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Clinical and Histopathological Study of Struma Ovarii: A Rare Ovarian Tumor

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Abstract Struma ovarii or monodermal teratoma is a specialized ovarian neoplasm which mainly constitutes mature thyroid tissue. It is a rare tumor which comprises 1% of all ovarian tumors and 2.7% of all dermoid tumors. Thyroid tissue can be observed in 5-15% of dermoid tumors but to designate the tumor as struma ovarii, it must comprise more than 50% of the ovarian tissue. This study was conducted in the Department of Pathology, Manipal Teaching Hospital in Pokhara, Nepal over a period of 10 years (Jan 2006 to Sep 2015). Age, clinical findings, pre-operative imaging diagnosis, size and side of the tumor, gross and microscopic findings along with type of surgery performed are included in the study. During this 10 years period, there were 7 cases of struma ovarii with age ranging from 26 to 56 years. 2 cases had tumor on the right and 4 cases had tumor on the left side while 1 case had bilateral struma ovarii. Initial presenting symptom was palpable mass, abdominal pain and vaginal bleeding. The size of the tumor ranged from 4 to 15 cm. The capsule was smooth and cut surface shows multiloculated cyst filled with greenish to pale brown gelatinous thick fluid. Microscopic examination revealed well encapsulated tumor composed entirely of thyroid follicles. Diagnoses of struma ovarii were made in all cases. The preoperative imaging may not exactly give the diagnosis. Clinically, lesser age group was more affected and left side is more commonly involved in our series, in contrary to other literature. Out of 7 cases, bilateral struma ovarii was seen in 1 patient. No malignant features were seen in any of these cases.

Keywords: *monodermal teratoma, ovarian tumor, struma ovarii*

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1. Introduction

Teratoma is neoplasm of germ cell origin arising from primordial germ cells and is composed of more than one germ cell layer structures. It can be divided into mature, immature and monodermal depending on its constituents. Monodermal teratomas are specialized ovarian neoplasms and include struma ovarii and carcinoid. Struma ovarii is the expression of dominant growth of thyroid tissue in a teratoma sometimes leading to even exclusion of all other components [1,2,3]. It is a relatively rare tumor which comprises only 1% of all ovarian tumors and 2.7% of all dermoid tumors [4]. Here, we describe clinico pathological features of 7 cases of struma ovarii and discuss relevant current literature.

2. Material and Methods

This is a retrospective hospital based study conducted at the Department of Pathology in Manipal Teaching Hospital, Pokhara, Nepal over a period of 10 years, from January 2006 to September 2015. All cases of struma ovarii reported on histopathology were retrieved and included in the study. Tissues were processed routinely and H&E stains were used in all cases.

3. Results

Table 1. Clinical Findings and Preoperative Imaging Diagnosis in All Cases

	No of patients	%
Initial Clinical Presentations		
Palpable Mass	5	71.4
Abdominal Pain	3	42.9
Vaginal Bleeding	1	14.3
Preoperative Imaging Diagnosis		
Dermoid Cyst	4	57.1
Benign Multiloculated Cyst	2	28.6
Struma Ovarii	1	14.3

During this period, 26,234 histopathological specimens were received which included 347 cases of ovarian tumors, out of which 140 cases were teratomas. A total of 7 cases of struma ovarii were reported. The age ranged from 26 to 56 years. Initial presenting symptoms were palpable mass

in 5 cases, abdominal pain in 3 cases and vaginal bleeding in 1 case. Preoperative imaging study findings suggested the diagnosis of dermoid cyst in 4 cases, benign multiloculated cyst in 2 cases and struma ovarii in 1 case (Table 1).

Routine preoperative check up was normal in all patients. 4 patients had left sided tumor and 2 patients had right sided tumor while 1 patient had bilateral struma ovarii. Total abdominal hysterectomy with bilateral salphingo-oophorectomy was performed in 5 cases while unilateral oophorectomy was done in 2 cases (Table 2). Operative and postoperative periods were uneventful in all 7 cases.

Table 2. Details of Individual Case Including Side Involved, Surgery Performed, Gross Tumor Size and Specific Microscopic Findings

Case No	Age (years)	Side	Surgery Performed	Gross Tumor Size (cm)	Microscopic Findings
1	56	Left	TAH with BSO	7	Thyroid follicles with cystic change
2	31	Left	TAH with BSO	12	Thyroid follicles with hemorrhage and hemosiderin laden macrophages
3	46	Right	TAH with BSO	15	Thyroid follicles with cystic change
4	48	Left	TAH with BSO	11	Thyroid follicles with cystic change
5	26	Right	Right ovarian cystectomy	12	Thyroid follicles with cystic change
6	36	Left	Left Ovarian Cystectomy	14	Thyroid follicles only
7	39	Bilateral	TAH with BSO	Right- 4 Left- 13	Thyroid follicles only

TAH with BSO: Total Abdominal Hysterectomy with Bilateral Salphingo-Oophorectomy

On gross, the size of the tumors ranged from 4 to 15 cm. The capsules were intact, smooth and grey white. Bosselation was seen in 5 cases with tumors larger than 10cm. Cut surface showed multiloculated cyst filled with greenish to pale brown gelatinous thick fluid (Figure 1).

Figure 1. Cut surface of the ovary showing multiloculated cyst filled with thick pale brown gelatinous material.

Multiple sections were studied in each case. Under microscopic examination, all cases showed well encapsulated tumor composed entirely of thyroid follicles lined by benign cuboidal to flattened epithelium and filled with colloid (Figure 2).

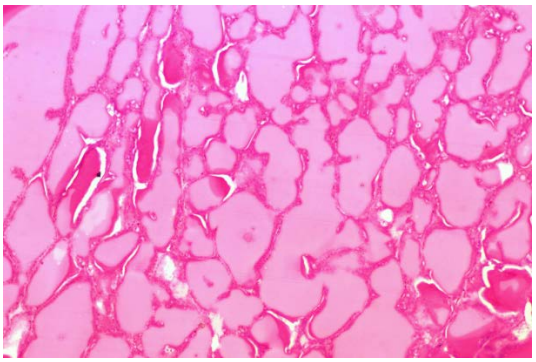

Figure 2. Microscopic picture showing thyroid follicles of varying sizes filled with colloid. (H&E, x100)

Thyroid follicles were separated by thin fibrous septae. Diagnosis of struma ovarii was made in all cases. In only one case, ectodermal tissues were seen composing less than 10% of the tumor. Post-operative retrospective thyroid function tests were done in all cases which were normal.

4. Discussions

Struma ovarii was first described by Boettlin in 1889 who observed the presence of thyroid tissue in ovaries and further reports were published by Von Kalden in 1895, Gottschalk in 1899 and Mayer in 1903. [5,6] However as it comprises only 1 % of all ovarian tumors, there has been paucity of DATA in literature. [7,8] Thyroid tissue is observed in 5-15% of ovarian teratomas but to qualify as struma ovarii, the proportion of thyroid must comprise more than 50% of overall tissue. [9] Due to scarcity of available data, no definite racial predilection of this tumor has been determined. In our series, struma ovarii comprised 5% of all dermoid cases. Yoo SC et al and Khediri Z et al in their series, reported struma ovarii cases to be 4.8% and 8.57% of all dermoid cysts respectively. [10,11] Struma ovarii occurs usually after the age of 40yrs. The peak age incidence of age is reported to be in 5th decade but few cases have also been reported in prepubertal and postmenopausal women. [1,11,12] This tumor is seen in only 17.6% of cases in patients below 30 yrs. [10] In this study, only 3(42.9%) cases were above 40yrs while 3(42.9%) cases were between 30 to 40 yrs and one case (14.3%) was of 26 yrs. Present DATA seem to be in contrary with the past DATA but corroborate with Khediri Z et al, who in their report of 3 cases, reported only 1 case each of 17 yrs, 31 yrs and above 40yrs olds. [11]

Large majority of struma ovarii cases present without any symptom. Yoo SC et al in their study noted that 41.2% cases had no presenting symptoms and tumors were discovered during routine ultrasound check up. [10] Clinical symptoms if present are mostly non-specific and include palpable abdominal mass, lower abdominal pain, vaginal bleeding, ascites, hydrothorax as well as in some cases elevated thyroid function and rarely thyroid neoplasms. [12,13,14,15] In the study, all patients presented with non-specific symptoms which is in accordance with other series. [10,11] In literature, 5-8% of cases with struma ovarii may have features of thyroid hyperfunction. [1,12,16,17] Kaur S et al presented a case of struma ovarii with preoperative high levels of T3 and

T4. [12] However in the study, 1 case who was radiologically diagnosed as struma ovarii had undergone thyroid function test which was within normal limit. Other patients did not have any hyperthyroid symptoms, so preoperative thyroid function tests were not carried out. However postoperative thyroid function test were normal. In the series of 34 cases presented by Yoo SC et al, preoperative thyroid function tests were not carried out in any case as there were no thyroid related symptoms.

USG is useful tool to diagnose and classify ovarian masses but is helpful in giving diagnosis of struma ovarii in only about 11.8% cases. [10] Preoperative imaging diagnosis includes dermoid cyst, endometrioma, benign cyst, normal ovary, malignancy and non- diagnostic in different studies. [10,11] In the study, only 1 case was radiologically suggested as struma ovarii while 4 cases were suggested as dermoid cysts and remaining 2 cases were suggested as benign multiloculated cyst. In struma ovarii, MRI typically shows multilocular cystic mass which shows variable signal intensity within the loculi. Some loculi show low intensity on T1 weighted images and very low intensity on T2 weighted images, corresponding histopathologically to gelatinous colloid material. [18] There was no case in the study who had undergone preoperative MRI. Thus, most cases of struma ovarii do not have definite clinical or imaging diagnostic features that differentiate it from other ovarian tumors. [11]

Most of the cases in literature are unilateral and right side is more commonly involved. [1] Khediri Z et al and Singhal S et al in their studies found all cases of the tumor on the right side. [1,11] Interestingly in this study 4 left sided cases, 2 right sided case and 1 case with bilateral struma ovarii were found. In literature, usually the size of the tumor varies from 0.5 to 10 cm. [1,19] Majority of the cases (75%) in this study presented with tumors greater than 10 cm. Grossly, they may appear as entirely cystic structure containing brown to green gelatinous material as it was found in 6 cases or as a nodule in a dermoid cyst as in one case of the study. [20] Microscopically it is known to be composed of normal or hyperplastic thyroid tissue and may show thyroid related changes including microfollicular adenoma, macrofollicular adenoma, trabecular pattern, solid pattern, oxyphilic cells and clear cells. [19] In this study, 4 cases showed cystic changes and 1 case showed hemosiderin laden macrophages. No papillary areas, nuclear atypia and Hurthle cell changes were seen in any of this study cases.

Bilateral struma ovarii is very rare and may be seen in only 6% of cases. [21] Rana V et al reported a case of bilateral non- functional struma ovarii in 70yrs old female who presented with pseudo-Meigs' syndrome. [22] We had a single case (14.3%) of bilateral struma ovarii in 39 yrs old female. The right ovary measured 4.5x4cm showing partially cystic areas and the left ovary measured 13x10 cm with multiloculated cyst filled with greenish to brownish gelatinous colloid like material. Malignant transformation of struma ovarii is rare (5-10%) which may be of papillary, follicular or mixed pattern. Papillary carcinoma is the commonest comprising 85% of malignant cases which shows characteristic ground glass nuclei. However, follicular carcinomas are difficult to diagnose as the struma ovarii lacks the thick capsule. [19,23] Tumors showing the morphology of papillary or follicular cancer and extra-ovarian spread of the lesions deserve the designation of malignant struma ovarii. Factors for the recurrence of tumor include the size of the tumor, ascites or adhesions during presentation or solid architecture. Presence of mitotic figures or vascular invasions is not very helpful in predicting prognosis. [19] Surgical resection is the treatment of choice in cases of struma ovarii. For women desiring further pregnancies, simple cystectomy or unilateral oophorectomy is the optimal treatment. [11] In the present study, 5 cases underwent total abdominal hysterectomy with bilateral salpingo–oophorectomy whose families were complete while 2 cases who underwent simple cystectomy who were young females.

5. Conclusion

Due to rarity of struma ovarii, detailed information of this entity is not easily available. Here, the clinical and pathological data of 7 cases have been presented. Preoperative imaging may not exactly give the diagnosis. Clinically, lesser age group was more affected and left side was more commonly involved in the study, in contrary to other literature. Out of 7 cases, bilateral struma ovarii was seen in 1 patient. No malignant features were seen in any of these cases.

Declaration of Conflicting Interests

The authors declare that there is no potential conflicts of interest with respect to the research, authorship and /or publication of this article.

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