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Pathology

ACTINOMYCOSIS WITH OVARIAN FIBROMA- A RARE ENTITY.

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ABSTRACT Actinomycosis is a chronic suppurative granulomatous inflammation caused by at least six known species; the most common being Actinomyces israelii.

Ovary is a rare site for pelvic actinomycosis and, it's association with ovarian fibroma, which is a benign tumor arising from stromal elements of ovary is not well documented.

Presenting herewith, one such rare case in a 35-year-old female who was clinically diagnosed as a case of bilateral solid-cystic ovarian mass which on gross showed features of fibroma of ovary and on microscopy multiple actinomycotic colonies were seen within the ovarian stroma.

Based on these findings, a rare diagnosis of Actinomycosis with ovarian fibroma was made.

KEYWORDS : Actinomycosis, pelvic inflammatory disease, tubo-ovarian mass.

Introduction:

Actinomycosis is a chronic, suppurative, granulomatous infection characterized by formation of abscesses, multiple draining sinuses and an appearance of tangled mycelial masses or granules in the discharges and tissue sections. [1] Pelvic actinomycosis is relatively rare and still rarer is ovarian actinomycosis as the ovary is said to be resistant to the surrounding inflammatory disease.

Actinomyces israelii is said to be the most common type among the 6-known species. [2]Out of the 3 forms of presentation, pelvic actinomycosis is rare and accounts for 3% of all actinomycotic infections in humans.[3]

The anatomical structure of the ovary is thought to resist surrounding inflammatory disease, hence making ovarian actinomycosis an even rarer entity. [3]Moreover, many times it has been found to mimic advanced pelvic malignancy. Therefore, diagnosis and timely recognition and management is often delayed leading to increased morbidity. [3] Ovarian fibroma is a rare, benign tumor that constitutes 3 % of all ovarian tumors with about one-third of the patients said to be asymptomatic. [4]The rest are often said to present with Meig's syndrome- i.e. ovarian mass along with hydrothorax and ascites.

Extensive literature search didn't reveal any association of fibroma with ovarian Actinomycosis and therefore it should be considered as a differential diagnosis of ovarian tumor with pelvic inflammatory disease.

Case Report:

A 35-year-old female, came with a complaint of on and off pain in the abdomen for 2 years with no aggravating or relieving factors. She was a known case of hypothyroidism and diabetes mellitus on irregular therapy. She had a history of tubal ligation 13 ½ years ago. There was no history of Intra Uterine Contraceptive Device usage.

On examination, a palpable lump was felt per abdomen. General physical examination and laboratory investigations including CA 125 were within normal limits. Ultrasound and Magnetic Resonance Imaging revealed a mixed solid-cystic lesion with internal vascularity in bilateral adnexae from which ovaries were not separately seen-suggestive of bilateral tubo-ovarian mass.

We received a specimen labelled as left ovarian mass, well-circumscribed soft tissue measuring: 3.5 x 3 x 2.5 centimeters. The external surface was well-encapsulated and nodular. Cut surface was solid, homogenous, grey-white with focal yellowish and myxoid areas. Microscopy showed a benign tumor of the ovary composed of closely packed spindle – shaped stromal cells arranged in a feather stitched pattern suggestive of fibroma. (Figure 1). Many foci within the tumor showed characteristic colonies of Actinomyces displaying radiating filaments with intense basophilic staining. (Figure 2) Colonies were branched and surrounded by eosinophilic, radiating, hyaline material representing the Splendore-Hoeppli phenomenon and dense mixed inflammatory infiltrate. Focal areas showed myxoid change, hyaline globules and numerous ill-formed epithelioid cell granulomas.

Special staining with PAS showed strong positivity for Actinomycosis. (Figure 3) Based on these features, a final diagnosis of Actinomycosis with ovarian fibroma was made.

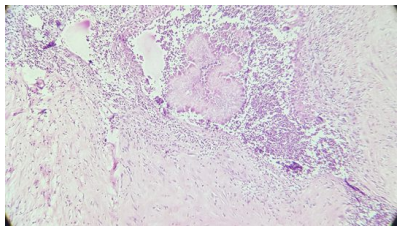

Fig 1: Actinomycotic colonies surrounded by dense inflammation in an ovarian fibroma

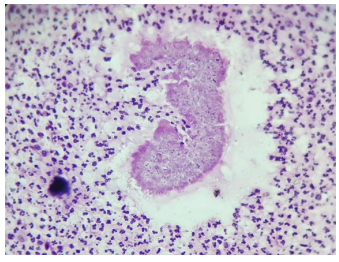

Figure 2: Colonies of Actinomyces surrounded by inflammation

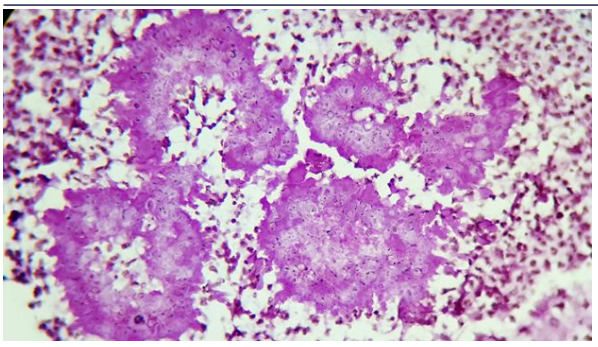

Figure 3: Colonies stained positive for PAS stain

Discussion:

Actinomycosis is a chronic, suppurative, granulomatous bacterial infection. In humans, Actinomyces israelii is the most common species known to cause infection. It is a slow growing, gram-positive, filamentous, non-spore forming, anaerobic, micro aerophilic bacteria. They are commensals, found frequently as normal inhabitant flora of the oropharynx, gastrointestinal tract and vagina of healthy individuals.

Since these infections are endogenous in origin, the organisms normally present are of low pathogenicity. They do not invade mucosal barriers and are known to multiply and spread only in case of injured and necrotic tissue.[1]

Before the antibiotic era, the fatality of the disease was nearly 80%, irrespective of the site of infection. [5]According to some reports, pelvic actinomycosis leading to genital tract involvement is usually secondary to infection in the gastrointestinal tract. However, in our case, no evidence of genital tract involvement was seen.Various studies have shown that Intra Uterine Device users are at an increased risk of contracting pelvic actinomycosis. The first report of such an association was by Barth in 1928. [3]It is said that retained tampons and pessaries are also associated with occurrence of pelvic actinomycosis in women.[1]

The clinical features of pelvic actinomycosis follow a prolonged, insidious course with vague, variable and often non-specific symptoms. It is reported that a delay in diagnosis may lead to a frozen-pelvis and rarely prove to be fatal. Hence, it must be considered as a differential in cases of tubo-ovarian masses.

Nearly 85% of women with pelvic actinomycosis have been reported to have had an Intra Uterine Device in place for more than 3 years. [3] However, our patient did not have a history of Intra Uterine Device usage. Clinical features in a case of pelvic actinomycosis are said to be vague, which includes long standing, dull abdominal pain, backache and vaginal discharge.

In the present case, patient complained of intermittent, dull abdominal ache for 2 years. It is said that radiological findings mimic those of malignancies. Due to this, pre-operative diagnosis of Actinomycosis is rarely made.[3]

Histological diagnosis is based on microscopic identification of actinomycotic colonies with Sulphur granules and dense inflammation. Culture is said to be the gold standard for identifying the organism but due to special handling and time required it is not usually employed. [2] In our case microscopy showed classical histomorphological features of actinomycosis and was confirmed on PAS stain. As the tissue was formalin fixed, culture was not done.

Complications of long standing Actinomycotic infection are said to include endometritis, salpingo-oophoritis, and tubo -ovarian abscess. [3]It is however imperative to rule out other infections which show presence of granules before a confirmatory diagnosis is made as the mode of treatment would depend on the correct identification of the organism. Gram stain is useful to differentiate between gram positive colonies of actinomycosis and gram-negative colonies of Pseudomonas / Proteus / Staphylococcus. [5]Fungal hyphae are broader than actinomycotic filaments [5] and therefore was ruled out in our case.Nocardia and Streptomyces have gram positive branching filaments but they do not show characteristic eosinophilic clubs that radiate around actinomycotic granules.

Once diagnosed, primary management is said to be by antibiotics and surgery whenever indicated. [3]

In our case post-operative antibiotics were given.

Ovarian fibroma is a rare, benign tumor that constitutes 3% of all ovarian tumors. [4] About one-third of the patients are said to be asymptomatic. [4] The mean age of the patients is 45 years.[4]

They often present clinically with Meig 's syndrome, a diagnosis of ovarian mass along with hydrothorax and ascites. None of the above features were seen in our patient. Extensive research showed no reports of an association of fibromas with actinomycosis and so we can conclude that the diagnosis of fibroma was an incidental finding and actinomycosis may be seen with other non-neoplastic or neoplastic lesions.

Hence, actinomycosis of the ovary should be considered in the differential diagnosis of patients with ovarian mass and suspected pelvic inflammatory disease.

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