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Case Report

Pathology Section

Xanthoma of Fallopian Tube in Young Female- An Uncommon Entity

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ABSTRACT

Xanthomas or xanthelasmas are rare benign tumours characterised by localised lipid deposits within an organ system which can be an important sign of systemic disease. These tumours have a predilection for skin and subcutaneous tissue, the most common visceral site being the Gastrointestinal (GI) tract. Xanthoma in female genital tract is a rare finding and should be considered as a differential diagnosis for abdominal pain in a reproductive age group. This is an unusual case of xanthoma of fallopian tube in a 27-year-old female presenting with lower abdominal pain. Ultrasonography (USG) revealed right hydrosalpinx. The histopathological examination of fallopian tube showed sheets of foamy macrophages with peripherally placed nucleus and abundant vacuolated cytoplasm in the lamina propria which was positive for Cluster of Differentiation 68 (CD68) and negative for Cytokeratins (CK). A final diagnosis of xanthoma of right fallopian tube was made. Fallopian tube xanthomas must be distinguished from xanthogranulomatous salpingitis, which is associated with an inflammatory cell infiltrate, often including giant cells and plasma cells.

Keywords: Female genital tract, Lower abdominal pain, Salpingitis, Xanthelasma, Xanthogranulomatous

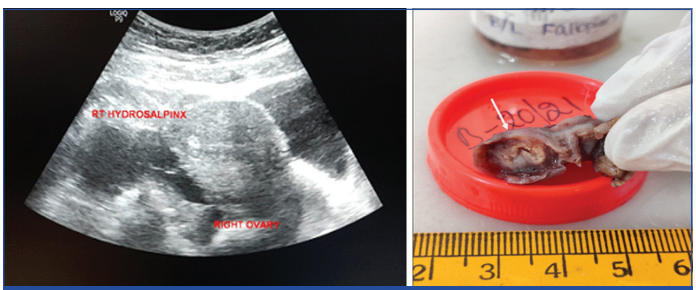
CASE REPORT

A 27-year-old female, with primigravida presented to the Gynaecology Outpatient Department (OPD) with complaints of intermittent right sided lower abdominal pain since one day. The patient had a surgical history of Lower Segment Cesarean Section (LSCS) six months back. There was absence of any significant family history and the haematological investigations were within normal limits.

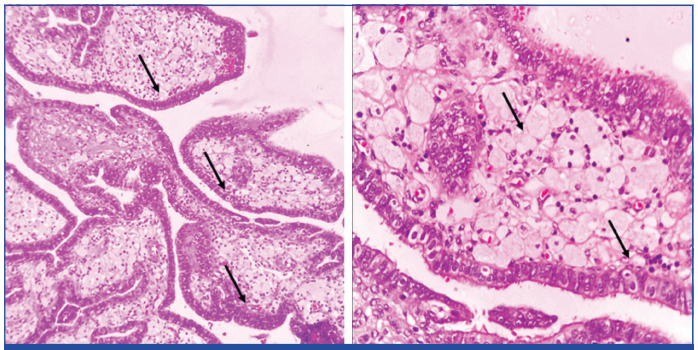
On examination, tenderness was noted on right lower quadrant of abdomen. USG abdomen and pelvis showed a cystic mass measuring 4×1 cm in the right fallopian tube and a radiological diagnosis of right hydrosalpinx was made [Table/Fig-1]. On exploratory laparotomy clear fluid was exuded from the distended right fallopian tube. The uterus, cervix and attached ovary were normal without any significant pathology. The fallopian tube was resected and sent for histopathological examination.

The tissue was fixed in 10% buffered formalin. Grossly, the specimen consisted of a single dilated grey brown tubular tissue measuring 5×1.5×1.2 cm. Cut section revealed dilated lumen with yellow to white plaques in the wall [Table/Fig-2]. After careful dissection, the tissue was processed and embedded in paraffin wax and stained with Haematoxylin and Eosin (H&E) followed by special stains i.e., Periodic Acid-Schiff (PAS), Ziehl-Neelsen (ZN) and Perls Prussian blue stains. Light microscopy exhibited hyperplastic tubal plicae lined by intact tubal epithelium with the sub-epithelial stroma showing sheets of foamy macrophages with peripherally placed nucleus and abundant vacuolated cytoplasm [Table/Fig-3,4]. There was absence of associated inflammatory infiltrate, giant cell reaction, necrosis or fungal elements. The foamy macrophages were positive for PAS stain and negative for Perls stain which ruled out pseudoxanthogranulomatous salpingitis. No acid fast bacilli were seen on ZN stain. A histopathological diagnosis of xanthoma of right fallopian tube was made after ruling out xanthogranulomatous and tubercular salpingitis.

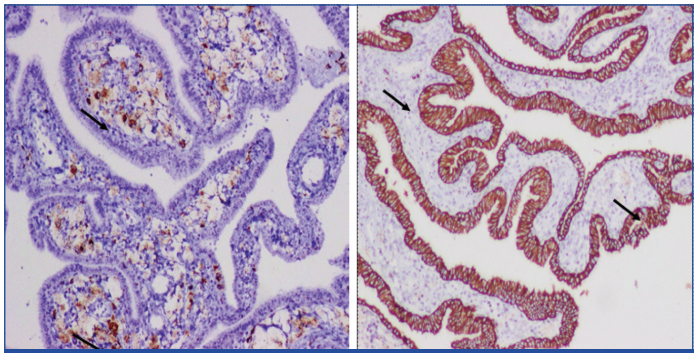
Immunohistochemistry further established the diagnosis of xanthoma with the foamy macrophages being positive for CD68 and negative for epithelial marker CK [Table/Fig-5,6]. After the diagnosis was made, the patient's serum cholesterol was assayed and found to be normal. The postoperative period was uneventful and the patient



[Table/Fig-1]: Ultrasound image showing hydrosalpinx of the right fallopian tube.
[Table/Fig-2]: Gross examination of fallopian tube showing dilated lumen and thickened wall with yellow to white plaques on it (Arrow). (Images from left to right)



[Table/Fig-3]: Hyperplastic tubal plicae line by intact tubal epithelium with the sub-epithelium showing sheets of foamy macrophages (Arrow) (H&E Stain, 10X).
[Table/Fig-4]: Sheets of foamy macrophages with abundant vacuolated cytoplasm and peripherally placed nucleus (Arrow) (H&E Stain, 40X). (Images from left to right)



[Table/Fig-5]: Foamy macrophages showing CD68 positivity (Arrow) (10X).
[Table/Fig-6]: Foamy macrophages showing CK negativity (Arrow) (10X). (Images from left to right)

was discharged with advice for follow-up and was doing well till the last follow-up which was one month postsurgery.

DISCUSSION

Xanthomas also known as xanthelasmas are rare reactive histiocytic proliferations characterised by localised lipid deposits in an organ or organ system which can be an important sign of systemic disease. They can involve any anatomical site with predilection towards the skin and subcutaneous tissue. Xanthomas can occur in any age group, in patients with predisposing systemic conditions like familial hypercholesterolemia, they tend to occur typically in second decade [1]. The incidence of xanthelasmas range from 0.3%-1.5% in India [2]. The most common visceral site is Gastrointestinal (GI) tract where they are incidentally discovered during the upper GI endoscopy. Xanthomas of perineum and fallopian tube are extremely rare. Grossly, they appear as yellow/cream coloured plaque or nodules and are histologically characterised by the presence of foamy histiocytes containing lipids [3]. Visceral accumulation of foamy macrophages is extremely rare, when occurs it is almost exclusively seen in GI tract, specifically stomach [4]. Xanthomas can occur in any age group, but in patients with predisposing systemic conditions like familial hypercholesterolemia they tend to occur typically in second decade [1].

They are non tumourous but reactive histiocytic proliferations that can occur in both hyperlipidemic and normolipidemic patients. The development can be attributed to leakage of lipid from the vasculature into the surrounding tissues, where these lipids are subsequently phagocytised by macrophages and "foamy" macrophages are formed due to accumulation of the undegraded cholesterol in these macrophages [5].

Xanthoma and xanthogranulomatous salpingitis are closely related to each other and are considered as close differentials [6]. Xanthogranulomatous salpingitis are rare inflammatory conditions characterised by aggregates of lipid laden foamy macrophages or histiocytes (xanthoma cells) which is a pathologic hallmark of the lesion. Along with the xanthoma cells, infiltration of acute or chronic inflammatory cells is also seen [7]. The foamy histiocytes admixed with plasma cells has been described as consistent finding irrespective of the age of the lesion [8,9]. Another close differential diagnosis is pseudoxanthogranulomatous salpingitis which is generally associated with long standing endometriosis and presence of histiocytes which stain positive for Perls Prussian blue due to presence of haemosiderin [10].

There are many reports on xanthogranulomatous salpingitis and oophoritis with pelvic endometriosis, Pelvic Inflammatory Disease (PID), Intrauterine Device (IUD), chronic endometritis and chemotherapy due to a breast malignancy as suggested causes [10-14]. However, only one case of isolated fallopian tube xanthoma has been reported in literature till date [4]. This is a rare case of fallopian tube xanthoma in a female aged 27 years with an unusual presentation of xanthoma at a rare site. The present case differs from the other xanthogranulomatous cases with regards to absence of any inflammatory infiltrate, giant cells, necrosis or haemorrhage and the lesion being solely composed of foamy macrophages which are positive for CD68. The histological appearance of the current lesion irrespective of the aetiology is closely related to gastric xanthelasma, thus, justifying the use of same terminology for a similar lesion in fallopian tube.

CONCLUSION(S)

A high degree of suspicion is required, and xanthomas can be considered in the differential diagnosis of lower abdominal pain in reproductive age group. It is difficult to diagnose this lesion because of the non specific nature of complaints in the majority of the cases, and the final diagnosis is usually elusive until the histopathology is performed. Mismanagement of this condition can be avoided by correct diagnosis and complete surgical excision. Till date no studies have shown recurrence after surgical excision.

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