cystadenoma

Serous cystadenoma can be divided into different types depending on the quantity of fibrous tissue present, including cystadenoma, cystadenofibroma, adenofibroma, papillary cystadenoma, papillary cystadenofibroma, and papillary adenofibroma. Serous adenofibromas are rare variant of serous surface epithelial tumors.⁶ Serous tumor with a solid, fibrous component is called as serous cystadenofibroma in which both serous and mucinous components are intermixed. Grossly these tumors are predominantly solid with dense fibrous cut surface and

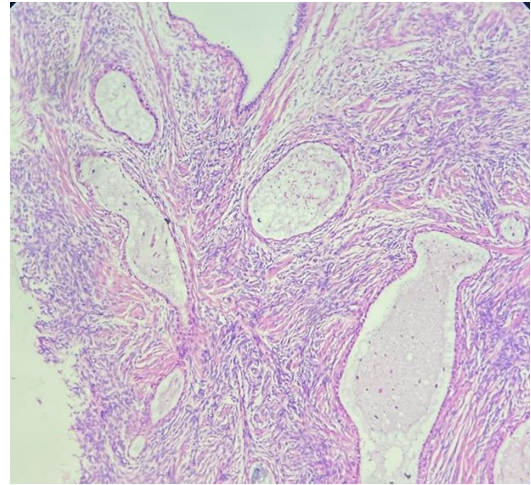


Figure 9: Left ovary showing seromucinous adenofibroma

sometimes appear as nodule in the wall of an endometriotic cyst.

3.2. Mucinous cystadenoma

Ovarian primary mucinous tumors are a diverse group of neoplasms made up of gastrointestinal-type cells that fall into three categories: benign, borderline, and malignant.⁷ Because CA-125 antigen is not raised in the majority of primary ovarian mucinous neoplasms, CA-19-9 antigen is utilized as a marker in ovarian mucinous tumors.⁸ Rupture of a stage I mucinous ovarian carcinoma may increase the likelihood of its recurrence, hence care should be given by the surgeon when entering the abdomen to remove the affected ovary intact without spilling the mucinous contents.⁹ As benign mucinous ovarian cystadenomas are restricted to the ovary, no additional treatment is necessary. Although appendectomy was once conducted for any benign lesions or ovarian tumor with mucinous histology, the available data suggest against doing so as long as the appendix looks normal and there is no sign of pseudomyxoma peritonei.¹⁰ Ovarian mucinous tumors are a unique histologic entity. Their etiology, pathologic features, molecular signature, and clinical behavior set them apart from other forms of epithelial ovarian malignancies.

3.3. Seromucinous adenofibroma

Seromucinous adenofibroma is a rare benign cystic neoplasm made up of glands and cysts that are lined by an admixture of bland müllerian type epithelium that includes ciliated, endocervical type mucinous, and endometrioid. About 1% of ovarian epithelial neoplasms are these benign tumours.⁵ They can be distinguished from the more frequent cystadenoma by their distinctively conspicuous fibromatous hypocellular stroma. Adenofibromas are extremely uncommon benign tumors with extremely

uncommon malignant potential. They arise from the ovarian stroma and germinal lining. Whether a tumor is solid, semisolid, or liquid depends on the ratios of the stromal and epithelial components as well as the secretory activity of the epithelial component. They have short, wide papillary projections, are multiloculated and enclosed, and can reach a size of 20 cm.¹¹ These lesions may occasionally be bilateral or unilateral. It may also penetrate extraovarian regions, such as the coelomic cavity and peritoneal cavity.⁵ In our case the Right ovary had two cystic structures but the left ovary had both solid and cystic component which favour our diagnosis of adenofibroma. On microscopy, presence of significant fibrous stroma separating the glands and cyst with no significant atypia of the lining cells led the diagnosis of seromucinous adenofibroma. The treatment of choice is complete surgical removal of the tumor. The prognosis is generally excellent with prompt and appropriate treatment.

3.4. Seromucinous borderline tumor

In contrast to benign ovarian tumors, borderline ovarian tumors have greater levels of epithelial proliferation and variable nuclear atypia without destructive stromal invasion.¹¹

10% to 20% of all epithelial ovarian cancers are borderline tumors occurs at a younger age, is typically bilateral in up to 40% of cases, and is linked to endometriosis.⁵ On gross examination, they show papillary projections on the inner surface of cysts. The cyst wall is frequently thicker and fibrous and 8 to 10 cm is the average size. Due to neutrophil infiltration, cyst material is frequently mucopurulent and can be hemorrhagic, serous, or mucinous.¹² Shappell et al. provided the initial description of seromucinous tumors in 2002. They discussed the clinicopathologic characteristics of 54 ovarian tumors that included papillary architecture, a predominance of ciliated serous type epithelium, and endocervical mucinous epithelium of which 34 (63%) were atypical proliferative (borderline), 5 (9%), intraepithelial, 8 (15%), microinvasive, and 7 (13%) were carcinoma. After initially including borderline tumor in the 2014 4th Edition of the WHO classification of Female Genital Tumors, "Seromucinous Carcinoma" has been eliminated as a unique entity in the 2020 5th Edition. The term "seromucinous carcinoma" has been eliminated since it was difficult to make a reliable diagnosis and had a large physical overlap with endometrioid carcinoma. It currently only recognize benign and borderline seromucinous tumors as separate entities. Complex cystic-solid masses are visible on MRI.²

Microscopically, SMBT exhibits a papillary structure with hierarchical branching, with stroma that are frequently bulbous and edematous, occasionally sclerotic. In the stroma, epithelium, or luminal spaces, many neutrophils are frequently seen. IHC stains CK7, ER, PR, CA125, and

PAX8 are typically positive in borderline seromucinous tumors while CK20, CDX2, and WT1 are typically negative.²

Usually, they appear as peritoneal implants. On the peritoneal surface, the implants may cause desmoplastic stromal reactions, and during surgical procedures, embedded clusters of mucin-producing tumor cells may be mistaken for disseminated adenocarcinoma on frozen sections. The 10-year survival rate for patients with SMBT is excellent, ranging 83-91%.²

4. Conclusion

Ovarian cancer continues to be a risk for woman, as the majority of patients will not be diagnosed at an early stage. These tumors has a wide range of age of presentation. Maintaining a heightened index of suspicion when approaching an ovarian mass can help in early diagnosis which will improve the patient's prognosis. Here we present four interesting cases of seromucinous tumors of ovary to highlight each of its histological features, among which seromucinous adenofibroma is a rare entity.

5. Source of Funding

None.

6. Conflict of Interest

None.


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Vinutha Gali, Associate Professor  <https://orcid.org/0000-0002-0021-0839>

Ajitha Rajalingam, Assistant Professor  <https://orcid.org/0000-0002-5852-665X>

J Thanka, Professor and Director  <https://orcid.org/0000-0002-7499-5552>

Author biography

Divya Dinesh, Post Graduate  <https://orcid.org/0009-0002-4780-0091>

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