

# Intra-operative consult for cystic struma ovarii—An experience in an academic medical center: A study of 53 cases over 21 years

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## ABSTRACT

Struma ovarii often escapes recognition during intraoperative consultation because of its rarity, subtle characteristic gross appearance, and lack of clinical suspicion. An intraoperative diagnosis of benign struma ovarii enables the general gynecologic surgeon to continue the planned surgery. However, a diagnosis of malignancy in a struma ovarii would alter the course of surgery with the involvement of a gynecologic oncology surgeon. We present here that our experience with intraoperative consultation for preoperatively undiagnosed struma ovarii presenting as an adnexal cystic or solid mass at our teaching hospital. Fifty-three cases of struma ovarii, 5.2% of all cystic teratoma of the same period, were diagnosed between January 1991 and March 2011. All intraoperative consultation reports, gross descriptions and final pathology reports were reviewed. The H&E stained slides and in selected cases, immunohistochemistry stained slides, were reviewed. Of the 53 cases of struma ovarii, intraoperative consultation was requested on 48 cases. Frozen section was done on 24 cases and only gross examination was felt appropriate in remaining 24 cases. 83% cases were diagnosed when a frozen section was done. None of the remaining 24 cases were recognized as struma by gross inspection. Our findings reveal that in a large number of cases the diagnosis of struma ovarii remained unrecognized during intraoperative consultation, indicating its often subtle/deceptive gross morphologic appearance. However, the purpose of the intraoperative consultation was served, as appropriate information was provided to the surgeon to guide the surgical manage-

ment.

**Keywords:** Struma Ovarii; Intraoperative Consult; Ovarian Cyst

## 1. INTRODUCTION

Struma ovarii, the most common monodermal teratoma, is a relatively uncommon tumor and comprises less than 3% of all ovarian teratomas [1]. Because of the rarity of this tumor, only two large series have been published so far in the English literature, both by Scully and his colleagues [2,3]. Another series of proliferative and histologically malignant struma ovarii was published by Tavassoli and her colleagues [4]. Most others are single case reports. Several other reports on various features of strumal carcinoid are available in the literature [5,6].

The term “struma ovarii” is reserved for a teratoma which is composed entirely or predominantly of thyroid tissue. The tumor is usually unilateral. However, there is one report in which the contralateral ovary with a teratoma contained thyroid tissue [7]. Struma ovarii has been reported to be present in association with other neoplasms. Association with mucinous cystadenoma [8], Brenner tumor [9] was reported. The other potential pitfall of struma ovarii, especially on frozen section, is that clear cell carcinoma may mimic struma ovarii [10]. The tumor may present as a solid or cystic lesion. When predominantly cystic, it may be unilocular or multilocular. When the cyst is large, the epithelial lining of the wall may become attenuated, making the diagnosis difficult on hematoxylin & eosin stained slides. Immunohistochemical staining for thyroglobulin is required to confirm the diagnosis in these cases [3]. The deceptive gross appearance, similarity to other benign ovarian lesions,

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and lack of preoperative clinical suspicion appears to be the reasons precluding recognition during intraoperative consultation. We describe here our experience with intraoperative consultation on adnexal masses without any preoperative diagnosis and 53 cases of struma ovarii diagnosed incidentally in our teaching hospital during the past 21-year period.

## 2. MATERIALS AND METHODS

A computer search identified all cases of struma ovarii from the files of the Department of Pathology, Women & Infants' Hospital of Rhode Island, from January 1991 and March 2011 after obtaining approval from the Institutional Review Board (#13-0005). Also identified were all cases of cystic teratomas diagnosed during the same time period. The frequency of struma ovarii among cystic ovarian teratomas was determined. All intraoperative consultation reports and final diagnoses were compiled. The gross description of the tumors and the association of struma ovarii with other tumors were noted. Hematoxylin and eosin stained slides and, when available, immuno-histochemical stained slides, were also reviewed. Only those teratomas consisting entirely or predominantly of thyroid tissue were included in the study as struma ovarii. Mere presence of thyroid tissue in a teratoma did not fulfill the diagnostic criteria, and were excluded. The radiological impression when available and pertinent clinical features were noted.

## 3. RESULTS

A total of 53 cases of struma ovarii and 1019 cases of cystic teratomas were identified during the 21-year period. Struma ovarii constituted approximately 5.2% of all mature cystic teratoma. The tumors occurred in women 12 to 81 (median 45) years of age. Sixteen patients were post menopausal, 29 patients were premenopausal and 8 patients were perimenopausal. Of the 53 cases, 50 were benign tumors and three strumal carcinoid tumors were diagnosed. All patients presented with an adnexal mass. Twenty nine of 53 cases involved the left ovary, 22 involved the right ovary and two cases had bilateral involvement. Thirty two of 53 cases were cystic and 2 cases were entirely solid and the remaining 19 cases had solid and cystic areas. Intraoperative consultation was sought for 48 cases; the remaining 5 cases were not seen perioperatively. Three tumors were associated with fibroma and one Brenner tumor respectively. Three others which were thought to be associated with serous cystadenoma and later were found to stain positively with thyroglobulin, indicating a cystic component of struma ovarii. All 48 cases were examined grossly and frozen sections were performed on 24 cases when it was deemed necessary by the pathologist. In one case, despite

the gross impression of serous cystadenofibroma, a frozen section was performed, which revealed struma ovarii. Twenty of 24 (83%) cases were diagnosed accurately by frozen section and none of the remaining 24 cases were recognized by gross inspection during intraoperative evaluation. The grossly examined cases were thought to be benign cysts in 21 cases and 3 as serous or mucinous cystadenoma/adenofibroma. One case on frozen was thought to be granulosa cell tumor, later confirmed by immunohistochemical stain as struma ovarii. The gross appearance, opinion of the radiologist, gross impression and frozen section diagnosis of all 53 cases are tabulated in **Table 1**.

## 4. DISCUSSION

The typical textbook description of struma ovarii refers to a unilateral tumor of varying size, usually measuring less than 10 cm in diameter. When associated with other elements, the size may be larger [11]. The exterior surface is smooth, although fibrous adhesions are not unusual. Upon sectioning, the cut surface may be entirely cystic or solid or both. The thyroid tissue is usually brown or green brown, predominantly solid, and gelatinous [12]. Hemorrhage, necrosis and foci of fibrosis may be present. Solid tumors are less glistening and fleshy.

In our review, we found a variety of gross appearances of struma ovarii, the majority of which did not fit the typical description found in the literature. Thirty two of 53 cases (60%) in our study were entirely cystic. The majority of these were multilocular and 4 (19%) were unilocular. Nineteen cases (36%) were cystic and solid and only two (3%) were entirely solid. Typical greenish or brown green cysts were present in only 4 (7.5%) cases. A gelatinous appearance was noted in 7 (13%) cases either in association with greenish cysts or in isolation. The characteristics of the cyst contents were also variable. The cyst contents in 18 of 53 cases (34%) were found to be clear or yellowish/serous; in 4 (7%) hemorrhagic; and in 1 (0.4%) case, mucinous. The appearance of the remaining 3 cyst contents was not known to us as the cysts were either collapsed when received or taken out in pieces during surgery. Of the 4 tumors with hemorrhagic cyst contents, only one was due to torsion. The reason for hemorrhage within the cyst was not clear in the other 3 cases. The two entirely solid tumors were thought to be a non-epithelial ovarian tumor on gross examination. However, after frozen section, a differential diagnosis of struma ovarii versus strumal carcinoid was rendered. It was felt during intraoperative consultation that additional sampling and special stains were necessary to confirm the diagnosis.

Absence of the typical gross appearance resulted in a failure to identify a significant number of cases of struma

**Table 1.** Master table with gross and intraoperative impression.

Case #	Age (yrs)	Radiology	Intraoperative consult		
			Gross Appearance	Gross Impression	Frozen section
1	33	Adnexal mass	19 cm unilocular cyst, left ovary, filled with yellowish fluid and a small mucoid area with greenish coloration.	Benign cyst, probably mucinous cystadenoma	Not done
2	38	Cystic adnexal mass	3 cm multilocular cyst, left ovary, smooth surface and filled with clear/mucinous fluid. No solid area	Benign cyst	Struma Ovarii
3	33	Un-homogeneous adnexal mass	6.5 cm multilocular cyst, left ovary, with smooth purplish surface and filled with yellow viscid mucin	O.R. consult not requested	NA
4	49	Not available	4.7 cm multilocular cyst, right ovary, one small cyst filled with sebaceous material	Cystic teratoma	Struma Ovarii
5	69	Not available	6.5 cm multilocular cyst, right ovary, with white pink surface and filled with yellowish, soft, gelatinous material, and focal calcification	Cystic teratoma	Struma Ovarii
6	45	Complex cystic adnexal mass	7.5 cm multilocular cyst, right ovary, with unremarkable surface and lined by blood clot. Gritty on cut section.	Benign cyst? teratoma	Struma Ovarii
7	65	Complex adnexal mass	6.5 cm multilocular cyst, right ovary, with unremarkable surface and filled with serous fluid and a small dark-brown gelatinous nodule	Serous cyst vs. Cystic teratoma	Struma Ovarii
8	37	12 cm adnexal mass	11 cm unilocular cyst, left ovary, with purple-maroon, smooth surface and filled with hemorrhagic material	Torsed benign cyst	Not done
9	19	Cystic and solid adnexal mass	10 cm multilocular cyst, left ovary, with tan-pink surface and congested vessels. Filled with hemorrhagic fluid. No solid area.	Benign multilocular cyst	Not done
10	24	11 cm cystic adnexal mass	13 cm multilocular cyst, left ovary, greenish surface and filled with yellowish/greenish fluid and congested vessels	Benign cyst	Not done
11	23	9.3 cm complex adnexal mass	10 cm multilocular cyst, right ovary, with unremarkable surface, and a 2 cm tan papillary nodule containing greenish cysts	Cystic teratoma Vs. Struma ovarii	Struma Ovarii
12	64	3.8 cm cystic and solid adnexal mass	4.5 cm unilocular cyst, right ovary, with unremarkable surface and filled with yellowish fluid. A 2 cm gelatinous nodule is present in one cyst	Benign cyst	Struma Ovarii
13	32	Pelvic mass and bilateral ovarian cysts	4.5 cm unilocular cyst, right ovary, with unremarkable surface and filled with yellowish fluid. A 2 cm gelatinous nodule is present in one cyst	O.R. consult not requested	NA
14	27	14 cm complex adnexal mass	8 cm cyst walls, in aggregate, right ovary, taken out in pieces with hemorrhagic wall. A 3 cm yellowish nodule present	Benign hemorrhagic cyst	Struma Ovarii
15	49	Left adnexal cyst	7.5 cm cystic and solid mass, left ovary, filled with yellow-green fluid. A 3 cm solid, tan-pink area is present	Benign serous cyst	Serous cyst and Struma ovarii
16	64	Pelvic mass	11 cm multilocular cystic and solid, left ovary, with tan bosselated area on surface. Cysts filled with yellow serous fluid and solid area is tan and fleshy	Serous cystadenofibroma	Struma Ovarii
17	28	Ovarian cyst	6.6 cm multilocular cyst, left ovary, with translucent wall and filled with yellowish fluid. Focally gritty to cut	Benign cyst	Struma Ovarii
18	33	Not available	8 cm rubbery to firm, solid tumor, left ovary, with yellowish surface. The cut surface is yellowish-brown, lobulated, finely granular and hemorrhagic center.	Non-epithelial tumor	D/D: Struma Vs. Carcinoid
19	36	Not available	14 multilocular cyst, left ovary, with yellowish surface and filled with hemorrhagic fluid	Benign cyst	Not done
20	64	Not available	11 cm multicystic and solid lesion, right ovary, with small nodules and hemorrhagic and fibrosis. Cysts are filled with yellowish fluid and solid fibrotic areas are intermixed with hemorrhage, necrosis and often flesh looking.	Cystadenofibroma	Cystadenofibroma
21	65	Not available	6 cm multilocular cyst, right ovary with smooth surface and filled with clear fluid	Serous cystadenoma	Not done
22	49	Ovarian mass	5.5 cm multilocular cystic and solid mass, left ovary, with glistening surface. Cysts filled with yellowish fluid and solid areas are yellowish with focal dark brown coloration	Cystic teratoma vs. Struma Ovarii	Struma Ovarii

## Continued

23	44	Ovarian mass	30.5 cm unilocular cyst, left ovary, filled with serous fluid	Serous cystadenoma	Not done
24	44	13.9 cm complex cyst	13 cm unilocular cyst, right ovary.	Hemorrhagic cyst	Not done
25	66	Pelvic mass	13 cm multicystic and solid mass left ovary. Cysts are filled with clear yellow fluid and solid areas are gelatinous.	Cystadenoma	Not done
26	46	14 cm left ovarian cyst	15 cm solid and multicystic lesion, left ovary. Cysts are filled with bloody fluid and solid tan area.	Benign cyst	Struma Ovarii
27	48	? Uterine mass	5 cm unilocular cyst, left ovary	Benign hemorrhagic cyst	Not done
28	31	Right ovarian cyst	5 cm cystic right ovary with occasional solid areas. Some cysts are filled with sebaceous materials and hairs	Benign cyst	Not done
29	49	Left adnexal mass	4 cm unilocular cyst filled with chocolate colored fluid. No solid area	Cystic teratoma	Not done
30	12	Ovarian mass	4.5 cm multicystic lesion, left ovary. Few cysts are filled with sebaceous material and hair	Benign hemorrhagic cyst	Not done
31	31	Pelvic mass	5 cm cystic lesion, right ovary, received in pieces.	Cystic teratoma	Not done
32	46	Pelvic mass	12 cm solid and cystic mass. Solid areas are tan, gelatinous, Left	Benign cyst	Not done
33	49	Left ovarian mass	5.5 cm multicystic and solid lesion, left ovary. Solid areas are maroon colored and cysts are hemorrhagic	? Struma ovarii	Struma Ovarii
34	64	Pelvic mass	11 cm solid and multi-cystic lesion, right ovary. Cysts are filled with clear fluid and solid areas hemorrhagic, somewhat fleshy and fibrotic	Adenofibroma	Not done
35	36	10 cm Pelvic mass	14 cm multilocular cysts, left ovary. Cysts are filled with hemorrhagic fluid	Benign cyst	Not done
36	81	Pelvic mass	21 cm multi-cystic lesion, Left	Ovarian cyst	Struma Ovarii
37	41	Right pelvic solid mass	9.3 cm solid ovarian mass. Cut surface is lobular and yellowish-tan granular with occasional cystic areas filled with waxy material.	?Malignant tumor	D/D: Sex Cord Vs. Germ cell tumor
38	49	Right ovarian cyst (? Dermoid)	6.5 cm multicystic lesion, right ovary. Occasional cysts are filled with sebaceous material and hair. A 1.5 cm pink nodule is present	Cystic teratoma	Not done
39	45	10 cm pelvic mass	18 cm solid and cystic tumor, right ovary. Solid part is yellowish tan with hemorrhage.	Neoplasm	Granulosa cell tumor
40	54	Right pelvic mass; ? Dermoid	8.5 cm multicystic right ovary. Occasional cysts are filled with sebaceous material. Solid fibrotic areas noted.	Mature cystic teratoma	Not done
41	37	Bilateral complex pelvic mass	3.5 × 2.5 cm cyst with thin wall	Favor dermoid cyst	Not done
42	54	Lobulated complex mass	18 × 13 cm cyst, multiloculated, Left	Favor dermoid cyst	Not done
43	58	Cystic pelvic mass	Cyst, 3.5 × 2.8 cm, right	Benign cyst	Cystic teratoma
44	69	Pelvic Cyst	9 × 7 cm cystic, left	Benign cyst	Struma Ovarii
45	66	Complex echogenic mass	4.5 × 3 cm solid mass, right	Primary vs. metastatic tumor	Carcinoid tumor
46	41	Mass, ? Teratoma	5 × 4 cm multicoated cyst, right	Dermoid cyst	Not done
47	42	Adnexal mass	11 × 10 cm cystic left ovary	Benign cyst	Not done
48	53	Complex ovarian cyst	7.5 × 4.5 cm Cystic, Left	O.R. consult not requested	NA
49	22	Cystic ovarian mass	5 × 2.5 cm, cystic, Right	O.R. consult not requested	NA
50	41	Ovarian cyst	3.5 × 2.5 cm, Cystic, Left	O.R. Consult not requested	NA
51	49	Ovarian cyst	6.5 × 5.2 cm Cystic tumor	Benign cyst	Struma Ovarii
52	81	Right pelvic mass	16 × 11 × 4 cm. The inner lining is yellowish/tan and smooth. Small cysts with yellow gelatinous material	Benign cystadenoma	Not done
53	22	6 cm ovarian cyst	Cystic and solid tumor with yellow-white plaque, Left	Carcinoid vs. Struma ovarii	Strumal carcinoid

ovarii during the intraoperative consultation. In our experience, 20 of 24 (83%) cases were recognized when frozen section was done. However, none of the remaining 24 cases was recognized as struma ovarii when examined by gross inspection only. A significant number, 21 of 24 (88%) were thought to be benign cysts and remaining 3 of 24 (12%) cases were thought to be serous or mucinous cystadenoma on gross examination and no frozen section was performed. Three cases (14%) were gritty to cut and felt to represent components of a mature cystic teratoma intraoperatively. On final sections, however, these were found to be calcifications in a struma ovarii.

Intraoperative consultation was requested in view of absence of preoperative diagnosis, as all cases were presented as adnexal masses of varying size. While performing the intraoperative consult some cases were diagnosed by gross impression only and others after a frozen section. Frozen section was performed in the latter cases to rule out any malignancy or in a few cases, at the request of the surgeon. In our experience, performing a frozen section may not always resolve the diagnostic dilemma, but often may help increase the diagnostic accuracy, as was found in one of our cases. Despite the gross appearance of a serous cystadenofibroma, a frozen section was performed and the diagnosis of struma ovarii was made. The only solid case in our series was felt to be either a struma ovarii or strumal carcinoid during intraoperative consultation. Frozen section examination did not resolve this issue. Frozen section of another case revealed serous adenofibroma, but additional permanent sections revealed struma ovarii. Presence of extensive fibrosis and selection of an inappropriate area in the tumor led to the erroneous diagnosis during intraoperative consultation. Two tumors were diagnosed on permanent hematoxylin and eosin stained section as serous cystadenoma, but later found to represent cystic struma ovarii. Both these two tumors were predominantly cystic with minor component suggestive of glands containing colloid. Thyroglobulin staining by immunohistochemistry is essential to confirm struma ovarii in these suspected cases. Special stain was performed to confirm that the larger cysts were also component of struma ovarii as suggested by Szyfelbein, *et al.* [3]. One also must pay attention not to miss clear cell carcinoma as it may mimic struma ovarii [10]. Association of struma ovarii with Brenner tumor is rare and one such case was identified in our series. Association with fibroma was not previously reported in the literature. We had one such association in our series.

It is interesting that all cases in our series were benign tumors in contrast to earlier reports. A possible explanation could be that all the current cases are hospital cases only, where as, the other published reports included con-

sultation cases.

The radiological impression of most of the cases was a complex adnexal cyst, which did not help the pathologist or the surgeon to arrive at a diagnosis of struma ovarii preoperatively. Struma ovarii, being a teratoma, could potentially be a bilateral tumor. Its proper recognition during intraoperative consultation may help the general surgeon to seek consult of a gynecologic oncologist to evaluate the contralateral ovary more closely. A diagnosis of benign cyst does not convey the appropriate message to a general surgeon. It is reported in the literature that the likelihood of a cystic teratoma being bilateral is 8% - 15% [13], while only 2% - 3% of mucinous cystadenomas and 12% - 20% of serous cystadenoma were bilateral [14]. In our series, one patient had bilateral teratoma at presentation (Case 13) and in another patient (Case 17) the contralateral ovary was found to be involved 6 years later. We believed the tumor in the contralateral ovary was not recognized during her initial surgery as struma ovarii was not diagnosed during intraoperative consultation and the finding may have been overlooked.

An accurate intraoperative diagnosis of benign struma ovarii is somewhat an academic exercise as it does not provide any crucial information to the surgeon so that he has to change his course of surgery. However, it is important for the surgeon to know that the tumor is benign. It is also important that struma ovarii should be differentiated from clear cell carcinoma on frozen section as the management is different in those two tumors. Despite the potential pitfall none of the cases in our series was over diagnosed as clear cell carcinoma.

It is worth mentioning here that none of the 53 cases had any preoperative diagnosis and intraoperative consultation was requested to arrive at a diagnosis and to rule in/out malignancy.

## 5. CONCLUSIONS

We have observed in our study that the diagnosis of struma ovarii escaped recognition in half of the cases during intraoperative consultation. It appears that doing frozen section improves the diagnostic accuracy of struma ovarii significantly when compared with gross examination.

In our series, we feel that the purpose of intraoperative consult was served as malignancy was excluded and useful information was provided to the surgeon in the operating room.

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