Case Series

Struma Ovarii: A Report of Three Cases and Literature Review

Pratibha Singh, Nitisha Lath, Shashank Shekhar, Manu Goyal, Meenakshi Gothwal, Garima Yadav, Pushpinder Khera[1]

Departments of Obstetrics Gynecology and [1]Radio-Diagnosis, All India Institute of Medical Sciences, Jodhpur, Rajasthan, India

Abstract

Struma ovarii is a variant of mature cystic teratoma, with predominant thyroid element. Confirmatory diagnosis is by histopathology. It may mimic as ovarian malignancy in some. It may be associated with ascites in minority; even CA-125 has been found to be raised in some cases. We here report three cases – two operated for suspected ovarian malignancy in a postmenopausal female and one case operated for persistent benign ovarian cyst with raised CA-125 in a young female. In all histopathological diagnosis was a benign struma ovarii. It is difficult to diagnose these cases preoperatively as there are no specific clinical, radiological or serum markers for these rare tumors in the absence of thyroid biology abnormality. Most cases are diagnosed on histopathology. These benign tumors many times require extensive staging laparotomy for suspected ovarian malignancy in the absence of preoperative diagnosis. Prognosis is good for these tumors. Subtle radiological signs and a clinical suspicion can avoid extensive staging laprotomies in these patients.

Keywords: *Monodermal ovarian teratoma, ovarian tumour, struma ovarii*

Introduction

Struma ovarii is a rare histological diagnosis, a variant of dermoid in which thyroid tissue constitute >50% of the component,[1] also called as monodermal ovarian teratoma where thyroid tissue predominates. This tumor was first described in 1889 by Boettlin. It comprises 1% of all ovarian tumor and 2.7% of all dermoid tumor.[2] It is mostly benign, with malignant transformation in just 5%.[3] Ascites may be associated in one-third of the cases.[4] Cases of struma ovarii with elevated CA-125 have rarely been reported. Struma ovarii rarely produces sufficient thyroid hormone to cause hyperthyroidism, or exceptionally become malignant, and thus managed as a thyroid cancer. Mostly, struma ovarii is managed through surgical removal of the ovarian cyst/tumor.

We present here three cases of struma ovarii reported in our institute, all having different presentations. First is a case of the 70-year-old female with an incidental finding of large complex 10 × 12 cm ovarian mass on imaging with suspected liver metastasis, but with normal CA-125. Second case is a young 35-year-old female with persistent ovarian cyst with raised CA-125 taken up for laparoscopic ovarian cystectomy. Third case is a 50-year-old female with chronic pain abdomen in which there was a large multiloculated heterogeneous ovarian cyst with raised CA-125. She was taken up for staging laparotomy for suspected epithelial ovarian cancer. In all the cases, the histological diagnosis came out to be struma ovarii. We are presenting these cases because of the rarity of the condition and varied clinical and radiological presentation.

Access this article online

Quick Response Code:

Website: www.jmidlifehealth.org

DOI: 10.4103/jmh.JMH_53_18

Case Reports

Case 1

Mrs X, 70 years old Postmenopausal female P4004 presented with vague mass per abdomen and palpitation for the past 4 months. She was nondiabetic and normotensive. She was on tablet Metoprolol, prescribed by a physician for palpitation, for the past 4 months. She was diagnosed to have sinus tachycardia, with no

Address for correspondence: Dr. Pratibha Singh, Department of Obstetrics Gynecology, All India Institute of Medical Sciences Residential Complex, Basni, Jodhpur, Rajasthan, India. E-mail: drpratibha69@hotmail.com

How to cite this article: Singh P, Lath N, Shekhar S, Goyal M, Gothwal M, Yadav G, *et al.* Struma ovarii: A report of three cases and literature review. J Mid-life Health 2018;9:225-9.

features of thyrotoxicosis, anemia, or fever. Her thyroid profile being normal. She was admitted to our institute for further workup and management. On examination, she had no pallor, icterus, or lymph node enlargement. There was mild tachycardia (108 bpm), respiratory, and cardiovascular examination was normal. On abdominal examination, 5 × 5 cm firm mass was felt in suprapubic region arising from pelvis, smooth surface, and nontender. Per speculum findings were suggestive of senile changes in vagina and cervix. On bimanual pelvic examination revealed a large firm mass 14 × 12 cm felt separately, posterior to the uterus. Ultrasound showed a large complex heterogeneous pelvic mass likely to be ovarian malignancy. Contrast-enhanced computed tomography abdomen was done, which revealed a complex solid cystic lesion in pelvis (11 × cm 10 cm × 6 cm) likely right ovarian malignant teratoma [Figure 1] with multiple heterogeneous attenuating masses in liver suspicious of metastasis [Figure 2]. Blood investigation including ovarian tumor markers was normal (S. TSH-3.2Miu/ml, CA 125-12.1, AFP 1.3, Beta hCG (11.1). Her electrocardiogram showed Sinus tachycardia with normal QRS complex. Two-dimensional echo done showed mild pulmonary arterial hypertension, normal left ventricle ejection function (65%). In view of the suspected advanced ovarian malignancy with liver nodule suspicious of metastasis, neoadjuvant chemotherapy was planned. Hence, fine-needle aspiration cytology (FNAC) from the liver nodule was planned. FNAC liver showed blood mixed aspirate, no malignancy.

The decision for surgery was taken for confirmation of diagnosis and debulking of the tumor. After preanesthetic checkup, exploratory laparotomy was done - abdomen opened by midline vertical incision.

Intraoperative findings were as follows:

1. Mild ascites (serous) 30–40 ml which was sent for cytology for malignant cells
2. Left ovarian multilobulated mass 12 cm × 10 cm with solid areas. Right ovary was healthy looking
3. Abdomen was explored in a clockwise manner. A polypoidal mass 4 cm × 3 cm felt over left lobe of liver; with omentum, bowel, GB, stomach, spleen found to be apparently normal.

Total abdominal hysterectomy + bilateral salpingo-oophorectomy + infracolic omentectomy + hepatic mass resection + multiple peritoneal biopsies done and sent for histopathological examination.

Histopathology of the left ovarian tumor showed variably sized thyroid follicles filled with colloid and lined by cuboidal to flattened epithelium. The histopathological report was benign struma ovarii of the left ovary. Liver mass came out to be cavernous hemangioma. Peritoneal fluid was negative for malignant cells.

Case 2

Mrs. Y 35-year-old female P2002 presented with a complaint of vague lower abdominal pain for the past 1 year with persistent left side ovarian cyst. The cyst was followed up by sonography which was not decreasing in size so planned for further workup and surgery.

Her menstrual cycles were regular with the normal flow with no dysmenorrhea. She had a history of excision of breast lump (fibroadenoma). There was no significant medical or drug history.

On examination, no pallor/lymphadenopathy/organomegaly or ascites was found. A mobile cystic mass of 8 cm × 8 cm arising from pelvis confirmed by bimanual pelvic examination. Rest of the systemic examination was normal. Ultrasonography showed a large left ovarian simple cyst of 8.4 cm × 6.6cm with

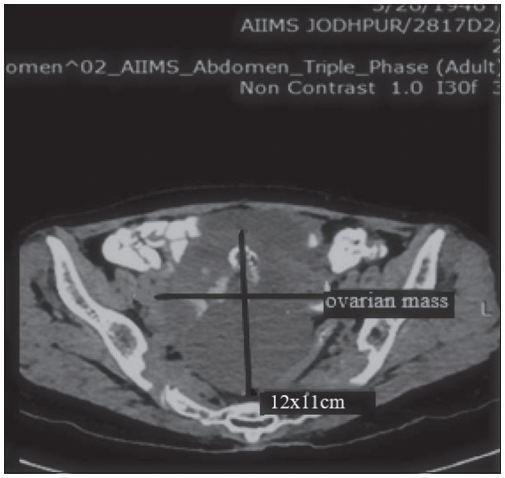

Figure 1: Complex pelvic mass 11 cm × 10 cm likely malignant teratoma

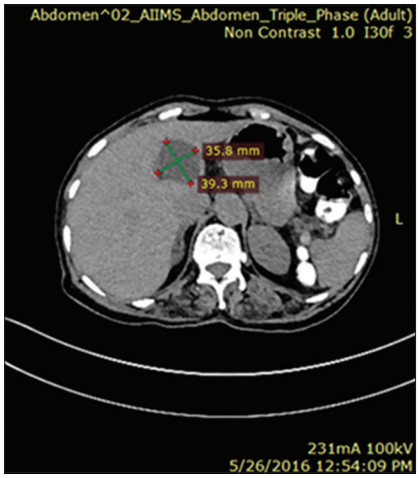

Figure 2: Triple phase computed tomography showing liver lesion

significant post-void residual urine (92 ml). CA-125 was raised (61.57 mIU/L). She was taken up for surgery.

Intraoperative findings were of left ovarian cyst of 8 cm × 8 cm with few fine septations and gelatinous material within the cyst. Endometriotic patches were also seen over anterior wall of uterus and postoperative day (POD). Right side tube and ovary were healthy looking. Laparoscopic left ovarian cystectomy was done, and cyst wall sent for histopathological examination. In the postoperative period, the patient had urinary retention on POD 1, urine culture sent was sterile. She responded to conservative management Histopathological diagnosis was of benign struma ovarii.

Case 3

Mrs. Z a 50 years' postmenopausal female presented with chronic pain in lower abdomen along with nausea and decreased appetite for 2–3 years. She was on antipsychotic medication for the past 4 years (tablet clonazepam 25 mg and tablet Mirtazapine 7.5 mg). On examination, no pallor or supraclavicular lymphadenopathy. Abdominal examination was normal. Bimanual examination revealed 8 cm × 10 cm cystic mass felt through left and anterior fornix. Per rectal examination showed rectal mucosa free with no nodularity in POD. Rest of the systemic examination was within normal limits.

Ultrasonography of abdomen showed gallbladder with 1 cm calculus at neck. A solid-cystic ovarian mass 10 cm × 7 cm × 7 cm, multiloculated, cystic component with fluid of different echogenicity suggestive of mucinous cystadenocarcinoma. Tumor markers were CA-125-43.9 mIu/L, CA 19-9-45.7, and CEA-1.79.

Magnetic resonance imaging (MRI) showed multiloculated cystic mass of 10 cm × 8 cm × 7 cm with multiple thin septa some of locules overtly hypointense on T2W suggesting hemorrhagic content, ovaries not visualized separately, and probably mucinous cystadenoma [Figure 3] MRI of Ovarian Mass. Omental thickening, lymphadenopathy and ascites was absent. Gallbladder has large 12 mm intraluminal calculus in the neck [Figure 4]. Bosniak class I cyst in the right kidney 3 cm × 2 cm was also present.

The surgical opinion was also sought in view of gallbladder calculus, and the patient was taken up for staging laparotomy. Intraoperative findings were - minimal hemorrhagic ascites (30–40cc) sent for fluid cytology for malignant cells. Large multiloculated left ovarian cyst (8 cm × 8 cm), which was adherent to surrounding structures. Dense adhesion was present, adhesiolysis was done and gallbladder identified, there was a 1 cm solitary stone found impacted in the neck of

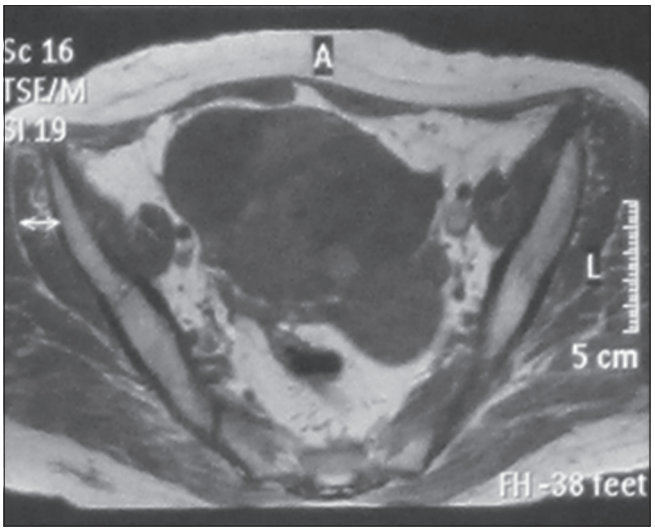

Figure 3: MRI of ovarian mass

gallbladder. Total abdominal hysterectomy with bilateral salpingo-oophorectomy with infracolic omentectomy done along with cholecystectomy. Specimen sent for histopathological examination. Peritoneal fluid was negative for malignant cells. On gross examination, cyst was greyish-white with multiloculated cysts with yellow to tan colored material. Microscopic examination showed tumor composed of predominantly variable sized thyroid follicles filled with colloid, reported as struma ovarii.

Discussion

Mature cystic teratoma (Dermoid cyst) constitute the majority of ovarian germ cell tumors and constitute 20% of ovarian tumors.[5] They are often discovered incidentally on physical or sonographic examination. They may contain hair, teeth, or bone and fatty material. Thyroid tissue is rarely found on histological examination, but if the thyroid tissue predominates (>50%) then the term struma ovarii is applied. It is not common to see hormonally active struma ovarii. Struma ovarii a very rare histological diagnosis is found in just 3% of ovarian teratoma, 2% of all germ cell tumors and 0.5% of all ovarian tumors.[6,7] Malignant transformation is uncommon, in only about 5% struma ovarii.[6,8]

Most patients of struma ovarii are in reproductive age, but it can be diagnosed at any age, even children.[6] Most cases are clinically asymptomatic. It may be associated with ascites, with or without pleural effusion (Pseudo-Meigs syndrome). The ectopic thyroid tissue explains why struma ovarii is sometimes associated with thyrotoxicosis.[6]

Macroscopically, the tumor is mostly solid or solid-cystic, and sometimes cystic with solid areas or protrusions.[9]

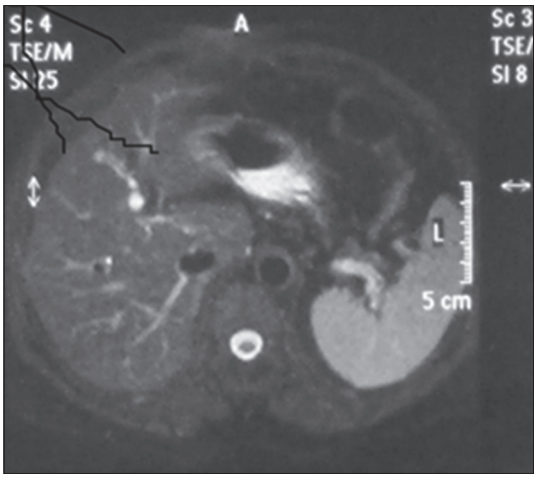

Figure 4: MRI abdomen showing liver, GB

The cut section may look greyish, with fleshy glistening appearance due to thyroid component. On microscopy, it is composed of mature thyroid tissue consisting of colloid containing follicles of various sizes.[6] Ultrasound is a primary modality for identification and characterization of any Ovarian mass. Mature cystic teratoma shows focal high echogenic nodules with heterogeneous internal echoes. Typical feature of struma ovarii on sonography is the presence of well-defined solid tissue with a smooth margin that is vascularized on Doppler study ("struma pearl").[10] An MRI can at times can be helpful due to its ability to distinguish between fluids and fat in the diffusion-weighted image The classic MRI appearance of struma ovarii includes multiple intra-cystic solid areas, representing thyroid tissue, that are of low-signal intensity on T2-weighted images and intermediate signal intensity on T1-weighted images.[11] These characteristic features are not very easily seen/interpreted on radiologic examination; most cases are diagnosed on histopathology reports after surgery. Many cases undergo unnecessary extensive surgery in suspicion of malignancy with its associated morbidity.

Once diagnosed, surgery is the primary modality of the management. Conservative surgery (cystectomy and oophorectomy) is recommended for struma ovarii especially if they have fertility potential, and laparoscopic approach should be the preferred route owing to obvious advantages of laparoscopic surgery.

Benign struma ovarii and malignant forms without metastasis have good prognosis. Ascites or pleural effusion if present disappears after surgery. Malignant cases should also undergo total thyroidectomy followed by radioiodine therapy (131I).[12,13] Serum thyroglobulin is used as tumor marker for follow-up in these malignant cases. As there has been only few reported malignant cases, there is no consistent data on the protocol of management of such cases.[14]

Conclusion

Struma ovarii can mimic ovarian malignancy clinically, when presented with a complex ovarian mass, with ascites and an elevated CA-125. Our 1st and 3rd cases were opened as a case of suspected malignant teratoma, but histopathology revealed it to be benign struma ovarii. The management of struma ovarii is by surgery. If "struma Pearl" can be identified preoperatively, extensive laparotomies may be avoided. Benign struma ovarii has good prognosis and survival without any significant long-term problems. Efforts should be made to diagnose this condition preoperatively, so as to avoid extensive laparotomies, as these benign cases can be managed very effectively by the laparoscopic approach, as was done in our second case.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Willemse PH, Oosterhuis JW, Aalders JG, Piers DA, Sleijfer DT, Vermey A, *et al.* Malignant struma ovarii treated by ovariectomy, thyroidectomy, and 131I administration. Cancer 1987;60:178-82.
2. Kim SJ, Pak K, Lim HJ, Seong SJ. Clinical diversity of struma ovarii. Korean J Obstet Gynwcol 2002;45:748-52.
3. Rosenblum NG, LiVolsi VA, Edmonds PR, Mikuta JJ. Malignant struma ovarii. Gynecol Oncol 1989;32:224-7.
4. Amr SS, Hassan AA. Struma ovarii with pseudo-Meigs' syndrome: Report of a case and review of the literature. Eur J Obstet Gynecol Reprod Biol 1994;55:205-8.
5. Peterson WF, Prevost EC, Edmunds FT, Hundley JM Jr., Morris FK. Benign cystic teratomas of the ovary; a clinico-statistical study of 1,007 cases with a review of the literature. Am J Obstet Gynecol 1955;70:368-82.
6. Talerman A. Germ cell tumors of the ovary. In: Kurman RJ, editor. Blaustein's Pathology of the Female Genital Tract. 3rd ed. New York, NY: Springer Verlag; 2001. p. 967-1033.
7. Yamashita Y, Hatanaka Y, Takahashi M, Miyazaki K, Okamura H. Struma ovarii: MR appearances. Abdom Imaging 1997;22:100-2.
8. Ayhan A, Yanik F, Tuncer R, Tuncer ZS, Ruacan S. Struma

ovarii. Int J Gynaecol Obstet 1993;42:143-6.
9. Roth LM, Talerman A. The enigma of struma ovarii. Pathology 2007;39:139-46.
10. Savelli L, Testa AC, Timmerman D, Paladini D, Ljungberg O, Valentin L, *et al.* Imaging of gynecological disease (4): Clinical and ultrasound characteristics of struma ovarii. Ultrasound Obstet Gynecol 2008;32:210-9.
11. Matsuki M, Kaji Y, Matsuo M, Kobashi Y. Struma ovarii: MRI findings. Br J Radiol 2000;73:87-90.
12. Mui MP, Tam KF, Tam FK, Ngan HY. Coexistence of struma ovarii with marked ascites and elevated CA-125 levels: Case report and literature review. Arch Gynecol Obstet 2009;279:753-7.
13. Cooper DS, Ridgway EC, Maloof F. Unusual types of hyperthyroidism. Clin Endocrinol Metab 1978;7:199-220.
14. Lara C, Cuenca D, Salame L, Padilla-Longoria R, Mercado M. A hormonally active malignant struma ovarii. Case Rep Oncol Med 2016;2016:2643470.