PATHOLOGICA 2020;112:224-228;
DOI: 10.32074/1591-951X-122

Case Report

Follicular carcinoma arising from struma ovarii. A case report

Faten Limaiem, Saadia Bouraoui

University of Tunis El Manar, Tunis Faculty of Medicine, Tunisia

Summary
Struma ovarii is a monodermal variant of ovarian teratoma. Thyroid-type carcinoma arising in struma ovarii is rare. The most common type is papillary carcinoma, followed by typical follicular carcinoma. A 75-year-old hypertensive patient consulted for the sensation of a painless pelvic mass that has been progressing for six months. The abdominopelvic ultrasound showed a right lateralized abdominopelvic mass measuring 14x13x8 cm with a solid and cystic double component. The patient underwent a unilateral right adnexectomy. Grossly, the tumor was encapsulated and lobulated. On cut sections, it was solid brown whitish in color and gelatinous. On histological examination, it was formed of follicular structures of variable size filled with a dense colloid. From this goiter a malignant tumor proliferation arose, arranged in sheets, trabeculae and follicular structures, and the tumor cells were cubic or polyhedral moderately atypical with few mitotic figures. There were no papillary-like nuclear features. There was focal capsular and vascular invasion. Immunohistochemical study showed positive immunostaining of tumor cells with TTF1. Postoperative course was uneventful. The exact prognosis of thyroid-type carcinoma arising in struma ovarii is still unclear because of its rarity, inadequate follow-up, and the absence of consensus in diagnosis and treatment.

Key words: ovary, malignant, neoplasm, struma ovarii, teratoma, follicular carcinoma

Received: April 13, 2020
Accepted: May 5, 2020

Correspondence
Faten Limaiem
Department of Pathology, Mongi Slim Hospital, La Marsa, Tunisia
Tel. +216 96 55 20 57
Fax +216 893 618
E-mail: fatenlimaiem@gmail.com

Conflict of interest
The Authors declare no conflict of interest.

How to cite this article: Limaiem F, Bouraoui S. Follicular carcinoma arising from struma ovarii. A case report. Pathologica 2020;112:224-228. https://doi.org/10.32074/1591-951X-122

Introduction

Struma ovarii is a rare monodermal variant of teratoma. It is composed predominantly of thyroid tissue (50%), and accounts for approximately 2.5 to 5% of ovarian teratomas [1,2]. Malignant struma ovarii is very rare accounting for less than 5% of the cases. Papillary and follicular carcinoma are the most frequent types of malignancy that arise in struma ovarii; other forms of thyroid carcinoma occur rarely [1,2].
In this paper, we report a new case of follicular carcinoma arising in struma ovarii. Our aim was to recall the clinicopathological features of this rare entity.

Clinical history

A 75-year-old hypertensive patient consulted for the sensation of a painless pelvic mass that has been progressing for six months. The abdominopelvic ultrasound showed a right lateralized abdominopelvic mass measuring 14x13x8 cm with a solid and cystic double component. The patient underwent a unilateral right adnexectomy. On gross examination, the tumor was encapsulated and lobulated (Fig. 1 A). On cut sections, it was solid brown whitish in color and gelatinous (Fig. 1 B).

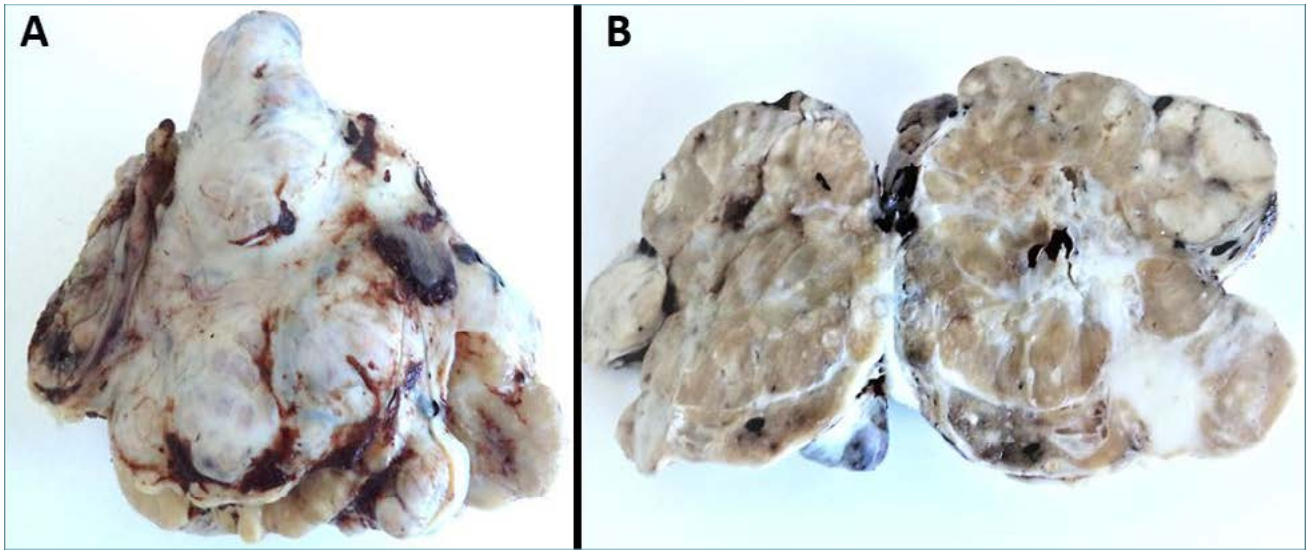

Figure 1. (A) Gross photograph of the surgical specimen showing an encapsulated ovarian mass (B). On cut sections, it was solid brown whitish in color and gelatinous.

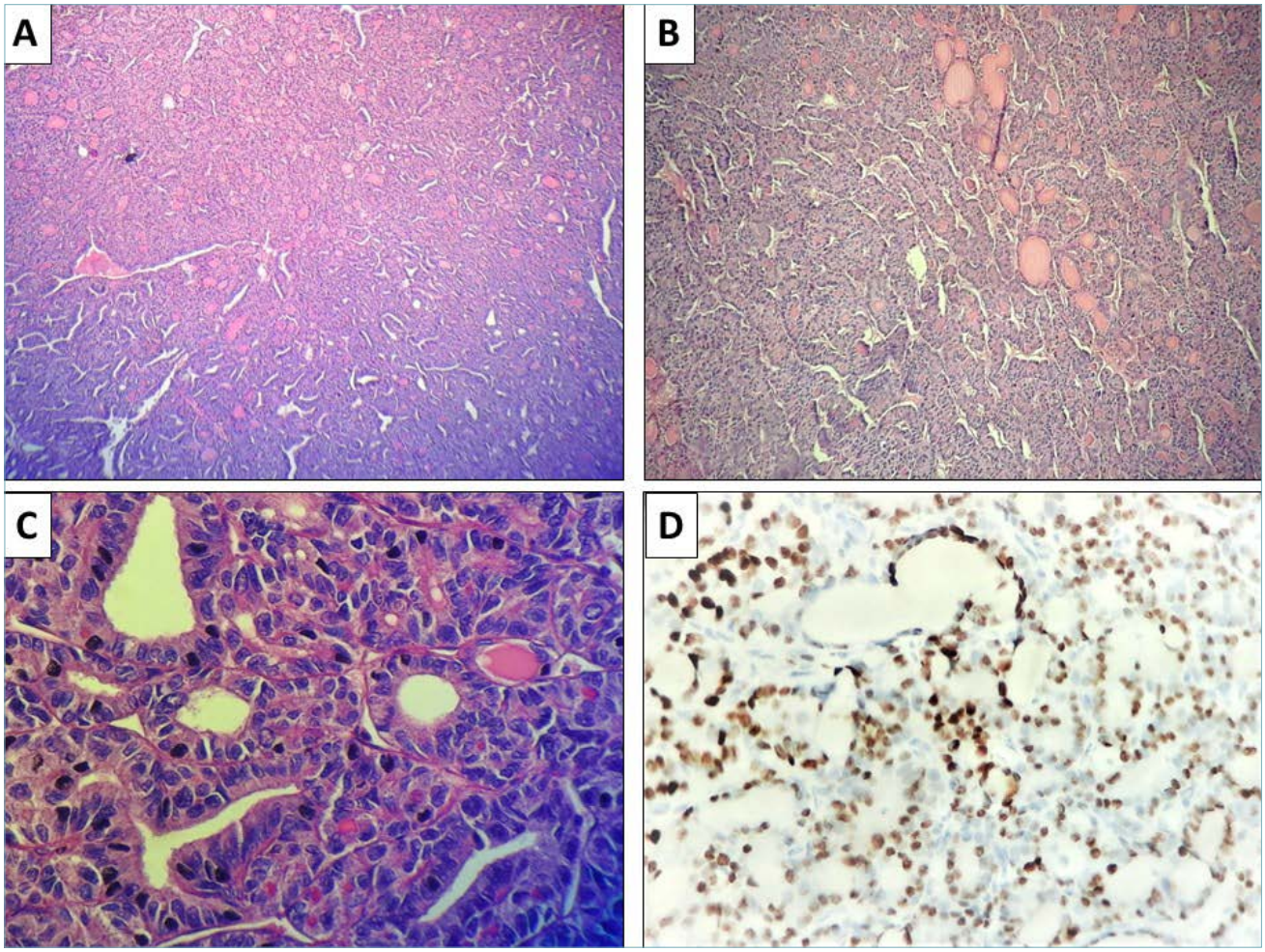

Figure 2. (A-B) Histologic section showing follicular carcinoma arising in struma ovarii. Tumor cells are arranged in follicles and trabeculae, (hematoxylin and eosin, x 40). (C). At higher magnification, the follicles contain pink-colored colloid. There are no nuclear features suggesting papillary carcinoma, (hematoxylin and eosin, x 400). (D) Immunohistochemical staining showing intense nuclear immunoreactivity of tumor cells for TTF1, (immunohistochemistry, x 400).

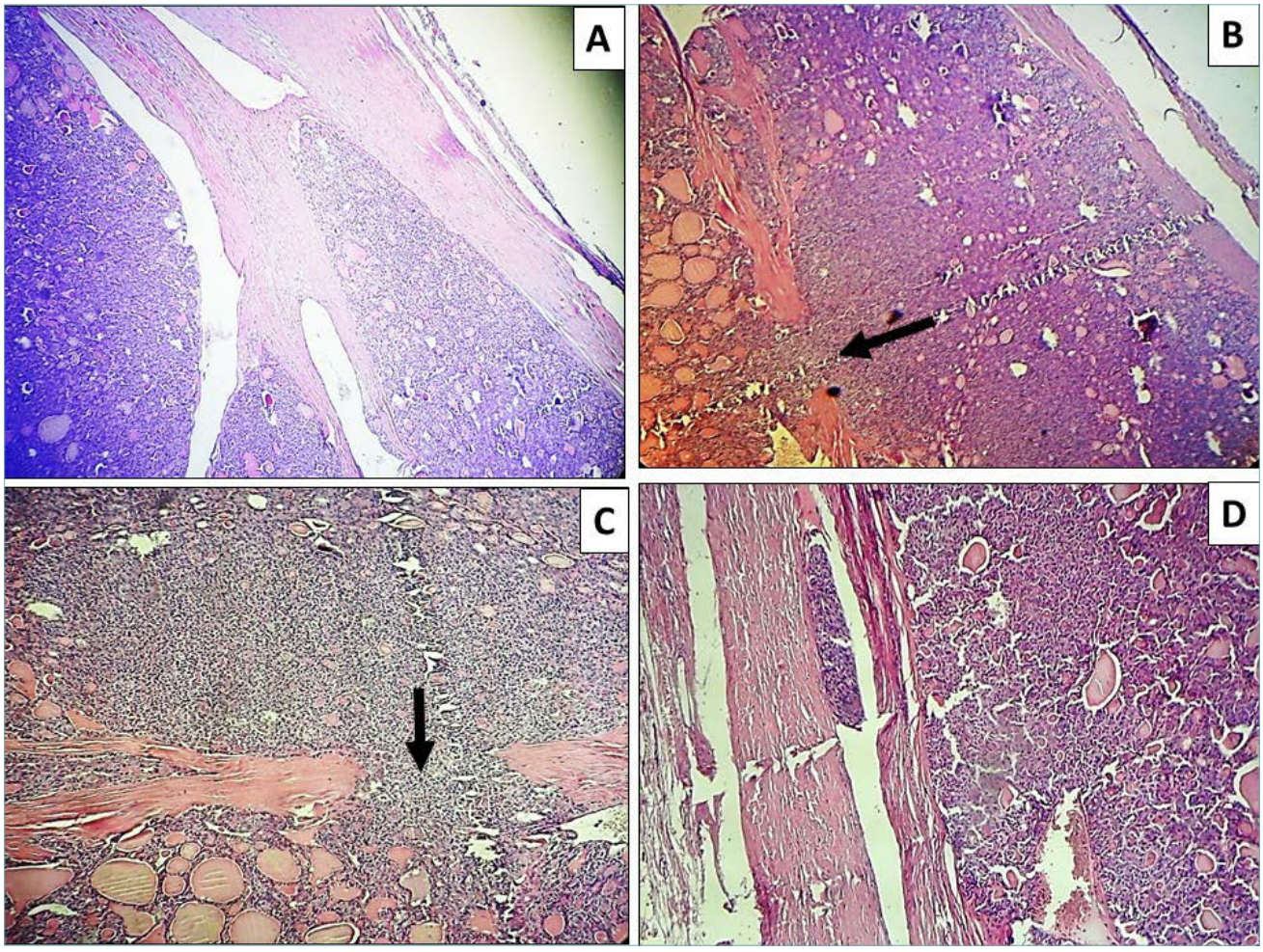

Figure 3. Histologic section showing follicular carcinoma arising in struma ovarii. At low magnification, there is a focal capsular invasion (arrow), (hematoxylin and eosin, x 40).

Intra-operative frozen section analysis was performed and concluded to an adult type granulosa cell ovarian tumor. Subsequently, the patient underwent total abdominal hysterectomy with bilateral salpingo-oopherectomy, without adjuvant treatment. On histological examination, the ovarian mass was formed of follicular structures of variable size filled with a dense colloid. From this goiter a malignant tumor proliferation arose, arranged in sheets, trabeculae and follicular structures (Fig. 2 A, B). The tumor cells were cubic or polyhedral moderately atypical with few mitotic figures (Fig. 2 C). There were no papillary-like nuclear features (Fig. 2 C). We noted focal capsular and limited vascular invasion < 4 vessels (Fig. 3 A-D). Immunohistochemical study showed positive immunostaining of tumor cells with TTF1 (Fig. 2 D). The final pathological diagnosis was malignant struma ovarii with minimally invasive follicular carcinoma. By analogy to follicular carcinoma of the thyroid, this follicular carcinoma arising in struma ovarii can be classified as minimally invasive with limited vascular invasion < 4 vessels.
Postoperative course was uneventful. After the operation, thyroid function tests and the thyroglobulin level were assessed. Thyroid stimulating hormone and free levels of thyroid hormones and thyroglobulin were within normal limits. The patient did not receive any adjuvant treatment. During the six-month follow-up period, the patient did not show evidence of recurrence or metastasis.

Discussion

Struma ovarii is a benign and relatively uncommon monodermal germ cell tumor with less than 200 cases reported in literature [3-5]. It can occur at any age,

with a peak of frequency in the fifth and sixth decades of life [6]. Patients with follicular carcinoma arising in struma ovarii range from 22 to 70 years old [7]. The most common presenting symptom of struma ovarii is a pelvic mass as was the case in our patient. Ascites has been reported in 15-20% of cases, but the presence of ascites does not necessarily indicate malignancy. Although struma ovarii is a neoplasm consisting of thyroid tissue, only 8% of patients present with hyperthyroidism [8]. The term struma ovarii should be reserved for tumors composed either exclusively or predominantly of thyroid tissue (> 50%) [9]. Struma ovarii also comprises cases of mature teratoma with less than 50% thyroid tissue that contain thyroid-associated malignancy [9]. The most common malignancy arising in struma ovarii is papillary thyroid carcinoma followed by follicular carcinoma [10]. Recently, some authors described a new entity, highly differentiated follicular carcinoma of ovarian origin, which is characterized by extra-ovarian dissemination of thyroid elements with an innocuous appearance that histologically resembles non-neoplastic thyroid tissue [11]. To the best of our knowledge, approximately 50 cases of malignant struma ovarii with follicular-type carcinoma have been reported in the English language literature [12]. Because of its rarity, there has been disagreement regarding the diagnosis and the treatment of malignant struma ovarii. Some authors have advocated that the histological diagnosis of malignant struma ovarii should follow the same guidelines as those for thyroid carcinomas [1].

The histological diagnosis of malignant struma ovarii with follicular carcinoma is often difficult, since it is frequently challenging to evaluate capsular invasion in the ovary. In these cases, the diagnosis of malignant struma ovarii is based on the evidence of malignant behaviors including the presence of vascular invasion, dissemination, or metastasis. The most reliable diagnostic criterion for follicular carcinoma is vascular invasion in struma ovarii. Adequate sampling is therefore crucial in this setting [11]. Histological changes of malignancy in struma often do not equate with clinically malignant behavior, and most cases of thyroid-type carcinoma arising in ovarian struma do not have a clinically aggressive course [9]. According to some authors, the identification of a specific molecular alteration in malignant struma ovarii with follicular carcinoma can be a useful biomarker for the prediction of malignant behavior of malignant struma ovarii with follicular carcinoma even when confined to the ovary. In one study, the authors investigated the tumor samples of malignant struma ovarii with follicular carcinoma for 50 cancer-related genes, including RAS, BRAF, p53, and PPARgPAX8 gene fusion by targeted DNA sequencing and fluorescence in situ hybridization, respectively. The main driver gene alterations in follicular thyroid carcinoma were not found in malignant struma ovari, which suggests the possibility of a different mechanism of tumorigenesis for malignant struma ovarii with follicular carcinoma [12]. The histological differential diagnosis of struma ovarii includes metastatic thyroid carcinoma to the ovary, stromal carcinoid, sex cord stromal tumor and melanoma.

The treatment of choice of malignant struma ovarii is contraversial. Management strategies include radical surgery with or without thyroidectomy, followed by adjuvant therapy (chemotherapy, external radiotherapy, thyroid suppression) [13]. Some authors have advocated that malignant struma ovarii should be treated with total abdominal hysterectomy, bilateral salpingo-oopherectomy, pelvicparaaortic lymph node sampling and thyroidectomy followed by I131 therapy [6]. According to these authors, thyroidectomy should be a part of radical surgery to exclude the possibility of a primary thyroid carcinoma [6].

The prognosis of thyroid-type carcinoma arising in struma ovarii is still unclear because of its rarity, inadequate follow-up, and the absence of consensus in diagnosis and treatment. However, limited case reports and a small-series review demonstrated a good prognosis.

Roth et al. [10] concluded that anaplastic carcinoma is the most aggressive tumor type, and highly differentiated follicular carcinoma of ovarian origin is the least aggressive. Of the more common tumor types, typical follicular carcinoma is more aggressive than papillary carcinoma. However, the conclusion that typical follicular carcinoma is more aggressive than papillary carcinoma is still not convincing because of the rarity, the limited case reports, and the variability between median and mean survival data [7]. Further investigation and studies with more cases are needed to reveal the survival difference between follicular and papillary carcinoma [7,10].

The metastatic potential in patients with struma ovarii is low. The most common metastatic sites include other pelvic structures, contralateral ovary, omentum, and mesentery [14]. In rare cases, thyroid tissue may spread to the peritoneal cavity in a condition termed peritoneal strumosis [15]. The peritoneal implants grow slowly and only rarely cause side effects such as the formation of adhesions and intestinal occlusion. Pathologic examination of the peritoneal implants shows multiple nodules of mature thyroid tissue of various sizes with features similar to those of struma ovarii [7].

In conclusion, malignant struma ovarii remains a challenge for the clinicians. Although prognosis of malignant struma ovarii is favorable, its clinical course is

unpredictable and consensus on its optimal treatment has not been clearly established. We need more data to determine the optimal diagnosis, management and follow-up protocols for this rare entity.

References

1 Devaney K, Snyder R, Norris HJ, et al. Proliferative and histologically malignant struma ovarii: a clinicopathologic study of 54 cases. Int J Gynecol Pathol 1993;12:333-43. https://doi.org/10.1097/00004347-199310000-00008

2 Yassa L, Sadow P, Marqusee E. Malignant struma ovarii. Nat Clin Pract Endocrinol Metab 2008;4:469-72. https://doi.org/10.1038/ncpendmet0887

3 Boyd JC, Williams BA, Rigby MH, et al. Malignant Struma ovarii in a 30-year old nulliparous patient. Thyroid Res 2017;10:3. https://doi.org/10.1186/s13044-017-0038-1

4 Goffredo P, Sawka AM, Pura J, et al. Malignant Struma ovarii: a population-level analysis of a large series of 68 patients. Thyroid 2015;25:211-5. https://doi.org/10.1089/thy.2014.0328

5 Marcy PY, Thariat J, Benisvy D, Azuar P et al. Lethal, malignant, metastatic Struma ovarii. Thyroid 2010;20:1037-40. https://doi.org10.1089/thy.2010.0022

6 Yücesoy G, Cakiroglu Y, Muezzinoglu B, et al. Malignant struma ovarii: a case report. J Korean Med Sci 2010;25:327-9. https://doi.org/10.3346/jkms.2010.25.2.327

7 Zhang X, Axiotis C. Thyroid-type carcinoma of struma ovarii. Arch Pathol Lab Med 2010;134:786-91. https://doi.org/10.1043/1543-2165-134.5.786

8 Dardik RB, Dardik M, Westra W, et al. Malignant struma ovarii; two case reports and review of the literature. Gynecol Oncol 1999;73:447-51. https://doi.org/10.1006/gyno.1999.5355

9 Roth LM, Talerman A. Recent advances in the pathology and classification of ovarian germ cell tumors. Int J Gynecol Pathol 2006;25:305-20. https://doi.org/10.1097/01.pgp.0000225844.59621.9d

10 Roth LM, Miller AW, Talerman A. Typical thyroid-type carcinoma arising in struma ovarii: a report of 4 cases and review of the literature. Int J Gynecol Pathol 2008;27:496-506. https://doi.org/10.1097/PGP.0b013e31816a74c6

11 Roth LM, Karseladze AI. Highly differentiated follicular carcinoma arising from struma ovarii: a report of three cases, a review of the literature, and a reassessment of so-called peritoneal strumosis. Int J Gynecol Pathol 2008;27:213-22.

12 1Tsukadaa T, Yoshidab H, Ishikawaa M, et al. Malignant struma ovarii presenting with follicular carcinoma: a case report with molecular analysis. Gynecol Oncol Rep 2019;30:100498. https://doi.org/10.1016/j.gore.2019.100498

13 Makani S, Kim W, Gaba AR. Struma ovarii with a focus of papillary thyroid cancer: a case report and review of the literature. Gynecol Oncol 2004;94:835-9. https://doi.org/10.1016/j.ygyno.2004.06.003

14 1Kabukcuoglu F, Baksu A, Yilmaz B, et al. Malignant struma ovarii. Pathol Oncol Res 2002;8:145-7. https://doi.org/10.1007/BF03033726

15 Brogsitter C, Wonsak A, Wurl K, et al. Peritoneal strumosis. Eur J Nucl Med Mol Imaging 2004;31:1057. https://doi.org/10.1007/s00259-004-1548-3