CASE REPORT

A Case of Cystic Struma Ovarii: A Rare Ovarian Tumor

Himleena Gautam1, Kamal Kathar2, Papari Goswami3, Alaka Goswami4

ABSTRACT

Background: Struma ovarii is a rare ovarian neoplasm, which contains thyroid tissue. It accounts for less than 5% of ovarian teratomas. These are mostly benign, occurring between 40 years and 60 years of age. Clinical and radiological features are inconclusive and mostly it is diagnosed by histopathology. About 5–8% cases have hyperthyroidism. Cystic struma ovarii is very rare with only 25 cases reported till now. It creates confusion in diagnosis, as even in histopathology, the cells are mostly like those resembling other cystic ovarian tumors with minimal thyroid follicles.

Case description: A 20-year-old girl came with complaints of abdominal discomfort and difficulty in squatting and lying supine. Clinically, a 28-week-size cystic tumor was palpated. Ultrasound showed features of a cystic benign tumor. Tumor markers were normal. Laparoscopic cystectomy was done. Histopathology showed cystic struma ovarii. At 6-month follow-up, the patient has been doing well.

Conclusion: Cystic struma ovarii is a rare diagnosis. Clinical, biochemical, and radiological features do not help. A careful and keen pathological examination is necessary so that thyroid follicles are not left while viewing. There is no clear consensus on follow-up of these patients, but benign tumors usually do not need extensive follow-up.

Clinical significance: It is a rare tumor and a good histopathological diagnosis is needed. Cystic tumors should be carefully examined. A proper correlation between clinical, biochemical, radiological, intraoperative, and histopathological findings may help us to consider this diagnosis when we get similar cases.

Keywords: Case report, Cystectomy, Cystic, Histopathology, Struma ovarii.

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BACKGROUND

Struma ovarii is a rare, mostly unilateral ovarian tumor, which is predominantly (>50%) composed of thyroid tissue. It belongs to the category of specialized or monodermal teratoma.1,2 It accounts for 0.5–1% of all ovarian tumors and 2–5% of ovarian teratomas.3–5 These lesions are mostly benign with around 5–10% cases of malignancy.2 Boettlin first described this highly specialized tumor in 1889. In 1933, Plaut confirmed that follicular cells of struma ovarii and ascites.6 Clinical and biochemical features of hyperthyroidism are uncommon, occurring in less than 5–8% of cases.4,6,7

Struma ovarii can be of solid or cystic type. Cystic type is quite rare, and may be difficult to diagnose due to the presence of minimal quantity of thyroid follicles, thus resulting in confusion with other cystic ovarian tumors.5 Mostly these are multilocular cysts, but about 10% are unilocular.8 Around 25 cases of cystic struma ovarii has been reported.

CASE DESCRIPTION

A 20-year-old female came to outpatient department with complaints of on and off abdomen pain for last 6 months, and discomfort in supine position and difficulty in squatting for last 1 month. The patient has history of delayed developmental milestones. She attained her menarche at 14 years. On examination, a 28-week-size midline cystic lump was felt with restricted mobility. It had smooth surface with a palpable lower pole. Preoperative investigations were normal with CA-125 of 40 IU/mL and thyroid-stimulating hormone of 2.73 IU/L. Ultrasoundography showed a 19.8 cm × 19.8 cm thick walled cyst of left ovary with viscid internal fluid, but with no septation or solid component, thus indicating a benign cyst. All other organs were found to be normal. Laparoscopy was done. A high supraumbilical primary port was given. A 20 cm × 18 cm smooth walled cyst was seen filling up the whole pelvic cavity, with no space for maneuvering the instruments (Fig. 1). On puncturing it with an aspiration needle, greenish yellow viscous fluid was aspirated, which was drained out prior to cystectomy. There was no spillage of cyst contents in the abdominal cavity (Fig. 2). Cystectomy was done. Right ovary looked normal. The cyst was taken out through the 10-mm port. Multiple fine septations were seen within the cyst. On gross examination, it was thought to be a mucinous cyst. However, the HPE report showed that it was a cystic struma ovarii. The biopsy specimen, the pathologist found flattened epithelium in many areas of the specimen. But in one area, thyroid follicles filled with colloid were seen, which were lined by flattened to cuboidal epithelium. Some of these were cystically dilated (Figs 3 and 4).

1,2,4Department of Obstetrics and Gynaecology, Apollo Hospitals, Guwahati, Assam, India
3Department of Pathology, Apollo Hospitals, Guwahati, Assam, India
Corresponding Author: Himleena Gautam, Department of Obstetrics and Gynaecology, Apollo Hospitals, Guwahati, Assam, India, Phone: +91 9678941859, e-mail: himleena@gmail.com


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Discussion

Though most cases of struma ovarii are found in late reproductive age group, there are reports from 16 to 83 years old.11,12 Our patient was 20 years old with a history of symptoms for 6 months. Many cases have no symptoms and are diagnosed incidentally on clinical examination or radiology. Others have nonspecific symptoms with less than 8% cases having hyperthyroidism. Our patient also had nonspecific symptoms such as heaviness, abdominal discomfort in supine, and squatting positions. However, clinically a huge cystic lump was palpable.

Preoperative diagnosis of this condition is very difficult. Ultrasonography may show the presence of “struma pearl,” which is a well-defined solid tissue with a smooth margin that is vascularized on Doppler study.13 On computed tomography scan intracystic lesion of ovarii is highly attenuated on precontrast scans and no or moderate cyst wall enhancement indicates the presence of viscid gelatinous colloid material.6 The classic magnetic resonance image appearance includes multiple intracystic solid areas, representing thyroid tissue, that are of low-signal intensity on T2-weighted images and intermediate signal intensity on T1-weighted images.14 However, these characteristic features are not very easily observed or interpreted on radiologic examination, and they are not specific. In addition, there is no definitive clinical or biochemical marker. Diagnosis is therefore by histopathological examination. However, presence of greenish or brown glary fluid may give a clue.15

Furthermore, struma ovarii causes more diagnostic dilemma when it is of cystic variety, as it is very rare and also sometimes confusing in histopathology. It is characterized by a prominent cystic configuration with minimal thyroid tissue or follicles and presence of nonspecific flat or cuboidal epithelium may mislead the diagnosis with other cystic tumors.6,10,15 Sometimes immunohistochemical staining for thyroglobulin may be required to arrive at a diagnosis.11 In the biopsy report of our patient too, an area showed well-defined thyroid follicles, but in most other places, flattened epithelium was reported.

Conservative surgery (cystectomy and oophorectomy) is recommended for benign struma ovarii especially in reproductive age group. Malignant cases may need radical surgery. Sometimes fertility preservation surgery is done, but these cases should also undergo total thyroidectomy followed by radiiodine...
therapy.\textsuperscript{16,17} Laparoscopic approach is preferred, but the surgeon must be careful not to rupture the tumor intra-abdominally to avoid dissemination.\textsuperscript{18} When laparoscopy is performed and malignant struma ovarii is confirmed postoperatively, a second staging procedure should be performed either via laparotomy or laparoscopically.\textsuperscript{19} Follow-up protocols are not clear in the literature, especially for benign cases. We have followed up the case after 6 months and have called her again at 1 year. However, extensive follow-up protocols are not advised, and probably not necessary.

**Conclusion and Clinical Significance**

Rarity of struma ovarii cases makes its diagnosis difficult. No specific clinical, radiological, and intraoperative findings are present to suggest its diagnosis pre- or perioperatively. It is a histopathological diagnosis. Cystic struma ovarii is very difficult to identify histopathologically because of the scarce thyroid follicles and features like other cystic tumors. Thus biopsy has to be done very keenly to diagnose it. Also in cystic tumors, this differential diagnosis needs to be kept in mind and discussed with the pathologist. It is a benign condition and usually does not need long-term follow-up.

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