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Case Report

An unusual case of pseudoxanthomatous salpingitis

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ABSTRACT

Pseudoxanthomatous salpingitis (PXS) is a rare condition which is characterized by the presence of pigment-laden histiocytes within the lamina propria of the fallopian tube. Till date, less than 30 cases of have been reported in the literature. Herein, we report an incidentally detected case of PXS. PXS has been linked with ovarian endometriosis. The pathogenesis of PXS is unclear but it might be a response by the macrophages to haemorrhage in the lumen of the fallopian tube. The importance of knowing about this entity lies in the fact that it can be preoperatively diagnosed as adnexal neoplasm sometimes mistakenly.

Keywords: Pigmentosis, Plicae, Pseudoxanthomatous, Brown black, Prussian blue

INTRODUCTION

Pseudoxanthomatous salpingitis (PXS) is a rare condition which is characterized by the presence of pigment-laden histiocytes within the lamina propria of the fallopian tube. Till date, less than 30 cases of have been reported in the literature. Herein, we report an incidentally detected case of PXS. PXS has been linked with ovarian endometriosis and has the propensity to destroy the involved tissues.[1] The pathogenesis of PXS is unclear but it might be a response by the macrophages to haemorrhage in the lumen of the fallopian tube. Hence, radiological examination and precise histopathological examination may enlighten the inflammatory pathology of the lesion and helps in preventing misdiagnosis because of the rarity of these conditions.

CASE REPORT

A 52 year old female presented with chief complaints of mass descending per vaginum since 1 year. Ultrasound abdomen and pelvis revealed a right sided hydrosalpinx and possibly cystocoele. Vaginal hysterectomy with bilateral salpingo-oophorectomy and pelvic floor repair was done. Intraoperatively, the uterus was found to be atrophic and right fallopian tube showed hydrosalpinx. Left fallopian tube and bilateral ovaries were normal. Histopathological examination showed numerous pigmented histiocytes with brown black intracytoplasmic pigment in the lamina propria of the fallopian tube (Figure 1-3). The brown black pigment was stained with Prussian blue (Special stain for iron Figure 4) which showed a blue colour.

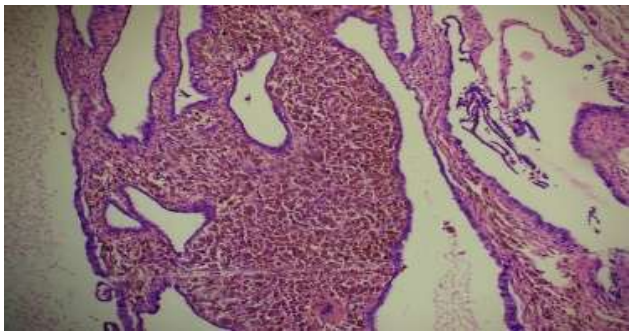

Figure 1: Fused plicae lined by tubal type epithelium and the lamina propria showing sheets of pigment laden macrophages (H and E 40x).

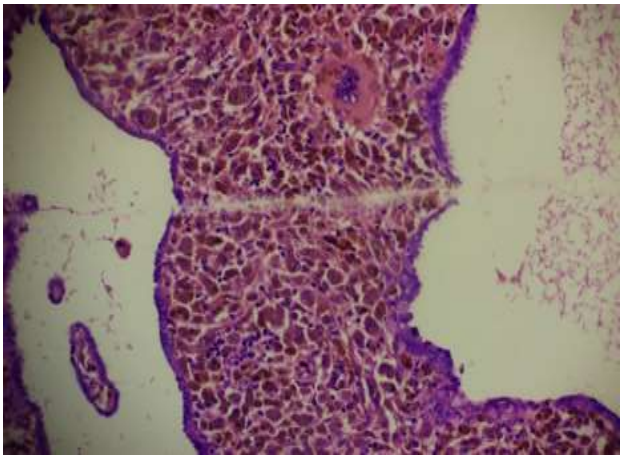

Figure 2: Fused plicae lined by tubal type epithelium and the lamina propria showing sheets of pigment laden macrophages and a foreign body type multinucleate giant cell (H and E 100x).

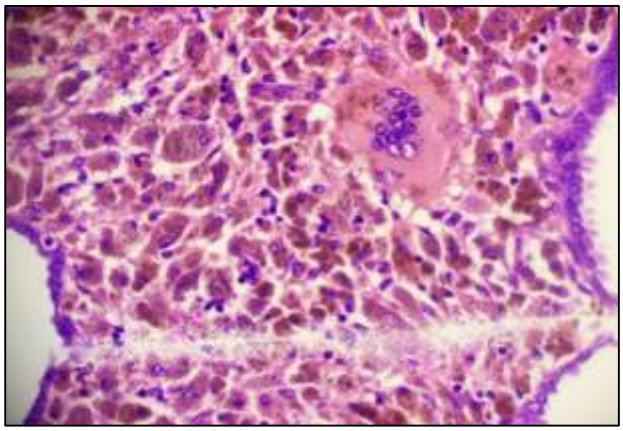

Figure 3: Fused plicae lined by tubal type epithelium with the lamina propria showing sheets of pigment laden macrophages and a foreign body type multinucleate giant cell (H and E 400x).

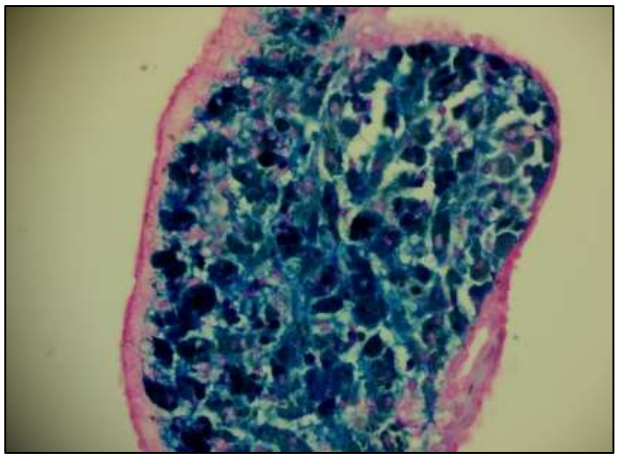

Figure 4: Fused plica lined by tubal type epithelium with the lamina propria showing sheets of pigment laden macrophages. Pigment highlighted by Prussian blue stain (H and E 400x).

DISCUSSION

Pigmentosis tubae is an uncommon condition characterised by the presence of pigment laden macrophages in the lamina propria of the fallopian tube. It was first described by Herrera et al.[2] Alternate terminologies for pigmentosis tubae includes PXS, xanthogranuloma, inflammatory pseudotumor, melanosis tubae, etc. It is usually associated with ovarian endometriosis but can be also associated with pelvic inflammatory disease (PID), intrauterine contraceptive devices (IUCD), radiation, etc. Grossly, tubal mucosa exhibits numerous small bulbous projections. On histopathological examination, distended plicae showing pigmented macrophages with or without other inflammatory cells can be noted. Different school of thoughts exists in the usage of terminologies like PXS and xanthomatous salpingitis. According to Furuya et al PXS appears as a xanthogranulomatous inflammation secondary to endometriosis whereas xanthomatous salpingitis is PID associated with xanthogranulomatous changes.[3] Few authors consider that both entities belong to a spectrum of changes caused by different etiologies. Various hypothesis has been proposed by various authors as to the occurence of PXS. Clement et al reported that spreading of blood in the mucosa of the uterine tube could be the etiology of PXS.[4] Repetitive haemorrhages of the cysts in ovarian endometriosis could reach the lumen of the tubes. Seidman et al proposed that the entry of blood to the lamina propria would happen only in cases with salpingitis and loss in the integrity of tubal mucosa.[5] Endometriotic foci at the mucosa or in the wall of the tube can be another mechanism involved in the pathogenesis of PXS. Idrees et al and Zorzi et al reported cases of PXS in association with fallopian tube mucosal endometriosis or parietal endometriosis. Histologic findings of PXS represent the final event of the clearance of the haemorrhagic material.[6] This occurs by histiocytic cells and the final products of degradation of blood are stored in the cytoplasm of the cells as lipofuscin or ceroid, the source of which could be probably by lipid peroxidation of lysosomes and cellular constituents. Iron-promoted lipid peroxidation may alter the lysosomal membranes and contribute to the excessive accumulation of lipofuscin-ceroid pigment in the pigmented cells of PXS.

CONCLUSION

PXS is a rare condition usually associated with ovarian endometriosis. The pathologic way of pseudoxanthomatous histiocytes to accumulate inside the plicae of the tubes is not clear and can be diverse in different cases. Both xanthogranulomatous salpingitis and PXS may cause follicle like structures and may be pre operatively diagnosed as adnexal neoplasm. Proper history, radiological examination and precise histopathological examination may enlighten the inflammatory pathology of the lesion and helps in

preventing misdiagnosis because of the rarity of these conditions.

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Ethical approval: Not required

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