

A Case of Struma Ovarii Diagnosed by Cytology during Laparoscopic Surgery

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Abstract

We report a case of pure struma ovarii tumor diagnosed by cytology during laparoscopic surgery. The patient was a 34-year-old Japanese woman, gravida 1, para 1, who had the left adnexal mass, and was pre-operatively diagnosed as left ovarian endometriotic cyst or mature cystic teratoma by magnetic resonance imaging findings. She underwent laparoscopy, and the content of the left ovarian cystic tumor was found to be yellow gelatinous material, suggesting mature cystic teratoma. The imprint cytology of the tumor showed benign glandular pattern, suggesting struma ovarii. Histopathological findings led us to the diagnosis of pure struma ovarii with positive reactions for thyroglobulin and thyroid transcription factor-1. No metastases or disseminated lesions were detected. The patient has no recurrent signs 7 months after the operation.

Keywords

Struma Ovarii, Cytology, Immunocytochemistry, Immunohistochemistry, Thyroglobulin, Thyroid Transcription Factor-1

1. Introduction

Struma ovarii is a rare type of ovarian tumor that has been reported to represent 0.5% - 1.0% of all ovarian tumors [1] [2] [3]. This tumor is the most common type of monodermal teratoma and comprises approximate 3.0% of all ovarian teratomas [1] [2] [4] [5]. It is defined as histologically characterized by the presence of thyroid-like glands at least 50%. Malignant transformation of the struma

ovariorum, which is associated with *BRAF* mutations and *RET/PTC* rearrangements [6] [7], as seen in papillary thyroid carcinoma of the eutopic thyroid, rarely occurs and its rate is reported to vary, ranging from 5% - 10% [8]. Struma ovarii is predominantly found in women between the ages of 40 and 60 years [9]. While magnetic resonance imaging (MRI) analysis has been reported to be useful for the differential diagnosis [10] [11] [12], the preoperative differential diagnosis between the struma ovarii and other malignant tumor is difficult due to the morphological similarities. In fact, only a few reports concerning cytological diagnosis of struma ovarii have been reported [13] [14], although immunocytochemistry might be a valuable tool for accurate diagnosis of the struma ovarii [14]. We present here a rare case of struma ovarii in a 34-year-old woman, and the imprint cytology during the laparoscopic surgery was useful for the diagnosis of the neoplasm. Immunocytochemical and immunohistochemical examinations of thyroglobulin were useful to confirm the above diagnosis.

2. Case Report

A 34-year-old Japanese woman, gravida 1 para 1 with normal menstruation cycle, was referred to the Department of Obstetrics & Gynecology, Gujo City Hospital, because of the left adnexal mass and secondary infertility introduced from a fertility clinic. Her sociodemographic and clinical characteristic of the patient was summarized in **Table 1**. Her medical history included her left ovarian cystectomy under a laparoscope at 32-year-old in other hospital in other prefecture. However, the patient and her family did not know the detailed information. Other diseases including thyroid tumor were not reported. Hyperthyroidism could not be detected. Magnetic resonance imaging (MRI) in our hospital revealed the left ovarian tumor containing cystic and solid parts, measuring $6.8 \times 5.2 \times 7.0$ cm. The cystic part showed variable signal intensity on the T2-weighted MRI scan (**Figure 1(A)**) and slightly high on T1-weighted (**Figure 1(B)**), suggesting the content being bleeding or gelatinous (highly viscous) and colloidal material. MRI examination did not detect enlarged lymph nodes, ascites and disseminated lesions in the peritoneum. Pelvic ultrasonography (USG) showed right polycystic ovary (**Figure 2(A)**) and left ovarian cystic tumor with papillary protrusions (**Figure 2(B)**) before operation. Clinical laboratory data, including tumor markers (CA125, 7.2 U/ml; CA19-9, 8.6 U/ml) showed within normal limitations. Based on the above findings, our working diagnosis was left ovarian endometriotic cyst or mature cystic teratoma.

Under the working diagnosis, the patient underwent laparoscopy one month after her first visit. She received laparoscopic left oophorectomy (**Figure 3**) and the tumor dropped into the ENDOPUCH®(Ethicon, TX) without rupture. Her right ovarian drilling was also performed for trigger ovulation in this patient with polycystic ovary syndrome, suggested by the pelvic USG before surgery (**Figure 2(A)**). Electrocautery or a laser is used to destroy parts of the ovaries. The content of the cystic tumor was yellowish gelatinous fluid with approximate

150 ml, suggesting a mature cystic teratoma. There was no peritoneal fluid (ascites). On the imprint cytology of the tumor, the tumor cells showed glandular pattern. The tumor was thus suggested to be struma ovarii arising from a mature cystic teratoma (**Figure 4(A)**), and immunocytochemistry performed after the operation revealed that the tumor cells were positive for thyroglobulin (**Figure 4(B)**). The patient made uneventful recovery, and was discharged on the post-operative third day. Pelvic USG revealed that the number of right ovarian follicles decreased (**Figure 2(C)**) and left ovarian tumor disappeared in her left adnexal lesion (**Figure 2(D)**).

The left ovarian mass having cystic and solid parts (**Figure 5**) was fixed in 4% buffered formalin, routinely processed, and embedded in paraffin for histological and immunohistochemical examinations. After which 3 - 4 µm-thick sections were stained with hematoxylin and eosin (H & E) for pathological diagnosis. In addition, immunohistochemistry using DAKO monoclonal antibodies, such as thyroglobulin (1:200 dilution, DAKO), thyroid transcription factor (TTF)-1 (1:200 dilution), inhibin (1:50 dilution) and calretinin (1:2000 dilution) was performed.

Histopathologically, the tumor consisted of normo- and macro-follicular thyroid tissue (**Figure 6(A)** and **Figure 6(B)**). Other components derived from germ layers except for thyroid tissue were also found. No atypical and/or immature cells were also recognized. Immunohistochemistry showed that glandular cells were positive for thyroglobulin (**Figure 6(C)**) and TTF-1 (**Figure 6(D)**), while they were negative for inhibin nor calretinin. Our final diagnosis was a pure struma ovarii in the left ovary. The patient has no recurrent signs 7 months after the operation.

3. Discussion

In the present case, the imprint cytology and immunocytochemical and immunohistochemical examinations for thyroglobulin and TTF-1 were useful for the diagnosis of the pure struma ovarii, as reported by Wei *et al.* [15]. Although the detailed pathological information on her ovarian cystectomy, which was done before two years ago, could not be known and the possibility of the mature cystic teratoma exists. Our case was diagnosed as pure struma ovarii at the operation. Only a few cases concerning cytology of struma ovarii have been reported [13] [14]: their cytologic findings included the presence of typically colloid with mosaic pattern, follicles, follicular cells alone and/or sheets of follicular cells [14]. However, these findings are rare [14], as found in our case. On the other hand, immunocytochemistry was reported to be valuable tool for accurate diagnosis for the struma ovarii [13] [14].

Most cases of struma ovarii were asymptomatic, as is the present case [3]. When there is struma ovarii larger than normal thyroid, the patient rarely has hyperthyroidism: thyroid hyperfunction is noted in 5% - 8% of cases [1]. Struma ovarii is defined as histologically characterized by the presence of thyroid-like

glands at least 50%, whereas pure struma ovarii has been reported in half number of all struma ovarii [1] [2]. Struma ovarii is predominantly found in women between the ages of 40 and 60 years [9]. The patient was 34 year-old and younger than predominant age distribution (Table 1).

Table 1. Sociodemographic and clinical characteristics of struma ovarii.

	Struma ovarii	Present case
Definition	-A mature teratoma composed either exclusively or predominantly of thyroid tissue	-Only thyroid tissue is present in the lt. ovarian tumor
Epidemiology	-The most common type of monodermal teratoma	
Age	-Reproductive years (mainly 40 - 60 years old)	-34 years old
Clinical manifestations	-Usually present with pain and/or a pelvic mass -Ascites (up to 1/3 of patients) -Rarely hyperthyroidism	-Secondary infertility -Polycystic ovary syndrome (rt ovary) -No ascites and hyperthyroidism
Macroscopic findings (Tumor size)	-Usually unilateral and solid (<10 cm)	-Lt. ovary occupied by cystic and solid tumor tissues (7 cm)
Serum CA125 level	-High level of CA125 is suggestive of malignancy	-Within normal limits (7.2 U/ml)
Histopathology	-Mature thyroid tissue is present predominantly (over 50%) in mature ovarian teratoma	-Thyroid tissue is present exclusively in lt. ovarian cystic tumor with papillary projection
Malignant transformation	-About 5% -Mostly papillary carcinoma with <i>BRAF</i> mutations and <i>RET/PTC</i> rearrangements	-None
Prognosis & follow-up time	-Most cases of typical struma ovarii are benign. -The outcome of histologically and biologically malignant thyroid type tumors in struma is favourable	-Well, 7 months after the operation

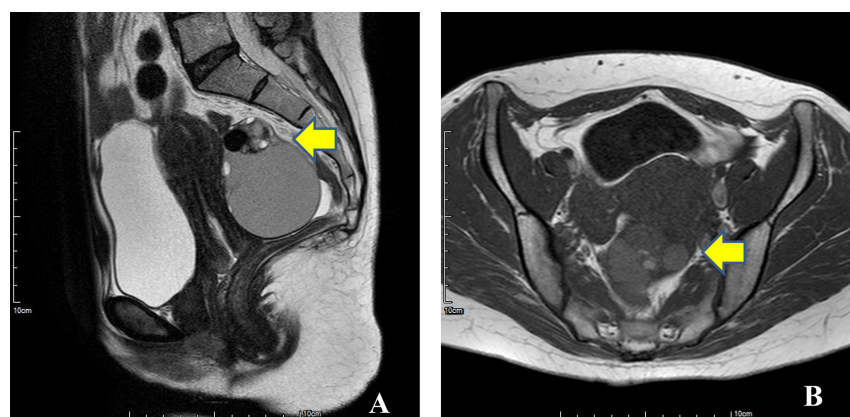


Figure 1. Pelvic MRI shows multiple small cysts with thick separations (arrow), some of them filled with colloidal material which show as low signal density in T2-weighted (A) and relative high (arrow) in T1-weighted (B).

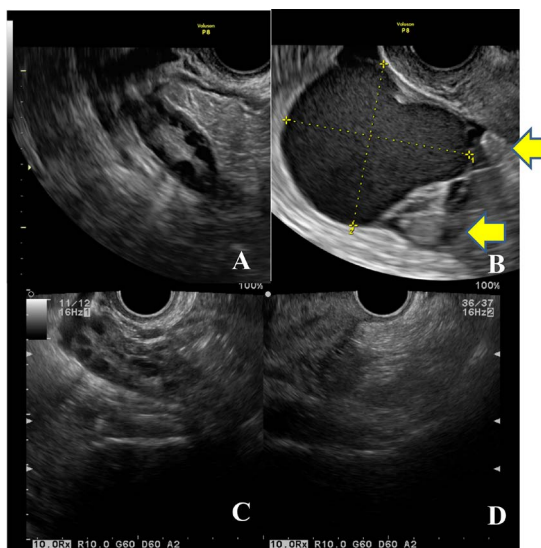


Figure 2. Pelvic ultrasonographic images before and after surgery. Before surgery, right ovary showed polycystic ovary syndrome (A) and left ovary showed large cystic tumor with papillary protrusions (arrows) (B). After surgery, right ovary showed normal appearance (C) and the adnexal tumor disappeared (D).

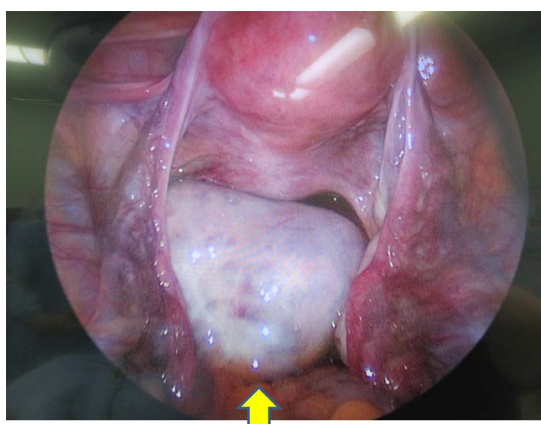


Figure 3. Laparoscopic findings of the left ovarian tumor (arrow).

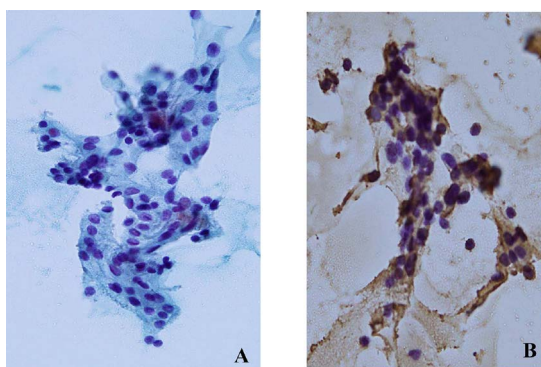


Figure 4. The imprint cytology on the left ovarian tumor. Glandular cells without atypia (A) are immunocytochemically positive for thyroglobulin (B). (A) Papanicolaou stain (original magnification, $\times 100$) and (B) thyroglobulin immunohistochemistry (original magnification, $\times 100$).

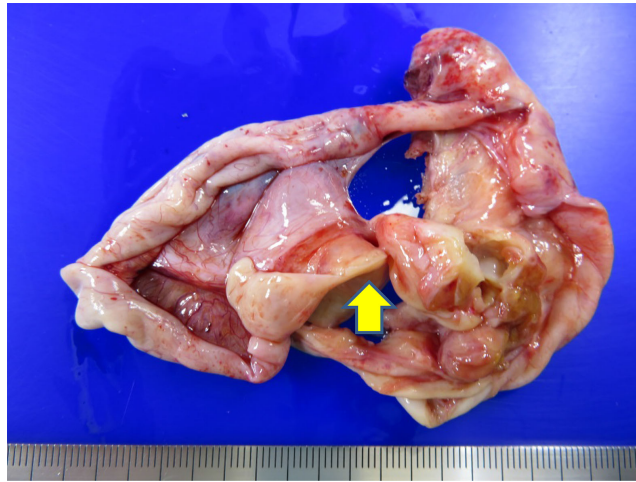


Figure 5. Macroscopic view of the left ovary. The tumor replaced the left ovary. The tumor consisted with solid (arrow) and cystic parts containing gelatinous and colloidal fluid.

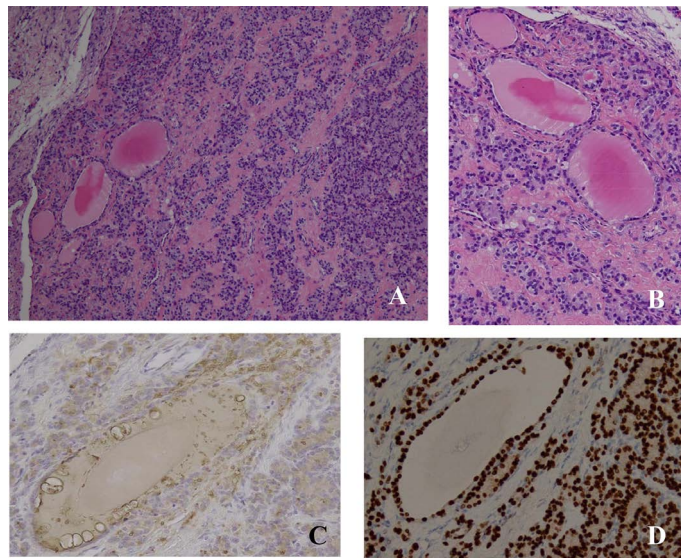


Figure 6. Histopathology of the left ovarian tumor. Microscopically, the tumor is consisted of colloid-containing follicles with varying sizes without nuclear atypia (A, $\times 20$; B, $\times 40$, H & E). Immunohistochemically, the tumor cell are positive for thyroglobulin (C, $\times 100$) and TTF-1 (D, $\times 100$).

Before operation, MRI findings are reported to be useful for the differential diagnosis for the struma ovarii [10] [11] [12]. MRI findings show multilocular cysts with solid components of variable signal density within the loculi. Signal intensity is slightly high on T1-weighted image and very low on T2-weighted image. These findings may suggest presence of hemorrhage or gelatinous (highly viscous) colloidal material of the content [10] [11] [12].

As described in previous reports, the key management of struma ovarii is surgical removal [1] [2] [4] [5]. Fertility-conserving surgery in most cases could be performed in the cases of their stated desire or potential for fertility. Laparos-

copic removal of large ovarian cysts has been well-established and technically feasible for struma ovarii [16] [17]. As struma ovarii is often diagnosed post-operatively, there is concern that the removal of potentially malignant ovarian struma ovarii via laparoscopy may not determine the accurate stage, and increase the possible port site metastasis and spillage of tumor cells into the abdominal cavity. Therefore, we considered that taking out of the ovarian cystic tumor with struma ovarii without rupture must be important, and imprint cytology was useful for diagnosis struma ovarii.

4. Conclusion

A case of pure struma ovarii tumor diagnosed by cytology during a laparoscopic surgery was reported. The patient was a 34-year-old Japanese woman underwent a laparoscopic left oophorectomy under a preoperative diagnosis of endometriotic cyst or mature cystic teratoma in the left ovary. The content of the left ovarian cystic tumor was yellow gelatinous, suggesting a presence of mature cystic teratoma. The imprint cytology of the tumor showed benign glandular pattern, like as a struma ovarii. Histopathological findings led to the diagnosis of pure struma ovarii with a strong positive reaction for thyroglobulin and TTF-1. No other metastases or disseminated lesions were detected. The patient has no recurrent signs 7 months after her operation.

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Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

Authors' Contributions

Authors' contributions	KN	MT	TI	SM	KK	SM	YY	TT
Research concept and design	y	n	n	n	n	n	n	n
Collection and/or assembly of data	y	y	y	n	y	y	y	n
Data analysis and interpretation	y	y	y	y	y	y	y	n
Writing the article	y	n	n	n	n	n	n	n
Critical revision of the article	y	n	n	n	n	n	n	y
Final approval of article	y	y	y	y	y	y	y	y

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