CASE REPORT

Struma Ovarii Presenting with Pseudo-Meig's Syndrome and Elevated CA 125 Levels



T. J. Simi Raj¹ · K. Chitrathara¹ · Pushpa Mahadevan¹ · P. Sanam¹ · Ananda Subramanian¹

Received: 1 August 2016/Revised: 21 September 2016/Accepted: 5 October 2016/Published online: 31 October 2016 © Association of Gynecologic Oncologists of India 2016

Abstract

Purpose Struma ovarii is a rare monodermal variant of ovarian teratoma accounting for only 2% of all mature teratomas. It is diagnosed when thyroid tissue is present in more than 50% of the specimen. Struma ovarii can rarely mimic advanced ovarian cancer and present with a complex ovarian mass, ascites and markedly elevated CA 125 levels. Only one such case is reported from India.

Method We report a case of 43-year-old lady with struma ovarii who presented with a right adnexal mass, gross ascites, vulval and pedal edema and elevated CA 125 levels. She underwent total hysterectomy and bilateral salpingo-oophorectomy.

Results There was immediate resolution of ascites and normalization of CA 125 levels following surgery. She is disease-free now, 3 years after surgery.

Conclusion Struma ovarii with pseudo-Meig's syndrome and raised CA 125 levels poses a diagnostic challenge and can be mistaken for advanced carcinoma ovary.

Keywords Struma ovarii \cdot Pseudo-Meig's syndrome \cdot Elevated CA-125 levels \cdot Ovarian cyst \cdot Ascites \cdot Hydrothorax

☑ T. J. Simi Raj drsimisooraj@gmail.com

Introduction

Struma ovarii is a rare monodermal variant of ovarian teratoma accounting for only 2% of all mature teratomas [1]. It was first described in 1899 by Boettlin. Many teratomas contain thyroid tissue; however, for the diagnosis of struma ovary, the thyroid tissue in it should be more than 50% [2]. It is usually benign. Malignancy is observed in about 5% of tumors [1]. Struma ovarii with pseudo-Meig's syndrome and raised CA 125 is a rare presentation, and it raises the suspicion of malignant ovarian tumor. Here, we report such a case.

Case History

Forty-three-year-old premenopausal para 1 living 1 lady was seen by medical oncology department with lower abdominal pain, abdominal distension, vulval edema and pedal edema. On examination, abdomen was grossly distended with ascites. CT abdomen revealed a heterogeneously enhancing soft tissue density lesion of size 9×8 cm in right adnexa with fat density lesion adjoining it with ascites and mild bilateral pleural effusion. CA 125 level was 941 U/L. Ascitic fluid for cytology and imagingguided FNAC of the pelvic mass were negative for malignant cells; hence, she was referred to surgical oncology. All details were reviewed. Ascitic fluid was drained to relieve dyspnoea and abdominal pain. Pelvic examination revealed an approximately 8×8 cm mobile mass in the right adnexa. The options of diagnostic laparoscopy/laparotomy were discussed with the patient, and laparotomy was decided. Hemoglobin was 10.2 mg%, and serum albumin level was 3.6 mg%. All the other preop investigations including chest X-ray (except for the



¹ Department of Surgical and Gynecological Oncology, Lakeshore Hospital and Research Centre, Nettoor, Kochi, Kerala, India

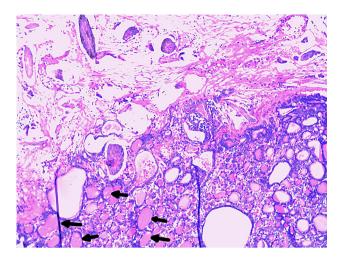


Fig. 1 Struma ovarii with follicles containing colloid material and hair shafts

pleural effusion) and ECG were within normal limits. Laparotomy revealed gross ascites and a $8 \times 6 \times 4$ cm size cystic mass with solid areas with bosselated surface replacing the right ovary. Due to some technical reasons, we could not send the specimen for frozen section. Hence, pelvic lymph node sampling and omental biopsy were done along with hysterectomy and bilateral salpingo-oophorectomy.

Gross histopathology of the surgical specimen revealed cysts filled with cheesy material, hair and solid nodules containing colloid material. Microscopy of the nodules showed varying-sized colloid-filled follicles lined by cuboidal epithelium and focal intraluminal folding of epithelium which was consistent with struma ovarii. Her thyroid function tests done postoperatively were normal. Ascites and pleural effusion resolved completely after surgery, and elevated CA 125 levels got normalized in 3 months. She comes for regular follow-up and is free of disease now, 3 years after surgery (Fig. 1).

Discussion

Struma ovarii can be associated with ascites in 1/3 of cases. When ascites and pleural effusion occurs with ovarian tumors other than thecoma/fibroma, it is known as pseudo-Meig's syndrome. Struma ovarii can present with pseudo-Meig's syndrome. However, ascites and pleural effusion along with elevated CA 125 which points to an advanced ovarian tumor is a rare occurrence with struma ovarii. Out of 11 such cases reported, only one case is reported from India [3]. Our case is reported as the second case from India. Vulval and pedal edema which can occur as part of gross ascites was also not seen in previously reported cases. Ours is the third case reported in less than 50 years. Our patient also had a right ovarian mass which is the preferred site of this tumor as evident from the table. Bilateral tumor was reported in only one case [3] (Table 1).

There are no unique identifying features in ultrasound scan or CT scan other than a multicystic pelvic mass [4]. Benign struma ovarii is managed by surgical resection which includes ovarian cystectomy or salpingo-oophorectomy with or without hysterectomy [5]. The associated thyroid hyperfunction, ascites or hydrothorax resolve spontaneously after excision [5]. Malignant struma ovarii is reported in 5–10% of cases and may be follicular, papillary or mixed in pattern. Desimone et al. [6] suggest treatment with thyroidectomy and I 131 as the first line of management for malignant struma ovarii. Our case of benign struma ovarii had a rare clinical presentation with ascites, pleural effusion and elevated CA 125, and she is free of disease 3 years after surgery.

Table 1 Features of reported struma ovarii associated with pseudo-Meig's syndrome and elevated CA 125 levels

Author et al. with year	Age in years	Post- menopausal	Presenting complaint	CA 125 levels in U/ml	Tumor size in maximum diameter (cm)	Site of tumor	Hyperthyroidism	References
Mitrou 2008	55	Yes	Abdominal mass, distension, weight loss	3803	23	Left	No	[7]
Paladini 2008	42	No	Abdominal distension, fever, diarrhea, vomiting	2548	11	Right	Yes	[8]
Rana 2009	70	Yes	Abdominal distension dyspnoea	284	7.5	Bilateral	No	[3]
Jiang 2010	46	No	Fatigue, anorexia, abdominal distension	1231	20	Right	No	[<mark>9</mark>]
Chengjuan 2014	52	No	Dyspnoea	1289	7	Right	No	[10]
Current report 2016	43	No	Abdominal pain, distension, vulval and pedal edema	941	8	Right	No	

Conclusion

Struma ovarii can rarely present with a complex ovarian mass, ascites and markedly elevated CA 125 levels. Only one such case is reported from India. Struma ovarii with pseudo-Meig's syndrome and elevated CA 125 levels can be included in the differential diagnosis of carcinoma ovary.

Compliance with Ethical Standards

Conflict of interest There is no conflict of interest for any of the authors. There are no relationships for any of the authors that may deem to influence the objectivity of the paper.

References

- Yücesoy G, Cakiroglu Y, Muezzinoglu B, et al. Malignant Struma Ovarii: a Case Report. J Korean Med Sci. 2010;25(2):327–9.
- Serov SF, Scully RE, Sobin LH. Histological typing of ovarian tumors. International histological classification of tumors. No.9. Geneva: World Health Organization; 1973.

- Rana V, Srinivas V, Bandyopadhyay S, et al. Bilateral benign nonfunctional struma ovarii with Pseudo-Meigs' syndrome. Indian J Pathol Microbiol. 2009;52(1):94–6.
- 4. Jung SI, Kim YJ, Lee MW, et al. CT findings. Abdom Imaging. 2008;33(6):740–3.
- Yoo SC, Chang KH, Lyu MO, et al. Clinical characteristics of struma ovarii. J. Gynecol Oncol. 2008;19(2):135–8.
- DeSimone CP, Lele SM, Modesitt SC. Malignant struma ovarii: a case report and analysis of cases reported in the literature with focus on survival and I131 therapy. Gynecol Oncol. 2003;89(3):543–8.
- Mitrou S, Manek S, Kehoe S. Cystic struma ovarii presenting as pseudo-Meigs' syndrome with elevated CA 125 levels. A case report and review of the literature. Int J Gynecol Cancer. 2008;18:372–5.
- Paladini D, Vassallo M, Sglavo G, et al. Struma ovarii associated with hyperthyroidism, elevated CA 125 and pseudo-Meigs syndrome may mimic advanced ovarian cancer. Ultrasound Obstet Gynecol. 2008;32:237–8.
- Jiang W, Lu X, Zhu ZL, et al. Struma ovarii associated with pseudo-Meigs' syndrome and elevated serum CA 125: a case report and review of the literature. J Ovarian Res. 2010;3:18.
- Jin C, Dong R, Bu H, et al. Coexistence of benign struma ovarii, pseudo-Meigs' syndrome and elevated serum CA 125: case report and review of the literature. Oncol Lett. 2015;9(4):1739–42.