

Autoimmune Oophoritis: An Incidental Finding on Hysterectomy

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Abstract

Autoimmune oophoritis is a rare autoimmune disease leading to premature ovarian failure and frequently associated with or at an increased risk of development of other autoimmune diseases. Hence detail clinical examination, investigations and follow up are mandatory especially in infertile young women and to find out other autoimmune disorders.

Herein, we report a case of autoimmune oophoritis as an incidental finding on hysterectomy in a 40-year-female.

Keywords: Autoimmune; Oophoritis; Premature Ovarian Failure.

Introduction

Premature ovarian failure is generally defined as secondary amenorrhoea and infertility before the age of 35 years. Autoimmune oophoritis is one of the rare disorders associated with distinctive changes in patients with premature ovarian failure after excluding other causes like gonadal maldevelopment, obvious chromosomal abnormalities, surgical, radiation induced or drug induced ablation of ovarian function. It is frequently associated with or at an increased risk of development of other autoimmune diseases [1].

Herein, we report a case of autoimmune oophoritis as an incidental finding on hysterectomy for suspected endometrial and ovarian pathology

Case Report

A 40-year-old multipara female presented with complaints of excessive bleeding per vagina and lower

abdominal pain since 3-4 months duration. Patient was multipara, completed family with three full term normal pregnancies and one miscarriage. Her menarche occurred at the age of 14 years with regular menses on 28-29 day cycle until last 3-4 months ago when her menses became excessive, irregular and painful. She also complained of pain in lower abdomen. Patient revealed past medical history of diabetes and hypothyroidism for which she is taking regular treatment since 3 years. Pelvic examination was unremarkable with anteverted uterus and impalpable both fallopian tubes and ovaries. Other systemic examination was unremarkable. Routine laboratory investigations showed microcytic hypochromic anaemia. Fasting blood sugar level was 125mg/dl, postprandial sugar-190 mg/dl. Past thyroid function test report reveal-TSH-10.5 uIU/ml, T3-0.60 ng/ml, T4-2.90 ug/dl. Pelvic ultrasound revealed normal sized right ovary and mild enlargement of left ovary with cystic areas. Patient underwent hysterectomy with left salpingo-oophorectomy for suspected endometrial and ovarian pathology and specimen was sent for histopathological examination. Grossly, uterus with cervix measured 10x7x4.5 cms. Left ovary measured 5x4x3.5 cms. Cut surface- endometrial cavity unremarkable, endocervix showed nabothian cyst. Myometrium showed roughened, congested, irregular trabeculations. Left ovary showed cysts ranging from 0.4 cm to 1.5 cms in diameter with yellow whitish

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areas [Figure-1].

Microscopy showed lymphoplasmacytic infiltrate surrounding theca cell layer of developing and atretic follicle and sparing primordial follicles [Figure-2]. Ovarian stroma showed dense inflammatory infiltrate consisting of lymphoplasmacytic cells [Figure-3]. At places foamy macrophages seen. Cervix showed chronic cervicitis with nabothian cyst and myometrium showed deep adenomyosis.

On histomorphological features patient was diagnosed as a case of lymphocytic oophoritis. Patient was known case of diabetes and hypothyroidism so advised further hormonal assays like FSH, LH, Prolactin, ANA, antithyroid antibodies, Anti-ds DNA to rule out autoimmune cause. FSH and LH levels were raised and antithyroid antibodies were positive. ANA was negative. On the basis of histomorphological and laboratory findings, final diagnosis of Autoimmune oophoritis was made. Patient was advised regular check-up.



Fig. 1: Cut surface of left ovary showed cysts ranging from 0.4-1.5 cms in diameter with yellow-white areas

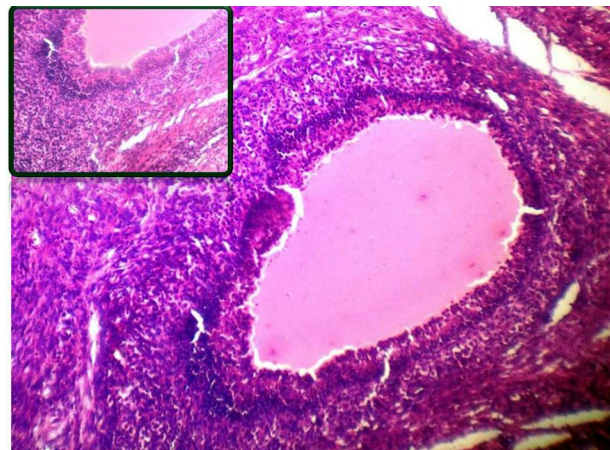


Fig. 2: Microscopy showed lymphoplasmacytic infiltrate surrounding the theca cell layer of developing follicle. (Haematoxylin and eosin, $\times 400$)

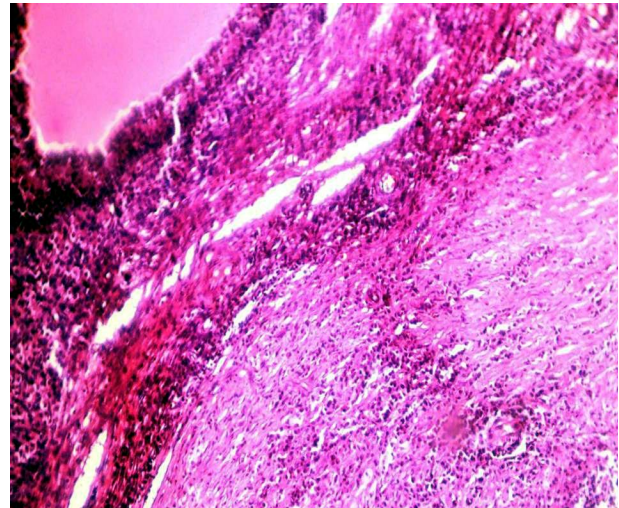


Fig. 3: Microscopy showed dense lymphoplasmacytic infiltrate in the ovarian stroma. (Haematoxylin and eosin, $\times 400$)

Discussion

Ovarian autoimmunity was first reported and documented serologically by Vallotton and Forbes in 1966[2]. Autoimmune oophoritis is a rare distinct cause of premature ovarian failure with secondary amenorrhea.

The ovarian failure is usually preceded or accompanied by one or more of the following autoimmune disorders: Idiopathic Addison disease, idiopathic hypoparathyroidism, myasthenia gravis, juvenile onset diabetes mellitus, SLE, Sicca syndrome, vitiligo, autoimmune haemolytic anaemia etc [3]. Our case also had associated hypothyroidism and diabetes mellitus. Most of the reported cases in the literature, patients had oligomenorrhea or amenorrhea; occasionally symptoms related to enlarged polycystic ovaries or abnormal vaginal bleeding have been the initial manifestations [1]. In our case patient presented with menorrhagia, the cause of which may be either adenomyosis or disturbed ovarian function. Autoimmune oophoritis was seen an incidental findings after hysterectomy in our case. The autoimmunity accounts for upto 30% of premature ovarian failure cases [4].

On gross examination ovaries are usually of normal size, but they may be enlarged and polycystic [1]. Histological examination revealed normal primordial follicles, infiltration of theca cell layer by lymphoplasmacytic cells. The intensity of infiltrate increases with degree of follicular maturation and as the follicle enlarged inflammatory cells and degenerating granulosa cells desquamate into lumen. Lymphocytes and plasma cells are the predominant cells, but eosinophils, histiocytes and sarcoid like

granuloma have also been described [5]. Destruction of Leydig cell by inflammation, lymphoid infiltrate in the ovarian hilus and sometime in a perineural distribution have been described. Histological evaluation of the ovaries in autoimmune oophoritis have been usually incomplete because the diagnosis should be established on the presence of autoantibodies to the ovarian tissue in patients with premature ovarian failure and other autoimmune diseases rather than histologic findings of the ovaries [6].

The proportion of patients with premature ovarian failure who have autoantibodies to the ovarian tissue as evidence of autoimmune oophoritis has varied from 0 to 100% [6]. Russell and Bannatyne suggested the hypothesis that a complex immune process consisting of humoral and cellular mechanism is involved in the pathogenesis of autoimmune oophoritis [7]. No accurate serological marker is available to diagnose patients with premature ovarian failure due to autoimmunity. Ovarian biopsy is currently the only diagnostic modality with certainty.

Literature review reveal use of hormone replacement therapy and steroid therapy especially in young patients who have not completed family for treatment of autoimmune oophoritis. There are multiple case reports in which corticosteroid therapy has resulted in pregnancies in women with autoimmune oophoritis and amenorrhea [8].

Conclusion

To conclude, diagnosis of premature ovarian failure

due to autoimmunity is helpful to restore ovarian function especially in young women with infertility or who have not completed family.

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