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Case Report

Benign struma ovarii-a rare monodermal ovarian teratoma-a case report

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ABSTRACT

Struma ovarii is a rare ovarian tumour, first described in 1889 by Boettlin. It is defined by the presence of thyroid tissue comprising of >50% of overall mass. It comprises 1% of all ovarian tumours and 2-5% of all ovarian teratomas. Preoperative diagnosis of struma ovarii is difficult because symptoms, clinical presentation and ultrasound are often similar to that of ovarian carcinoma. Hence most of the patients are diagnosed post operatively. Most cases of strum aovarii are benign and can be treated by excision of the ovary or by unilateral salpingo-oophorectomy. In a small number of cases, there are complications, the most important being the development of malignancy or ascites associated with pleural effusion producing a pseudo-Meigs' syndrome. This is a case of struma ovary presented with features of pseudo-Meig's syndrome. A 68 year old post-menopausal woman presented with acute abdomen and respiratory distress with an ultrasound diagnosis of ovarian torsion, ascites and pleural effusion and found to have atrial thrombus on evaluation. She had undergone staging laparotomy, TAH+ BSO, omental biopsy and peritoneal fluid cytology. Histopathology revealed predominantly benign and mature colloid filled thyroid follicles of varying sizes lined by cuboidal epithelium surrounded by lymphocytic infiltrate and congested blood vessels, which was suggestive of struma ovary.

Keywords: Struma ovary, Teratoma, Pseudo-Meigs syndrome

INTRODUCTION

Struma ovarii is considered as one-sided development of a teratoma, in which the thyroid tissue has over grown all other tissues, or one in which only the thyroid tissue has developed. The term struma ovarii should be reserved for tumours composed either entirely or predominantly of thyroid tissue. It is uncommon and comprises only 2.7% of ovarian teratomas. The age distribution of patients with struma ovarii is generally the same as that of patients with mature cystic teratoma and ranges from 6 to74 years and majority are in the reproductive age group.[1,3] Clinical findings are like those observed in patients with mature cystic teratoma. The only differences are that in some cases struma ovarii is associated with enlargement of the thyroid gland, and in other cases there is clinical evidence that the struma ovary is responsible for the development of thyrotoxicosis, although this has not been confirmed preoperatively by laboratory tests.[1-3]

Struma ovarii is usually unilateral but is often associated with mature cystic teratoma and measures less than 10 cm in diameter.[1,3] Rarely there will be an associated cystadenoma in the contralateral ovary.[1,3] The surface is usually smooth. Cut surface of the tumour may be composed entirely of light tan, glistening thyroid tissue, haemorrhage, necrosis, and foci of fibrosis. Microscopically the tumour is composed of mature thyroid tissue consisting of acini of various sizes, lined by a single layer of columnar or flattened epithelium. The acini contain eosinophilic, PAS-positive colloid. Thyroglobulin can be identified in epithelial cells by immunohistochemistry. Most cases of struma ovarii are benign and can be treated by excision of the ovary or by

unilateral salpingo-oophorectomy. In a small number of cases, there are complications, the most important being the development of malignancy and the presence of ascites alone or ascites associated with pleural effusion producing a pseudo-Meigs' syndrome.[11] Ascites may be found in 17%of cases of struma ovarii, and its presence does not indicate that the tumour is malignant.[12] The cause of the ascites and pleural effusion has not been fully elucidated. In most reported cases, excision of the tumour led to complete remission.

This is a case of a rare benign struma ovary, composed of thyroid tissue mainly and presented with features of pseudo-Meig's syndrome and atrial thrombus.

CASE REPORT

A 68-year-old P3L3, presented with lower abdominal pain of 2 months duration. She had attained menopause 20 years ago. Examination of abdomen showed a cystic mass of 5×6 cm in the lower abdomen. Bimanual examination showed that cystic mass is felt through the right fornix. Ultrasound revealed a multiloculated cystic lesion showing internal echoes, of size 8.3×6.5 cm in the right adnexae and atrophic uterus with an endometrial thickness of 3 mm.

MRI (Figure 1 A-C) revealed a well-defined complex multiloculated lesion in the right adnexa-possibly ovarian (ORADS-MRI-score 4), intermediate risk. Tumour markers were within normal limits (CA125-5.39, CEA-1.65, TSH -2.4).

Preoperative workup was done and ECHO showed a right atrial thrombus and she was heparinised. She had no history of hypertension, diabetes or thyroid abnormality.

The lesion is showing predominant TIFS hypointense and T2 hyperintense areas within with few locules showing TI hyperintense/T2 hypo and isointense contents within. Few thick internal septations noted. No areas of diffusion restriction noted. Few curvilinear blooming foci noted in GRE sequences-suggestive of septal calcifications. Right ovary not separately imaged. On post contrast study, heterogeneous post contrast enhancement noted with enhancement of the wall, septa with few enhancing mural nodules largest measuring 10×7.7 mm. One of the thick septae shows tiny papillary projections possibly ovarian - ORADS 4 - (intermediate risk).

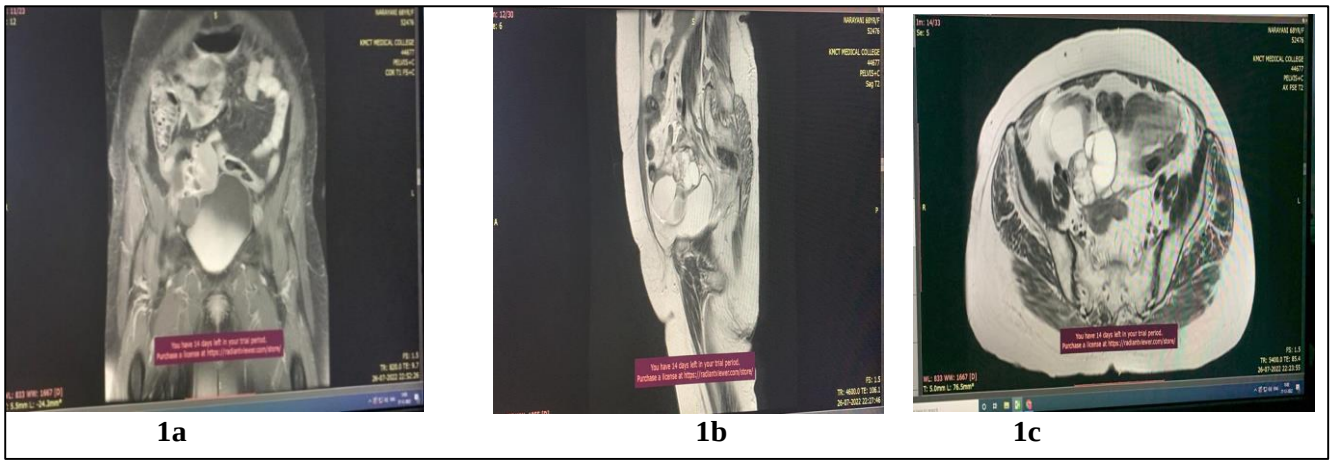

Figure 1 (A-C): A well-defined multi-loculated altered signal intensity lesion measuring 6.3x7.2×5.7 cm (AP×TR×CC) noted in the right adnexae.

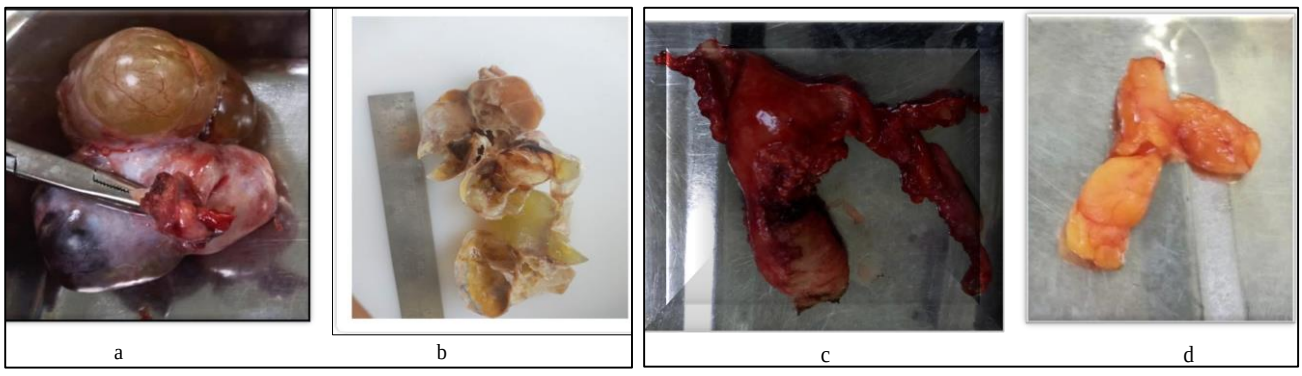

Figure 2 (A and B): Right ovary with complex cyst with solid and cystic area with torsion and areas of haemorrhage and mucin, uterus with left ovary, omental biopsy.

She had undergone staging laparotomy, TAH with BSO, omental biopsy and peritoneal fluid cytology. Findings were a right multiloculated cyst of 10×8×8 cm of variable consistency with solid and cystic area and showed features of torsion-twice around its pedicle. It also showed areas of haemorrhage and mucinous areas (Figure 2 A and B). Uterus and opposite ovary were normal (Figure 2 C). There was minimal ascites. Visualised parts of other intra-abdominal organs were normal.

Post operatively she had respiratory distress and SpO_2 fall. Chest X-ray showed bilateral pleural effusion with mild atelectasis. Her TFT was repeated post operatively and found to have minimally raised T4 levels. Symptoms subsided later over a course of a week.

Histopathological report showed that right ovary was partly cystic and partly solid, focally lined by flattened to low cuboidal epithelium along with few hemosiderin laden macrophages. Underlying stroma showed predominantly mature and benign colloid filled thyroid follicles of varying sizes lined by cuboidal epithelium surrounded by lymphocytic infiltrate, congested blood vessels, colloid laden macrophages and focii of calcification. Also noted focii of mature bony fragments and bony spicules. Periphery showed normal ovarian parenchyma. Opposite ovary, uterus and fallopian tubes were normal

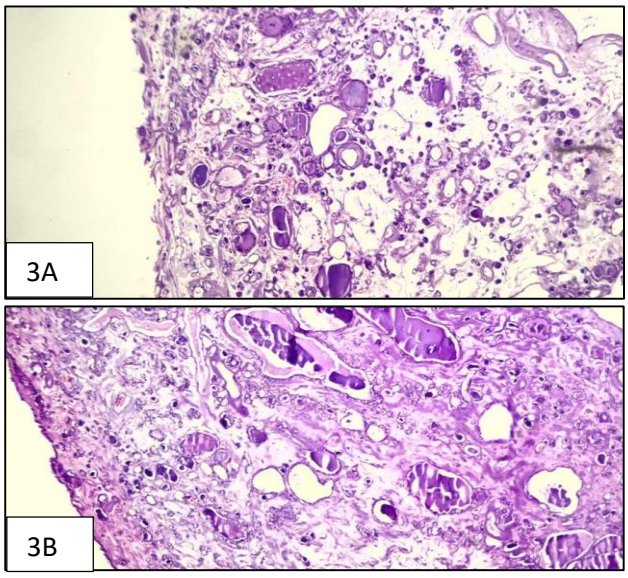

Figure 3 (A and B): Section from right ovarian cyst shows partly cystic and partly solid mass, focally lined by flattened to low cuboidal epithelium along with few hemosiderin laden macrophages. Underlying stroma showed predominantly mature and benign colloid filled thyroid follicles of varying sizes lined by cuboidal epithelium surrounded by lymphocytic infiltrate, congested blood vessels, colloid laden cysts, macrophages and foci of calcification. Also, noted foci of mature bony fragments and bony spicules. Periphery showed normal ovarian parenchyma.

DISCUSSION

Struma ovary was first described in 1889 by Boettlin, followed by Ludwig Pick who suggested these are the ovarian goiters.[5,6] In 1933, Plaut showed that the thyroid tissue in struma ovary is morphologically, pharmacologically, and biochemically identical to that of the cervical thyroid gland.[7] Smith reviewed the literature and found that approximately 5% had extra ovarian dissemination.[2] In 1976 Yannopoulos and associates comprehensively reviewed the literature regarding malignant struma ovary.[9] In the latest world health organisation classification, struma ovary and malignant thyroid tumours arising within the struma are included in the thyroid tumour group under the heading monodermal teratoma and somatic-type tumours associated with dermoid cysts.[10] It is the most common monodermal teratoma, accounting for 3% of all ovarian teratomas.[11]

Struma ovary is defined as ovarian teratoma that is mainly composed predominantly (>50%) or entirely of thyroid tissue or forms a macroscopically recognizable mature cystic teratoma.[12] Also included in this category are <50% thyroid tissue, if functional, histologically or biologically malignant thyroid tissue.[8] The peak incidence of struma ovary is in the 5th decade. But cases are reported in the post-menopausal women. In addition to the usual signs and symptoms of pelvic mass, additional ascites occurs in one third of cases and occasionally have Meig's syndrome. Clinical evidence of hyperthyroidism occurs in 5% of cases.[4] Thyrotoxic crisis is rare, but life-threatening complication following excision of struma in which hyperthyroidism was not diagnosed pre operatively.[13]

This was a case of 68-year-old postmenopausal lady, presented with ovarian torsion. TFT was normal preoperatively. There was minimal ascites and bilateral pleural effusion. Post operatively she had hyperthyroidism, pleural effusion, and atelectasis. The histopathological report showed that it is benign struma ovary.

Clinical findings

Clinical findings were similar to those observed in patients with mature cystic teratoma, presented with abdominal pain, abdominal distension and torsion of the bulky ovary. But are rare in a postmenopausal population as we discussed here.

The only differences are that in some cases struma ovarii is associated with enlargement of the thyroid gland, and in other cases there is clinical evidence that the struma ovarii is responsible for the development of thyrotoxicosis, although this has not been confirmed preoperatively by laboratory tests.[1-3] This patient had a borderline elevation of FT4 levels, which might have precipitated hyperthyroidism and arrhythmia. This may be responsible for developing atrial thrombus. Rarely the

removal of the ovary can cause some symptoms of thyroid storm also.

Some cases of thyrotoxicosis have been associated with struma ovarii, which has also shown evidence of thyroid hyperactivity. The ectopic thyroid tissue presents within struma ovarii, therefore, may be the subject of the same physiological and pathological changes as thyroid gland.[2]

Gross findings

Struma ovarii is usually unilateral, but is often associated with mature cystic teratoma and rarely with a cystadenoma in the contralateral ovary.[1,3] In some cases, the teratoma present in the contralateral ovary contained thyroid tissue. Struma ovarii varies in size but usually measures less than 10 cm in diameter. The surface is usually smooth and, before sectioning, the tumour shows similar appearances to mature cystic teratoma. Occasionally, adhesions may be present. The cut surface of the tumour may be composed entirely of light tan and glistening thyroid tissue. Haemorrhage, necrosis, and foci of fibrosis may be present. We were having a similar picture in this patient. Solid tumours with small amounts of colloid appear less glistening and fleshier.

Microscopic findings

The tumour is composed of mature thyroid tissue consisting of acini of various sizes, lined by a single layer of columnar or flattened epithelium.[15]The acini contain eosinophilic, PAS-positive colloid. The intensity of the staining may vary.[16]The acini, which maybe large, containing a large amount of colloid, or may be small. Thyroglobulin can be identified in the epithelial cells by immuno-histochemistry. Occasionally, the lining of the acini may be columnar, containing small papillary projections not unlike those seen in hyperactive thyroid gland. Sometimes the appearances may resemble a nodular adenomatous goiter. Adenoma-like lesions may also be observed. Struma ovarii showing appearances suggestive of Hashimoto's thyroiditis has also been seen.

Clinical behaviour and treatment

Most cases of struma ovarii are benign and can be treated by ovariotomy or by unilateral salpingo-oophorectomy. In a small number of cases, there are complications, the most important being the development of malignancy and the presence of ascites or ascites associated with pleural effusion producing a pseudo-Meigs' syndrome. Ascites may be found in 17% of cases of struma ovarii, and its presence does not indicate that the tumour is malignant.[2]The cause of the ascites and pleural effusion has not been fully elucidated. In most of the cases, excision of the tumour led to complete remission.

This patient had minimal pleural effusion and moderate ascites associated with the tumour. Malignant change in struma ovarii is uncommon. The criteria for malignancy in struma ovarii are the same as those for thyroid tumors. Treatment consists of surgery and administration of radioactive iodine (I^{131}) and other agents used in the treatment of thyroid malignancy, including radiation therapy. Struma ovarii may be associated with extra ovarian extension caused either by rupture of the tumour or by local spread. In such cases, the peritoneal cavity contains tumour deposits, which may be numerous and are composed of mature thyroid tissue. The condition is benign and is termed benign strumosis.

It is only rarely associated with untoward side effects, which are mainly caused by the formation of adhesions. Benign strumosis may be treated by excision of the tumor deposits or by administration of radioactive iodine (I^{131}).

CONCLUSION

Struma ovary is the most common type of monodermal ovarian teratoma, accounting for nearly 3% of all ovarian teratoma. Although it has been more than 100 years since the first description of struma ovarii, several aspects remain enigmatic. Better recognition of the wide range of macroscopic and histological feature of struma will improve accuracy of diagnosis. Precise criteria should be used for the histological diagnosis of malignancy in cases of struma confined to the ovarii with consideration of the proliferative changes that can mimic cancer. More precise description of the histological changes using terminology applicable to the thyroid gland and long term follow up will improve our knowledge of this entity. This case is important because of its unique features although the presentation was a pelvic mass in a post-menopausal woman, unusual presentation such as hyperthyroidism, ascites and Meigs syndrome made it difficult to diagnose struma ovary clinically.

In this patient, we suspected a cardiac pathology and ovarian malignancy in the beginning, but final histopathological report came as Struma ovary then only we could account for the symptoms.

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