CEOG Clinical and Experimental Obstetrics & Gynecology

Struma ovarii complicating pregnancy: a case report

F.V. Oreopulu[1], C. Sofoudis[2], S. Voulgaridou[1], S. Stasinopoulou[3], S. Fountoulis[4], G.P. Fragulidis[4]

[1] "Mitera Maternity General Hospital", Athens; [2] 2nd Department of Obstetrics and Gynecology, [3] Department of Pathology, [4] 2nd Department of Surgery, "Aretaieion Hospital", University of Athens Medical School, Athens (Greece)

Summary

Struma ovarii represents an ovarian mature teratoma with thyroid tissue comprising more than 50% of the ovarian tumor. It is a rare condition, representing approximately 1% of all ovarian tumors with a potential malignant transformation 5% to10%. When it is combined with pregnancy, it renders its management in this circumstance is extreme challenging. The authors present a case of benign struma ovarii diagnosed as a right adnexal mass during first trimester of pregnancy with an uneventful clinical course.

Key words: Pregnancy; Struma ovarii; Adnexal mass; Ovarian cyst.

Introduction

Struma ovarii is a monodermal ovarian teratoma composed mainly of differentiated thyroid tissue. It occurs rarely, representing less than 1% of all ovarian tumors. Generally benign, although the malignant cases count approximately 5-10 % [1], it is most common in ages between 40 and 60 years. The symptoms are similar to other ovarian tumors and rarely hyperthyroidism is the presenting symptom, seen in 5-8% of patients with struma ovarii. [2]. It can be rarely diagnosed in mucinous cystadenoma or combined with Brenner tumor [3].

During pregnancy, with the increased use of imaging studies, the detection rate of adnexal masses is approximately 2.5 % [4]. Consequently, despite that most of them are benign, great attention regarding the diagnostic and therapeutic approach must be given. Thus, although the optimal management of an adnexal mass during pregnancy is laparotomy to exclude malignancy, serial imaging studies as transvaginal ultrasonography and serum tumor markers can distinguish it and may early recognize malignant transformation.

The authors present a case of benign struma ovarii first diagnosed as a right adnexal mass during the first trimester of pregnancy with an uneventful delivery and clinical course.

Case Report

A 36-year-old Caucasian female (para 1, gravida 0) with free family, obstetrical, and gynecologic history presented to the present department complaining of amenorrhea for a period of two months and severe hypogastric pain.

A physical examination revealed an enlarged and painful right adnexa during palpation and an enlarged size of the uterus. The transvaginal ultrasound and the increased B-HCG certified the progressive pregnancy with the presence of one endometrial gestational sac and embryonic heartbeat (8w5d). Additionally, corpus luteum of the left ovary and a 8 x 10 cm right adnexal mass were diagnosed (Figure 1). The adnexal mass revealed no signs of malignancy such as septal spaces and papillary protrusions and there was no free peritoneal fluid. The tumor markers such as Ca-125 were negative. A complete blood count revealed mild leukocytosis (WBC 14,000, neutrophils 90%). A regular consultation was scheduled. During the follow up at the 12th week of gestation, the patient had no clinical symptoms and the transvaginal ultrasound revealed a decreased size of right adnexa (5 x 9 cm) (Figure 2). The size and structure of the mass remained stable throughout pregnancy and there were no clinical signs or symptoms indicating torsion or rupture of the cyst. The tumor markers remained free of malignancy. The patient had a normal delivery of a healthy baby boy (38w5d), weight 3,450 gr, and Apgar score of 9 in the first minute and 10 in the fifth minute.

Figure 1. — Right adnexal mass in first trimester of pregnancy (8w5d).

Revised manuscript accepted for publication August 4, 2014

Figure 2. — Right adnexal mass in second trimester of pregnancy (16w).

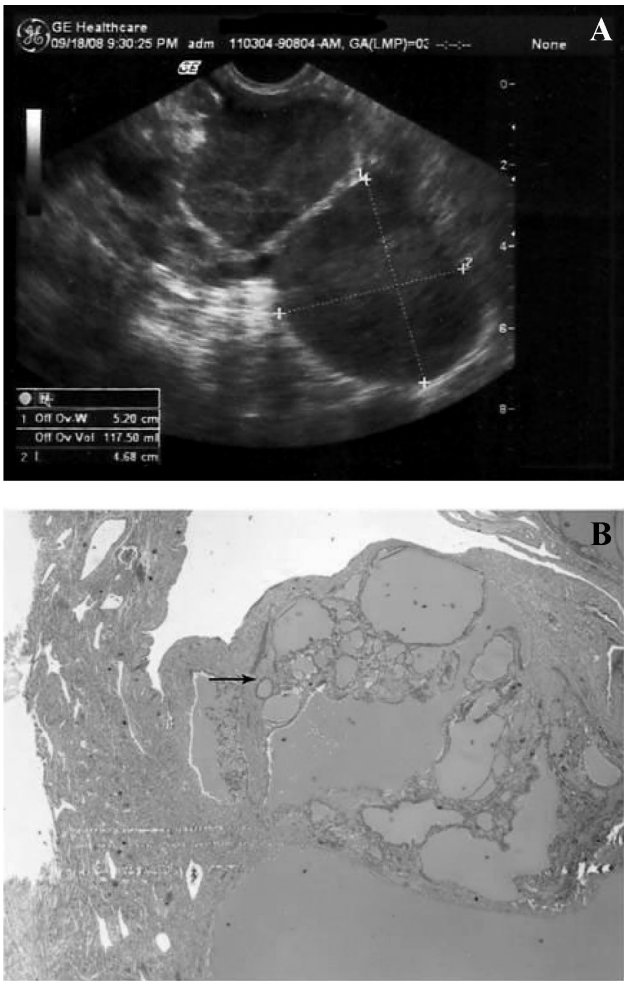

Figure 3. — A) Right adnexal mass postpartum (11 x 5 cm). B) Histological section of ovarian monodermal teratoma, showing thyroid follicles filled with colloid (arrow) (HE x25).

Post-delivery, the transvaginal ultrasound revealed an increased size of the adnexal mass (5 x 11 cm) with mucus like appearance with normal laboratory tests and serum tumor markers (Figure 3A). The patient underwent an exploratory laparotomy where a 12.5 x 7.5 x 6 cm tumor of the right adnexa was recognized. The left adnexa appeared normal. Patient underwent en bloc resection of the right adnexa with the tumor. Tissue biopsy from the left ovary and peritoneal lavage of Douglas space for cytological examination was performed. The frozen section of the right adnexa was negative for malignancy. The histological examination revealed the presence of ovarian monodermal teratoma covered with thyroid follicles filled with colloid (Figure 3B) representing right struma ovarii. The cytological examination was free of malignancy.

The postoperative course was uneventful and the patient was discharged from the hospital on the third postoperative day. Making the diagnosis of the struma ovarii postoperatively, she underwent thyroid function tests as well as ultrasonography of the thyroid gland which were normal. Four months after the exploratory laparotomy the patient became pregnant again and delivered a full-term healthy baby girl.

Discussion

Adnexal masses correlated with pregnancy vary with regards to the imaging findings and clinical symptoms. Between 1% and 2 % of pregnant women will develop an ovarian mass which will be diagnosed sonographically and 1% to 3% of these will be malignantly transformed. [5, 6] Some adnexal masses were diagnosed accidentally during pregnancy and others due to the clinical symptoms they present. The sign of pain indicates in most cases torsion or rupture of the ovarian mass. Torsion can lead to venous and lymphatic blockade which can develop stasis, venous congestion, and necrosis. The incidence is five per 10,000 pregnancies. [7]

The most often types of adnexal masses correlated with pregnancy are cystic teratomas, para-ovarian cysts, serous cystadenomas, and luteomas. [8] Struma ovarii is a monodermal ovarian teratoma, composed mainly of differentiated thyroid tissue, while benign and malignant stumps

ovarii represents 2% and 0.3% of teratomas, respectively. [9] It is most common in ages between 40 and 60 years and rarely occurs before puberty. There are reports of papillary hyperplasia, adenomas or Hashimoto's thyroiditis. [10] Stuma ovarii not only can be present as part of benign cystic teratomas, but also as mucinous cystadenomas or Brenner tumors. [11,12] The histology of a struma ovarii represents coexistence of ovarian follicles with thyroid follicles filled with colloid.

Clinical symptoms and clinical characteristics of an adnexal mass is hardly diagnosed during pregnancy. Generally, they remain asymptomatic and may be misdiagnosed with pregnancy related symptoms or complications. If they are diagnosed early, many studies suggest the safety of excision during the first semester of pregnancy. On the contrary, other study suggests a close follow up to avoid the possibility of spontaneous miscarriage. [13, 14] Cysts of six-cm diameter in reproductive age women diagnosed sonographically with no signs of malignancy can be treated conservatively as they may spontaneous decreased. Sonographically symptoms of malignancy and spontaneously increased size of an adnexal mass suggest surgical approach. When the ultrasound findings are not indicative, the help of MRI is irreplaceable. In addition, although serum tumors markers are useful diagnostic tools, yet in many cases they are inaccurate for malignancy during pregnancy. Thus, a careful follow up with a consideration should be performed in these cases.

Surgical resection of struma ovarii continues to be the optimal management concerning the benign cases. Concerning the malignant cases, metastatic or recurrent disease, surgery following with adjuvant radioiodine is the definitive treatment. [15-17]

Conclusion

The incidence of struma ovarii during pregnancy is very rare and most of the time is diagnosed incidentally with sonograph. It should be considered in differential diagnosis of an adnexal mass especially during pregnancy. For the vast majority of patients, as in the present case, it is benign and the prognosis is excellent.

References

[1] Devaney K., Snyder R., Norris H.J., Tavassoli F.A.: "Proliferative and histologically malignant struma ovarii: a clinicopathologic study of 54 cases". *Int. J. Gynecol. Pathol.*, 1993, *12*, 333.

[2] Ayhan A., Yank F., Tuncer S.: "Struma ovarii". *Int. J. Gynecol. Obstet.*, 1993, *42*, 143.

[3] Scully R.E., Young R.H., Clement P.B.: "Monodermal teratomas. Tumors of the ovary, maldeveloped gonads, fallopian tube and broad ligament". *In:* Scully R.E., Young R.H., Clement P.B., (eds). *Atlas of tumor pathology*. Third series, fascicle 23. Washington DC: Armed Forces Institute of Pathology, 1998, 27.

[4] Bernard L.M., Klebba K.P., Gray D.L., Mutch D.G.: "Predictors of persistence of adnexal masses in pregnancy". *Obstet. Gynecol.*, 1999, *93*, 585.

[5] Nelson M.J., Cavalieri R., Graham D., Sanders R.C.: "Cysts in pregnancy discovered by sonography". *J. Clin. Ultrasound*, 1986, *14*, 509.

[6] Bernard L.M., Klebba P.K., Gray D.L., Mutch D.G.: "Predictors of persistence of adnexal masses in pregnancy". *Obstet. Gyneol.*, 1999, *93*, 585.

[7] Yen C.F., Lin S.L., Murk W., Wang C.J., Lee C.L., Soong Y.K., Arici A.: "Risk analysis of torsion and malignancy for adnexal masses during pregnancy". *Fertil. Steril.*, 2009, *91*, 1895.

[8] Lee C.H., Raman S., Sivanesaratnam V.: "Torsion of ovarian tumors: a clinicopathological study". *Int. J. Gyneacol. Obstet.*, 1989, *28*, 21.

[9] Dardik R.B., Dardik M., Westra W., Montz F.J.: "Malignant stuma ovarii: two cases reports and a review of the literature". *Gynecol. Oncol.*, 1999, *73*, 447.

[10] Berendt-Obolonczyk M., Siekierska-Hellmann M., Wojtylak S., Obolonczyk L., Sworczak K.: "From struma ovarii to Hashimoto disease—an unusual diagnosis of primary hypothyroidism: case report". *Gynecol. Endocrinol.*, 2012, *28*, 43.

[11] Terada T., Tateoka K.: "Ovarian cystic tumor composed of Brenner tumor and struma ovarii". *Int. J. Clin. Exp. Pathol.*, 2012, *5*, 274.

[12] Sibio S., Borrini F., Sammartino P., Accarpio F., Biacchi D., Caprio G., *et al.*: "Predominant Brenner tumor combined with struma ovarii containing a papillary microcarcinoma associated with benign peritoneal strumosis: report of a case and histologic features". *Endocr. Pathol.*, 2010, *21*, 199

[13] Ko M.L., Lai T.H., Chen S.C.: "Laparoscopic management of complicated adnexal masses in the first trimester of pregnancy". *Fertil. Steril.*, 2009, *92*, 283.

[14] Koo Y.J., Lee J.E., Lim K.T., Shim J.U., Mok J.E., Kim T.J.: "A 10-year experience of laparoscopic surgery for adnexal masses during pregnancy". *Int. J. Gynecol. Obstet.*, 2011, *113*, 36.

[15] Yoo S.C., Chang K.H., Lyu M.O., Chang S.J., Ryu H.S., Kim H.S.: "Clinical characteristics of struma ovarii". *J. Gynecol. Oncol.*, 2008, *19*, 135.

[16] Krishnamurthy A., Ramshankar V., Vaidyalingam V., Majhi U.: "Synchronous papillary carcinoma thyroid with malignant struma ovarii: A management dilemma". *Indian J. Nucl. Med.,* 2013, *28*, 243.

[17] Leong A., Roche P.J., Paliouras Rochon L., Trifiro M., Tamilia M.: "Coexistence of malignant struma ovarii and cervical papillary thyroid carcinoma". *J. Clin. Endocrinol. Metab.*, 2013, *98*, 4599.

Address reprint requests to:
C. SOFOUDIS, M.D.
Ippokratous 209,
11472, Athens (Greece)
e-mail: chrisostomos.sofoudis@gmail.com