Metachronous ovarian mature teratomas with concomitant polycystic ovarian syndrome

M. Le Donne¹, M. Lentini¹, E. Sturlese¹, A. Ardizzone¹, S. Benvenga²

¹Department of Human Pathology in Adulthood and Childhood “G. Barresi”, University of Messina, Messina, Italy
²Department of Clinical and Experimental Medicine, Master Program on Childhood, Adolescent and Women’s Endocrine Health, University of Messina, Messina, Italy

ABSTRACT — A 22 year-old woman presented to our clinic with secondary amenorrhoea, pain of the right adnexal region. At ultrasound, the right ovary presented a multilocular complex mass of 10.4x8.5x7.3 cm enucleated and removed on laparoscopy. The histological diagnosis was mature cystic teratoma. Because of persisting amenorrhoea, the endocrine evaluation performed was consistent with polycystic ovary syndrome (PCOS). Eight months later, two small left ovarian masses were removed by a second laparoscopy and the pathological diagnosis was again mature cystic teratoma. Treatment with myoinositol (MI) 550 mg + D-chiroinositol (DCI) 13.8 mg + folic acid 200 µg twice a day achieved persistent normalization of the endocrine and metabolic values over the 24 months of follow-up. To our knowledge this is an unusual case of multiple and bilateral metachronous ovarian teratomas with a co-existing PCOS.

KEYWORDS
Mature cystic teratoma, metachronous, Hyperandrogenism, Polycystic ovarian syndrome, Myoinositol, D-chiroinositol.

INTRODUCTION

Mature cystic teratoma is the most common germ cell tumor. It accounts for 10% to 20% of all ovarian masses¹. More than 80% of mature cystic teratomas develop during the reproductive age²³. It can be multiple, with bilaterality reported in 10-15% of cases⁴, and metachronous⁵⁶. Polycystic Ovary Syndrome (PCOS) is a complex disease characterized by two of these three conditions according to the Rotterdam criteria: hyperandrogenism, chronic anovulation and polycystic ovarian morphology⁷. We report an unusual case of multiple and bilateral metachronous ovarian teratomas with a co-existing PCOS.

CASE REPORT

A 22 year-old woman, para 0, gravida 0, presented to our clinic with secondary amenorrhoea, pain of the right adnexal region and previous diagnosis of PCOS. Oral oestro-progestins were started two months earlier. On clinical examination, she had normal Body Mass Index (BMI = 23.5 Kg/m²), no hirsutism or acne. Gynecological examination showed an anteverted uterus of normal volume; the left ovary was normal whilst the right ovary was occupied by a mobile mass of approximately 10 cm in diameter. At ultrasound, the left ovary was normal, while the right ovary presented a multilocular complex mass of 10.4x8.5x7.3 cm (Figure 1A). Serum carcinoembryonic antigen, alpha-fetoprotein, CA-125 and β-human chorionic gonadotropin were negative. The mass was enucleated and removed on laparoscopy. Histological examination showed a cyst containing variable proportions of all three germ layers derivatives, consistent with the diagnosis of mature cystic teratoma (Figure 1B). Because of persisting postoperative amenorrhoea, the patient underwent endocrine evaluation. Laboratory results showed elevated LH/FSH.
ed two small areas of 3.7x2.9 cm and 1.4x1.5 cm, both without significant vascular spots by color Doppler imaging. At magnetic resonance imaging, these areas showed distinct fat component, modest shared fluid blood (“floating ball”), and small imaging foci of diffusion restriction. Laparoscopy was performed, and the two complex masses were enucleated and removed (Figure 1D). Pathological diagnosis was again mature cystic teratoma.

DISCUSSION

Ovarian mature teratoma, a type of germ cell tumor, is composed of mature histologic structures of ectodermal, mesodermal and endodermal origin. It is one of the most common kinds of ovarian tumor, with a frequency of approximately 20%. Although most mature teratomas are asymptomatic, the most common symptoms for this disease are abdominal pain, the presence of an abdominal mass and in very rare cases symptomatology is related to hormonal secretion. Only nine cases of androgen-producing ovarian cystic teratomas have been reported, and they should be suspected in the case of a rapid onset
Metachronous ovarian mature teratomas with concomitant polycystic ovarian syndrome

CONCLUSIONS

Therapy based on MI+DCI (40:1) improves the serum baseline hormones and OGTT-stimulated insulin values in patients with co-existing PCOS in the remaining ovary tissue. To our knowledge this is the first case of multiple and bilateral metachronous ovarian teratomas (two of which localized in the same ovary) with a co-existing PCOS.

ACKNOWLEDGMENTS:
We thank Prof. Fabrizio Guarneri for helping to set up the hormonal and insulinemia graph.

CONFLICTS OF INTEREST:
The Authors declare that there are no conflicts of interest.

REFERENCES