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Case Report

Struma ovarii with serous cyst adenoma: a rare case report

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ABSTRACT

Struma ovarii is a rare ovarian teratoma of mono dermal variant constituting 1-4% of all benign teratoma. Any ovarian teratoma is called struma ovarii only when it consists of at least 50% thyroid tissue. Its coexistence is extremely uncommon with serous cystadenoma with only a handful of cases reported till date. Hereby we present a rare case of 36 years old women with hypothyroidism presented to our OPD with pain abdomen for past 3 months and ovarian mass. Clinicoradiologically possibility of an endometrioma was considered. Histopathological examination revealed coexisting double pathologies; of which struma ovarii was an incidental finding. This case is important, not only being rare, but it also highlights the importance of careful and extensive histopathological examination even in a seemingly simple cystic lesion of the ovary to avoid missing concomitant focal pathologies.

Keywords: Ovarian torsion, Struma ovarii, Serous cystadenoma, Salpingo-oopherctomy, Ovarian tumor, adenoma, Ovarian neoplasm

INTRODUCTION

Struma ovarii is a monodermal teratoma and was originally described by Boettlin on 1889 as a rare form of ovarian tumor.[1] Struma ovarii originates from a single germ cell after first meiotic division and it means "ovarian goitre". It constitutes of only 2.7% of all benign ovarian teratoma.[3]

Any ovarian teratoma is called struma ovarii only when it consists of at least 50% thyroid tissue and thyrotoxicosis, which is thought to be caused by ectopic thyroid hormone synthesis, it is a symptom that only 8% of these tumors exhibit. 5-10% of these cases can undergo malignant transformation.[2-4] Struma ovarii undergoing torsion may not present with feature of thyrotoxicosis hence histopathology remains the main source of confirmation of the diagnosis.

A coexistence of struma ovarii with serous cystadenoma has only been reported in fewer than 10 cases so far.[5-10] Due to its rarity, there is no consensus on the best management of struma ovarii and each case must be individualized.

CASE REPORT

A thirty-six-year-old P3L3 presented to our gynaecology outpatient department with the complaints of dysmenorrhea for the past 3 months. Pain was gradual in onset, progressive, dull aching, radiating to back, mainly confined in left hypochondrium region. About 10 days back, she had an acute onset of pain for which she contacted some private practitioner where she was prescribed some anti-inflammatory drugs along with antibiotics, but her symptoms were not relieved completely and therefore, she visited our hospital with the above complaints.

She was a known case of hypothyroidism since 4 years. She was currently euthyroid and not on any medications. Her menstrual cycle was regular. There was no significant surgical or family history.

On examination, her general physical examination was normal. On per abdomen examination mild tenderness was present in left iliac fossa. There was no rebound tenderness, guarding or rigidity. Her per speculum examination was within normal limits. On per vaginal examination, uterus was anteverted, normal size, mobile, non-tender. Right fornix was free; ~8×9 cm soft cystic mass was felt in left fornix separately from the uterus with mild tenderness and restricted mobility. Her per rectal examination also confirmed the same findings. All other systemic examinations were within normal limits. She was admitted for further workup and management.

Ultrasound scan of whole abdomen and pelvis revealed uterus WNL with ET ~8.1mm, large left ovarian endometrioma ~71×68 mm with right adnexa clear and no peritoneal fluid. She had MRI done which was also suggestive of left ovarian hemorrhagic cyst ~90×59×90 mm with D/D of ovarian torsion with venous congestion. Tumor marker CA125 came out to be 26.60 U/ml.

She was planned for laparotomy with provisional diagnosis of left sided ovarian torsion. Intra operatively hemorrhagic peritoneal fluid was sent for cytology. There was ~10×9 cm bluish colored cyst on left adnexa with a twisted pedicle (3 turns of torsion across axis) with vascularity. Fimbrial end of this tube was adherent over swelling with flimsy adhesions. Right side was normal. She was then proceeded for left sided salpingo-oopherectomy along with right sided tubal ligation as patient has completed her family and was willing for sterilization.

Her post op course was uneventful and was discharged to follow up with histopathology report and repeat thyroid profile.

Peritoneal fluid cytology smear showed clusters to sheets of cuboidal cells having bland nuclear chromatin with scanty cytoplasm. No atypia seen. Suggestive of serous ovarian tumour with further advise for histopathology evaluation. Obtained tissues were sent for histopathological examination in separate containers. Gross examination of left salpingo-oopherectomy revealed ovarian cyst of ~11×7.5×5 cm with attached fallopian tube measuring 5 cm in length. On cut section, cyst was filled with colloid like gelatinous fluid with wall thickness of 1 mm.

Microscopic examination showed cyst wall thrown into extensive branching and tufting hierarchy pattern of papillary projections (Figure 1 A) lined by single layer of serous epithelium (tall columnar ciliated cells) with fibrovascular core (Figure 1 B) with a foci showing thyroid like micro-macro follicles filled with colloid which is lined by flattened cuboidal epithelium (Figure 1 C and D). No nuclear atypia or stromal invasion was seen. Findings were suggestive of struma ovarii coexisting with serous cyst adenoma ovary. Post operative period was uneventful. Later she was lost to follow up.

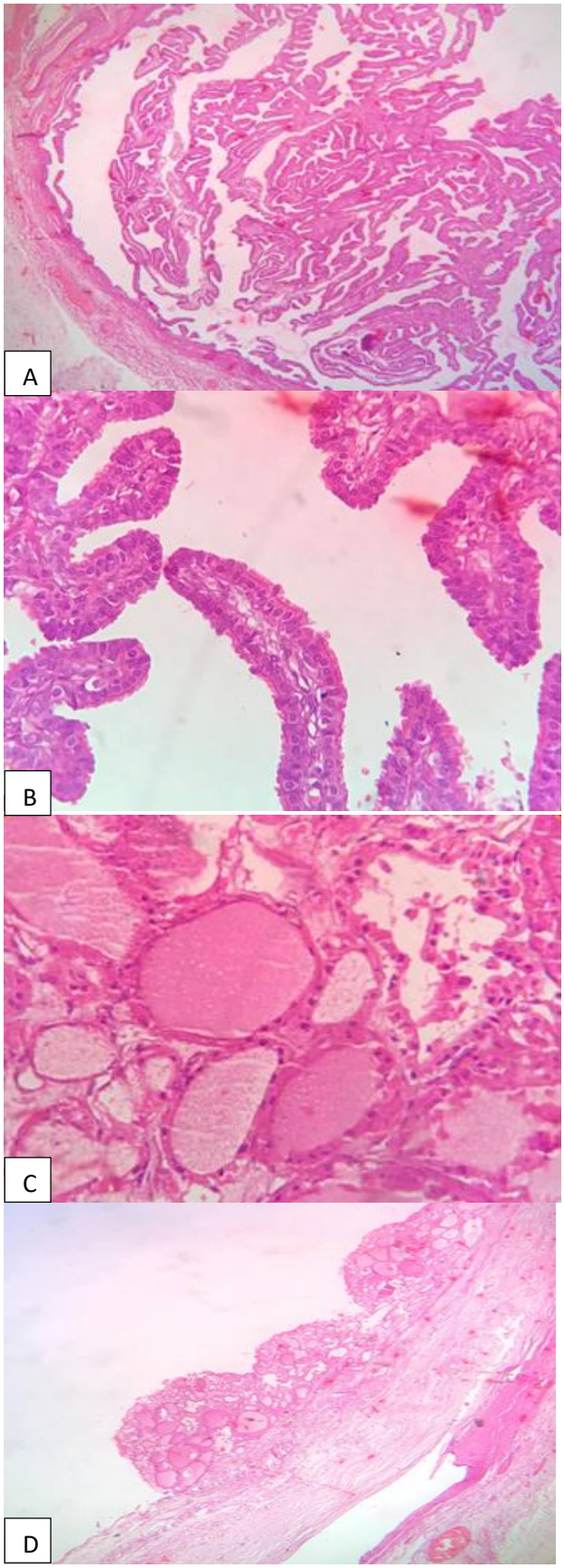

Figure 1 (A-D): Papillary projections, tall columnar ciliated cells, micro-macro follicles and flattened cuboidal.

DISCUSSION

Struma ovarii is a highly specified mono dermal teratoma consisting of predominantly thyroid ectopic tissue. these cystic tumors are typically slow growing, measuring between 5 and 10 cm and most often are unilateral. Of these malignant transformations occurs in <10% in the form of papillary and follicular carcinoma similar to thyroid malignancies.

Grossly struma ovarii has solid or cystic areas. Solid area shows glistening tan brown nodules. Cystic variety consists of clear to green-brown fluid and often can undergo torsion, but cyst rupture is rare presumably because their thick cyst wall resists rupture compared with other ovarian neoplasms.[5]

While serous cystadenoma's development can be attributed to ovarian surface epithelium metaplasia, teratoma is a tumor of the germ cells.[11,12] A different scenario would be that the ovum developed in a Graafian follicle that did not burst, resulting in the teratoma, and the mucinous cystadenoma (or serous cystadenoma in our instance) developed from the follicular lining's "epithelial metaplasia".[13] Due to the small number of occurrences reported in the literature, this uncommon cohabitation is not explained by a straightforward explanation. Pelvic mass presenting as struma ovarii with serous cyst-adenoma in ipsilateral adnexa is a rare entity with only 7 case reports so far.[5-10] Diagnosis of coexisting epithelial tumour is quite difficult because struma ovarii can show extensive pale cuboidal epithelium with lined cystic areas with fibrous hyalinized small papillae which can be misinterpreted as serous cyst adenoma. Thyroglobulin and thyroid transcription factor-1 levels on immunohistochemistry is the deciding factor in these difficult cases.

In our case, histopathological confirmation of struma ovarii with co-existing serous cyst adenoma consisting of tall columnar ciliated cells was a crucial diagnostic entity to confirm this rare encounter. It is difficult to ascertain the modality of diagnosis and its treatment because of constricted literature. It is very important to keep in mind all the possible differentials and a thorough pre operative evaluation along with extensive tumour sampling to confirm the rare presentation of ovarian tumours.

CONCLUSION

Diagnosing struma ovarii before surgery is challenging. Histopathology and immunohistochemistry remain the only sole modality for confirming struma ovarii as there was various differential diagnosis on imaging modalities. Surgery and histopathological examination remain the treatment for this rare entity along with further follow up.

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