Perplexing Multifocal Abdominopelvic Mass - turned out to be actinomycosis

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Abstract

A 34 year old female presented with complaints of lower abdominal pain and abnormal uterine bleeding for 2 months. On examination subcutaneous swelling on anterior abdominal wall and bilateral pelvic adnexal mass was noted. The lesion was clinically and radiologically, proposed as tumor. Postoperatively on histopathological examination it was diagnosed as multifocal abdominopelvic actinomycosis. Patient was successfully treated with parenteral antibiotics and disease free at follow-up. The nonspecific clinical and radiological features of Actinomycosis are more likely to be misinterpreted as tumor, while making delay in accurate diagnosis. Therefore use of modern ancillary investigatory procedures can help in early appropriate diagnosis and spare the patient from morbid surgical procedures.

Keywords: Actinomycosis, Multifocal, Abdominopelvic, Misinterpretation, Tumor.

Introduction

Abdominopelvic mass in young female can be due to varied etiology and may present with overlapping clinical features, which delays the definite diagnosis and subsequent patient management. Abdominopelvic actinomycosis accounts about 10 – 20% of the reported cases.(1) Its prompt clinical diagnosis is difficult because of its rarity and paucity of clinical symptoms even with extensive organ involvement. Hence it is rightly designated as "most misdiagnosed disease".(2) In this report, we present a case of multifocal abdominopelvic actinomycosis in young women, who had clinical and radiological features suggestive of neoplasm.

Case Presentation

34 years young female presented with complaints of lower abdominal pain and abnormal uterine bleeding for 2 months. Patient is a known case of uncontrolled type 2 diabetes mellitus and hypertension for 3 years. Patient had two full term normal institutional delivery. She gave history of intrauterine contraceptive device usage after first child birth for short duration of 6 months and puerperal sterilization after second child birth.

Firm, tender, subcutaneous mass in the anterior abdominal wall measuring 5x6cm was noted on per abdomen examination. On gynecological examination pedunculated mass in both adnexa was noted. Clinically differential diagnosis of desmoid tumor and endometriosis was made.

On radiological examination, enhancing lesion indenting dome of bladder with linear extension to infraumblical region and infiltration of abdominopelvic wall was noted. Radiological differential diagnosis of urachal remnant malignancy and endometriosis was given.

Fine needle aspiration cytology of subcutaneous swelling in anterior abdominal wall was done. Smears studied showed acute and chronic inflammatory cells of neutrophils, lymphocytes, cyst macrophages with foci of fibrosis in a fibrinopurulent background (Fig. 1a & b). Therefore, the impression of infective lesion was given.

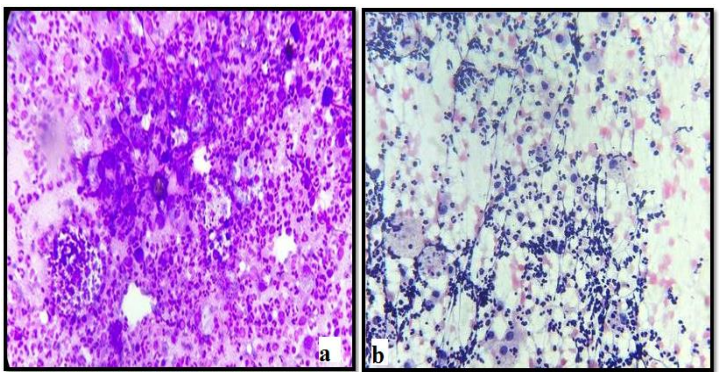

Fig. 1: A- Smears studied showed acute and chronic inflammatory cells of neutrophils, lymphocytes, cyst macrophages with foci of fibrosis in a fibrinopurulent background (Giemsa stain 10x). B- Papanicolaou stain 10x

Patient was taken up for surgical procedure of total abdominal hysterectomy with bilateral salphingectomy and excision of subcutaneous mass in anterior abdominal wall which was extending intraperitoneally up to dome of bladder. Histopathologically, macroscopic examination, of subcutaneous swelling was 5x6cm, firm skin covered nodule, cut surface showing solid fibrous region. Grossly, hysterectomy specimen was measuring 10 x9 x5 cm; cervix and endometrium were unremarkable. Both side tube with ovary were enlarged each measuring 6 X4.8X4.5cm and 6.5X5.3X4 cm respectively and its external surface was congested. Cut surface of tubes and ovaries showed yellowish pus filled areas along with fibrotic region (Fig. 2a & b).

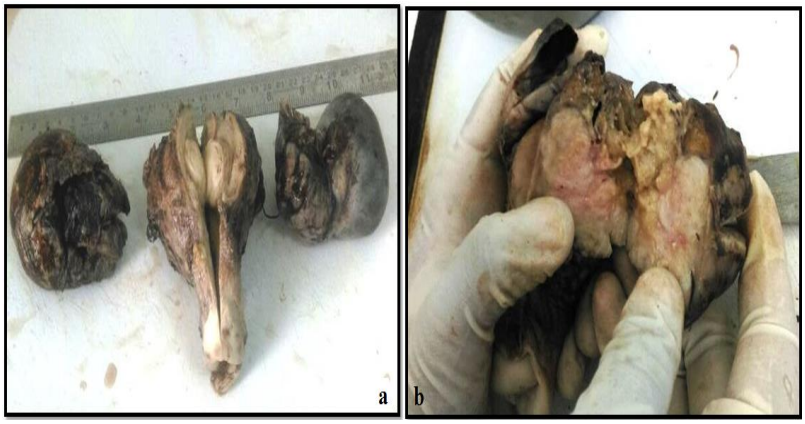

Fig. 2: A- Macroscopic examination of hysterectomy specimen was measuring 10X9X5 cm; cervix and endometrium were unremarkable. Both tubes and ovaries were enlarged and its external surface was congested. B- Cut surface of tubes and ovaries showed yellowish pus filled areas along with fibrous region

Microscopic examination of fallopian tubes, ovaries and separately excised subcutaneous mass showed intense inflammatory reaction surrounding filamentous basophilic structure, covered by eosinophilic club like material (Fig. 3a-d). Inflammatory cells like neutrophils, macrophages and foreign body type of giant cells along with granulation tissue. Uterine endomyometrium and cervix were unremarkable. Based on these features, diagnosis of multifocal abdominopelvic actinomycosis was made. Actinomyces colonies showed positivity with histochemical stains (Fig. 4a) like PAS, GMS, and Gram's stain and negativity with modified Ziehl Neelson stain (Fig. 4b).

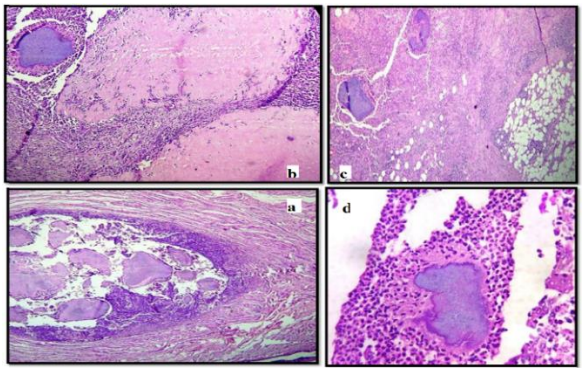

Fig. 3: Microscopic examination of mass showed intense inflammatory reaction surrounding filamentous basophilic structure, covered by eosinophilic club like material a) Fallopian tube (H&E 10x) b) Ovary (H&E 10x) c) Subcutaneous tissue (H&E 10x) d) High power magnification of actinomyces lesion (H&E 40x)

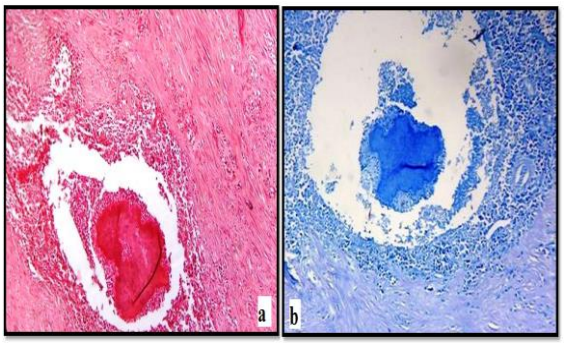

Fig. 4: Actinomyces colonies showed positivity with histochemical stains a) PAS 10x b) Gram's stain 10x

Discussion

This is a case of 34 year young women with multifocal abdominopelvic mass, histopathologically proven to be actinomycosis infection but clinically, radiologically mimicking a tumor.

Actinomycosis is a chronic disease caused by anaerobic or microaerophilic bacteria, primarily belonging to actinomyces genus. It is a normal flora and colonizes in mouth, colon, and vagina. It is a low virulent organism which can cause disease in any part of the body, only when normal mucosal barrier is disrupted and spreads to surrounding tissues regardless of tissue planes due to release of proteolytic enzymes. Similarly, ovarian actinomycosis occurs only when its mucosa is broken by

ovulation.[3] Actinomycosis clinically presents as abscess, fistula or mass like lesion, mimicking benign or malignant tumor.[4]

Abdominopelvic actinomycosis accounts for 10 - 20% reported cases.[1] Pelvic actinomycosis is rare accounting for 3% of all human actinomycotic infections.[3] Ovarian actinomycosis is rarer because structure of an ovary is resistant to surrounding inflammatory disease.[5] A slight male preponderance is noted 1.5:1 to 3:1. Actinomycosis is most common in patients between 4th to 6th decade of life. There is no known racial, environmental or geographic predisposing factor.[6] Actinomycosis of female genital tract was thought to originate from ascending infection of bacteria.[7] Curtis et al reported the longer the use of intrauterine device, greater the risk for actinomycosis of genital tract. Other risk factors include abdominal surgeries, tuboovarian abscess and ruptured appendicitis.[8,9] There are literatures mentioning the isolated ovarian actinomycosis can occur without usage of intrauterine devices.[10]

Actinomycosis is designated as most misdiagnosed disease and is listed as rare disease by the office of rare disease (ORD) of the national institute of health (NIH).[13] It is named such because of atypical presentation, radiological features and nonspecific clinical presentation.

One of the characteristics of actinomycosis is the lack of immediate tissue reaction after implantation of the organism. It usually requires 6weeks or longer for an actinomycotic swelling to breakdown and discharge pus.[6] The sulphur granule measures 0.4 – 4mm. The size of bacterium renders lymphatic spread impossible; hence regional lymphadenopathy is uncommon or develops late.[12] Similar to our case, Yoo Kyung Lee reported pelvic actinomycosis without fever, leukocytosis or elevated ESR.[13]

Preoperative diagnosis with FNAC may be impossible as the actinomycotic lesion is surrounded by intense fibrosis and extensive inflammatory tissue,[14] in our case it was reported as infective lesion. Negative culture, cannot exclude diagnosis of actinomycosis. Negative culture may result from previous antibiotic usage, improper specimen collection and transport techniques or insufficient incubation period.[15] In our case, culture was not done as patient was on antibiotics prior to surgery.

Unfortunately, in most cases, actinomycosis is diagnosed after surgery with classical histopathological feature of sulphur granules formed by colonies of organisms giving an amorphous appearance at center, surrounded by rosette of clubbed filaments and dense neutrophil aggregation.[16]

Special stains PAS and GMS are required to differentiate pseudoactinomyces granules of Nocardiosis and Streptomyces from actinomycosis. In the former, PAS & GMS show negative reaction whereas later show positive.[17]

Conclusion

Preoperative diagnosis of multifocal abdominopelvic mass remains difficult as clinical features and radiological features are nonspecific. Definite diagnosis is usually achieved by histopathological examination and histochemical stains highlighting pathognomonic sulphur granules.

Atypical presentation of multifocal abdominopelvic actinomycosis mass may be mistaken for tumor, but still it should be one of the differential diagnosis, as actinomycosis is entirely curable with parenteral antibiotics and debridement of infected tissue. Preoperative investigations like CT guided core biopsy or incision biopsy followed by frozen section examination of the tissue can minimize risk of overtreatment and unnecessary complications.

References

1. Cintron JR, Del Pino A, Duarte B, Wood D. Abdominal actinomycosis. Dis Colon Rectum. Jan 1996;39:105-8.
2. Harrison's Principles of Internal Medicine. 17thedition,Chapter 156:996.
3. Simsek A, Perek A, Cakcak IE, Durgun AV. Pelvic actinomycosis presenting as a malignant pelvic mass: A case report. J Med Case Reports 2011;5:40.
4. Kumar N, Das P, Kumar d, Kriplani A, Ray R. Pelvic actinomycosis mimicking: an advanced ovarian cancer. Indian J Pathol Microbiol. 2010 Jan-Mar;53(1):164-5.
5. Koshiyama M, Yashida M, Fujii H, Nanno H, Hayashi M, Tauchi K, et al. Ovarian actinomycosis complicated by diabetes mellitus simulating an advanced ovarian carcinoma. Eur J Obstet Gynaecol Reprod Biol 1999;87:95-9.
6. Rania Rodan. Unusual Presentation of Actinomycosis Misdiagnosed as Severe Periodontal Destruction. JRMS Mar 2012;19(1):53-56.
7. Nozawa H, Yoshinao Y, Yasuhiko M, Arita S, Aisaka K. Pelvic actinomycosis presenting with a large abscess and bowel stenosis with marked response to conservative treatment: a case report. Journal of medical case reports. 2007,1:141.
8. Curtis EM, Pine L. Actinomyces in the vagina of women with and without intrauterine contraceptive devices. Am J Obstet Gynecol 1981;140:880–884.
9. Garner JP, Macdonald M, Kumar PK. Abdominalactinomycosis. Int J Surg. 2007;5(6):441-448.
10. Marwah S, Marwah N, Singh I, Singh S, Gupta A and Jaiswal TS: Ovarian actinomycosis in absence of intrauterine contraceptive device: an unusual presentation. Acta Obstet Gynecol Scand 84:602-607,2005.
11. Talwar OP, Ghosh A, Pradhan S, et al. A prospective and retrospective study of actinomycosis in last five years at Manipal Teaching Hospital, Pokhara, Nepal. Kathmandu University Medical Journal 2007;5(4):488-491.
12. Belmont MJ, Behar PM, Wax MK. Atypical presentation of actinomycosis. Head and Neck 1999;21:264-268.
13. Lee YK, Bae JM, Park YJ, Park SY, Jung SY. Pelvic Actinomycosis with hydronephrosis and colon stricture simulating an advanced ovarian cancer. J Gynecol Oncol Vol.19, No.2: 154-156, June 2008.
14. Teresa Pusiol, Doriana Morichetti, Corrado Pedrazzani and Francesco Ricci. Abdominal – Pelvic Actinomycosis Mimicking Malignant Neoplasm. Infectious diseases in Obstetrics and Gynecology 2011, vol 2011:1-5; doi: 10.1155/2011/747059.

15. Sugano S, Matuda T, Suzuki T, Makino H, Iinuma M, Ishii K, Ohe K, Mogami K. Hepatic actinomycosis: case report and review of the literature in Japan. J Gastroenterol 1997;32:672-676.
16. Hung-Wei Chi, Ya-Sung Yang, Te-Yu Lin, Jung – Chung Lin, Feng – Yee Chang and Ning – Chi Wang. Tubo – ovarian Actinomycosis Complicated with Hepatic Abscess. J Med Sci 2010;30(6):261–264.
17. S. Kamprath, A. Merker, R. K¨uhne-Heid, and A. Schneider, "Abdominal actinomycosis with IUD," *Zentralbl Gynakol Journal*, vol. 119, no. 1, pp. 21–24, 1997.