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Original Article

Enterovesical Fistula Associated with Pelvic Actinomycosis: Case Report and Literature Review

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Abstract **Actinomycosis is a rare chronic, suppurative infection caused by the bacteria of the Actinomyces species. This chronic infection may present with a variety of symptoms. Florid abscess formation with fistulation, abundant granulation and dense surrounding fibrosis are common. Diagnosis prior to, or even during surgery is rare and the findings are usually mistaken for inflammatory process or malignancy. Abdominal actinomycosis has been recognized for over 150 years yet is largely unknown to most clinicians. It was once described as "the most misdiagnosed disease".**

We report here a 59-year-old woman who presented with enterovesical fistula secondary to pelvic actinomycosis. Its recognition is important as clinical and radiological findings may mimic malignancy and lead to radical and unnecessary surgery. A high index of suspicion for this rare but devastating condition must be maintained. To our knowledge, this is the second reported case of enterovesical fistula secondary to pelvic actinomycosis in the English language literature. The literature on abdomino-pelvic actinomycosis was reviewed.

INTRODUCTION

A fistula is an abnormal communication between two epithelialized surface.[1,2] After Rufus of Ephesus described the passage of feces and air through the urethra in 200 AD[3], Cripps H in 1888 published the first series of what is now known as enterovesical fistula.[4]

Enterovesical fistula is a rare and serious condition and can cause significant morbidity. Actinomycosis is a rare chronic infectious disease resulting in suppurative and granulomatous inflammation caused by Actinomyces species.[5] Actinomyces is a gram-positive, anaerobic filamentous bacteria.[5-7] It is normally present in the GI tract and female genital tract.[5,6,8-10] The three main clinical forms of Actinomycosis are cervicofacial (31%-65%), abdomino/pelvic (20%-36%), and thoracic (15%-33%).[5,7,11-14]

Actinomycosis was first described by Israel J in 1878[15] and it was first diagnosed in a live patient by Ponficke in 1879.[16,17] Clinical symptoms and signs of pelvic actinomycosis are not specific.[5] It has a propensity to mimic malignancy due to its capacity to form a mass lesion and to invade surrounding tissues.[5,18,19] The association between actinomycosis and enterovesical fistula is extremely rare.

Herein, we report a case of enterovesical fistula associated with pelvic actinomycosis that was treated successfully with surgical resection followed by

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antibiotic therapy. To our knowledge, this is the second reported case in the English language literature.

Case Report

A 59-year-old woman presented with a history of lower abdominal pain for one month. She also passed urine frequently, and afterwards flatus often escaped. The urine was turbid, having yellowish granular precipitate, with fecal odor. She denied any previous abdominal surgery or use of intrauterine device. She had a history of blunt abdominal trauma at the lower abdomen from a fall two months ago without any treatment. Physical examination revealed mild lower abdominal tenderness without mass. She had no fever. Per rectal examination was normal.

On insertion of the urethral catheter, feculent fluid was obtained and the diagnosis of enterovesical fistula was established. Abnormal laboratory tests included mild leukocytosis (white blood cell count of 11,400 /mm^3) and pyuria. Colonoscopy yielded normal findings. Computerized tomography (CT) showed large mass (5 cm in diameter) at the anterior abdomino-pelvic region with invasion to right rectus abdominis muscle, ileum and urinary bladder (right anterolateral wall). The differential diagnosis from CT scan was abdominal sarcoma or chronic abscess.

Surgery was performed with the diagnosis of enterovesical fistula possibly from malignancy. Laparotomy revealed a firm mass (5 cm in diameter) at the anterior abdominal wall with dense adhesion and invasion to urinary bladder and ileum, suggesting a malignant tumor. The involved ileum, urinary bladder, and anterior abdominal wall mass were resected en bloc. Primary anastomosis of the remaining portions of the small bowel was performed with staples. The bladder was repaired with absorbable sutures in two layers. Urinary catheter was retained for two weeks. No post-operative cystogram was performed at the time of catheter removal as it has been established that epithelialization of the bladder mucosa is completed within one week.[20] Cut surface of the resected specimen revealed ill-defined, firm, creamy yellow mass in the mesenteric fat of the ileum. The mass directly invaded the ileum, bladder and anterior abdominal wall (Figures 1, 2, and 3). Pathological examination showed

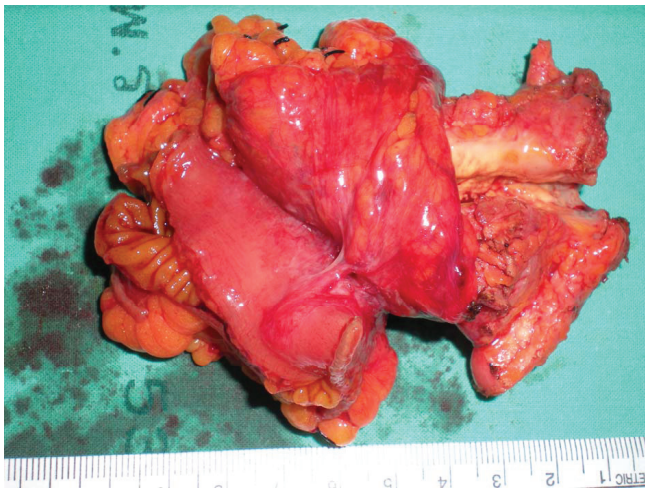

Figure 1 The resected specimen consisting of a segment of ileum, parts of the bladder and anterior abdominal wall.

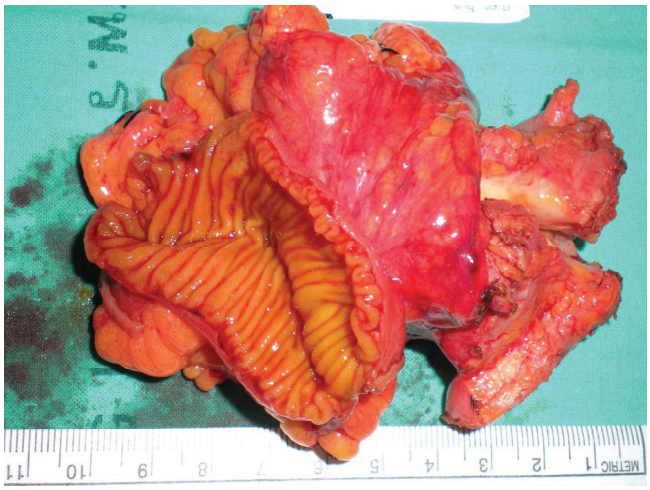

Figure 2 The resected specimen showing normal ileal mucosa.

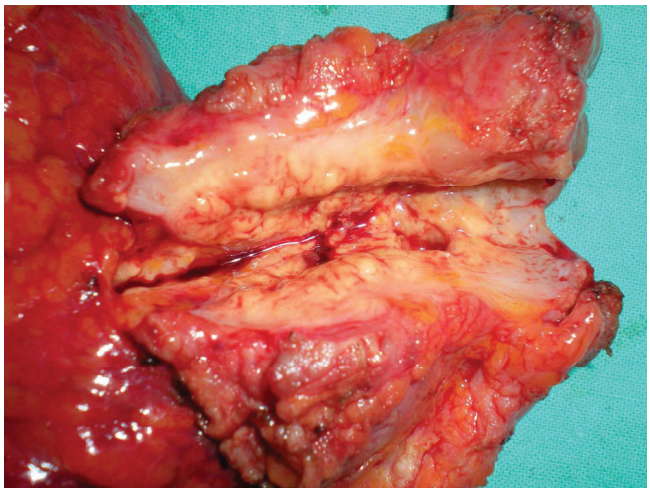

Figure 3 A cross-sectional view of the abdominal mass in the resected specimen.

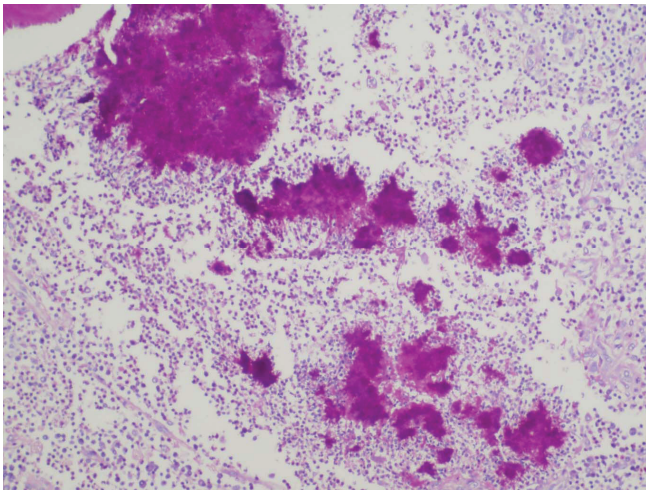

Figure 4 Microscopic view of a section of the abdominal mass, showing inflammatory cells and clusters of actinomyces (Hematoxylin & eosin stain, low power magnification).

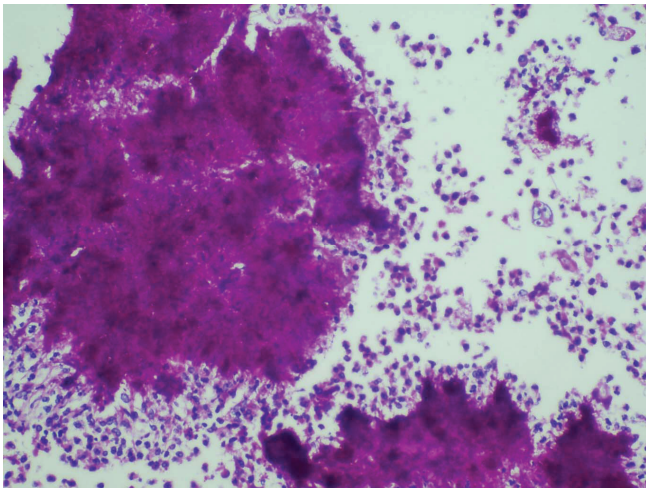

Figure 5 A high power magnification of the actinomyces cluster in the same section shown in figure 4.

actinomycosis with suppurative granulomatous inflammation involving ileum and urinary bladder wall (Figures 4 and 5). The post-operative recovery was uneventful. After surgery, the patient was given oral penicillin for 2 months. She remained well on the last follow-up.

Discussion

The intestinal origin of the enterovesical fistula include the sigmoid colon (68%-88%), rectum (47%-13%) and ileum (7.2%-10%).[21-23] Common causes of colovesical fistula include diverticulitis and colon cancer.[24,25] Most rectovesical fistulae are caused by rectal tumor or trauma.[24,26] Crohn's disease more commonly causes ileovesical fistula.[24] Uncommon causes of the fistula include pelvic irradiation, foreign bodies (eg. urinary catheter, fish or chicken bone in the bowel), infection (eg. tuberculosis, syphilis) and bladder cancer.[21,25]

Enterovesical fistulae are diagnosed primarily by clinical findings.[3,21,27] Pathognomonic features include fecaluria, pneumaturia, and recurrent urinary tract infection.[27] The majority of patients present with urological symptoms. Presenting features include fecaluria (40%-93%), pneumonia (40%-85%), recurrent urinary tract infection (47%-73%), systemic sepsis (14%), and passage of urine per rectum (up to 40%).[21-23,28,29]

Cystoscopy, colonoscopy and CT have been the mainstays of investigation, but they have less than ideal sensitivity and specificity.[24] These investigations do not always define or confirm the presence of a fistula.[22] There is little evidence supporting a specific investigative algorithm.[30] Nontheless, the CT scan remains the current standard for diagnosis and identifying the underlying etiology and to facilitate surgical planning.[21,24] A CT scan should be performed before bladder instrumentation,[21] and used especially for those patients in whom an extracolonic mass is suspected or those with malignancy.[22]

All patients undergoing enterovesical fistula surgery should have a pre-operative colonic evaluation. Colonoscopy may not demonstrate the fistula but will reveal the cause as well as exclude other pathologies within the colon.[22] Enterovesical fistula rarely close spontaneously (2%) and surgery is generally required.[21,22] It is recognized that without surgical intervention 75% of patients may die from sepsis within 5 years.[31]

Abdominal actinomycosis has been recognized for over 150 years yet remains largely unknown to most clinicians.[5,15] In 2008, about 34 actinomyces species are recognized, 20% of which have been recovered from the human body.[32] Actinomycosis in the human is most commonly caused by Actinomyces israelii.[5,7,33,34] They are normal commensal bacteria of the human GI tract and female genital tract.[13,35,36] This bacteria does not invade intact mucous membrane,[37] but becomes pathogenic in the presence of damaged mucosal barrier. Factors that precipitate abdominal actino-

mycosis include previous abdominal surgery, bowel perforation, and foreign bodies such as intrauterine devices.[38]

Henderson, in 1973, first reported an increase in pelvic actinomycosis in women who used intrauterine contraceptive device (IUD).[39] Although frequently cited, the association between IUD use and development of pelvic actinomycosis is controversial. The risk of pelvic actinomycosis resulting from IUD use is very low. Only about 92 reported cases exist in the published English language literature since 1996, despite 30 million patient-years of IUD use.[40] Definite risk factors for developing pelvic actinomycosis have not yet been established. In the majority of cases, as in the present report, there is no obvious portal of entry, and the pathogenesis of the disease is not always clear. The only possible predisposing factor in our case is blunt trauma at lower abdomen two months before the onset of symptom.

Actinomycosis produces granulomatous inflammation and mass formation,[34] later undergoing a softening process and abscess formation which may rupture into the adjacent bowel and bladder producing enterovesical fistula. The diagnosis of actinomycosis is usually ascertained after histopathological examination of the resected specimens.[13-15,21,32] The possibility of achieving a positive culture is only 13%-30%.[11,14,15] The high failure rate is due to various factors including prior antibiotic therapy, overgrowth of concomitant organism or inadequate methodology for anaerobic culture.[32] Generally, a negative culture result does not exclude actinomycosis.[15]

The diagnosis of pelvic actinomycosis is rarely made pre-operatively.[11,15,33] The absence of surgical planes and extensive spreading to adjacent structures and organs, as in the present case, is due to infiltrative tissue damage as a result of the release of proteolytic enzymes by the organism. This phenomenon may result in the condition of being mistaken for pelvic malignancy or "frozen pelvis"[10,13] and unnecessary radical surgery may be performed with high morbidity and occasional fatality.[41] Awareness of this entity may enable the surgeon to minimize major organ resections. Our report should help raise the awareness of this rare condition, to be included in the differential diagnosis of enterovesical fistula.

Uncomplicated actinomycosis can be medically treated by antibiotics. A prolonged treatment course is required because of the poor penetration of antibiotics into the fibrotic tissues.[5,33] Actinomyces species are sensitive to penicillin, but the duration of treatment varies from several weeks to months to achieve permanent recovery.[5] The recommended antibiotic regimen after surgery is oral penicillin 2-4 gm/day for 3-12 months.[11,16,33,34,42] However, if the area of actinomycosis infection can be completely removed surgically, a shorter period of antibiotic therapy could be effective.[33,42]

Even though the diagnosis of abdominal actinomycosis can be made before surgery, surgical resection is usually required. Surgery is valuable as a therapeutic adjunct and helps relieve the symptoms and removes the infected fibrotic tissue. Surgical treatment of this entity should include the resection of all infected tissue, and be combined with antibiotic therapy postoperatively. Piper et al. in 1969 reported the first case of ileovesical fistula associated with actinomycosis in a 69-year-old female.[43] To our knowledge, this is the second reported case in the English language literature.

Conclusion

Enterovesical fistula associated with actinomycosis is an extremely rare condition. Our report may help increase the awareness of this rare but important and curable disease. Maintaining a high index of suspicion for this condition will help prevent unnecessary radical surgery for presumed malignancies. Even though pelvic actinomycosis is rare, it should be included in the differential diagnosis of the cause of enterovesical fistula.

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References

1. Schecter WP, Hirshberg A, Chang DS, et al. Enteric fistulas: principles of management. J Am Coll Surg 2009;209:484-91.
2. Pickhardt PJ, Bhalla S, Balfe DM. Acquired gastrointestinal fistulas: classification, etiologies, and imaging evaluation. Radiology 2002;224:9-23.

3. Ferguson GG, Lee EW, Hunt SR, et al. Management of the bladder during surgical treatment of enterovesical fistulas from benign bowel disease. J Am Coll Surg 2008;207:569-72.
4. Cripps H. Passage of gas and feces through the urethra: colectomy, recovery, remarks. Lancet 1888;2:619-20.
5. Choi MM, Beak JH, Lee JN, et al. Clinical features of abdominopelvic actinomycosis:report of twenty cases and literature review. Yonsei Med J 2009;50:555-9.
6. Kaszuba M, Tomaszewska R, Pityñski K, et al. Actinomycosis mimicking advanced cancer. Pol Arch Med Wewn 2008; 118:581-4.
7. Choi MH, Hong DG, Seong WJ, et al. Pelvic actinomycosis confirmed after surgery:single center experience. Arch Gynecol Obstet 2009 Jun 30. (Epub ahead of print)
8. Fernández-Aceñero MJ, Silvestre V, Fernández-Roldán R, et al. Gastric actinomycosis:a rare complication after gastric bypass for morbid obesity. Obes Surg 2004;14:1012-5.
9. Kayikcioglu F, Akif Akgul M, Haberal A, et al. Actinomyces infection in female genital tract. Eur J Obstet Gynecol Reprod Biol 2005;118:77-80.
10. Devendra K, Chen CM. Pelvic actinomycosis masquerading as an acute abdomen from a small bowel perforation. Singapore Med J 2008;49:158-9.
11. Isik B, Aydin E, Sogutlu G, et al. Abdominal actinomycosis simulating malignancy of the right colon. Dig Dis Sci 2005;50:1312-4.
12. Ferrari TC, Couto CA, Murta-Oliveira C, et al. Actinomycosis of the colon: a rare form of presentation. Scand J Gastroenterol 2000;35:108-9.
13. Wang YH, Tsai HC, Lee SS, et al. Clinical manifestations of actinomycosis in Southern Taiwan. J Microbiol Immunol Infect 2007;40:487-92.
14. Marella VK, Hakimian O, Wise GJ, et al. Pelvic actinomycosis. Urologic perspective. Int Braz J Urol 2004;30:367-76.
15. Taga S. Diagnosis and therapy of pelvic actinomycosis. J Obstet Gynaecol Res 2007;33:882-5.
16. Wagenlehner FM, Mohren B, Naber KG, et al. Abdominal actinomycosis. Clin Microbiol Infect 2003;9:881-5.
17. Stringer MD, Cameron AE. Abdominal actinomycosis:a forgotten disease? Br J Hosp Med 1987;38:125-7.
18. Atay Y, Altintas A, Tuncer I, et al. Ovarian actinomycosis mimicking malignancy. Eur J Gynaecol Oncol 2005;26:663-4.
19. Sehouli J, Stupin JH, Schlieper U et al. Actinomycotic inflammatory disease and misdiagnosis of ovarian cancer. A case report. Anticancer Res 2006;26:1727-31.
20. Hepperlen TW, Stinson W, Hutson J, et al. Epithelialization after cystotomy. Invest Urol 1975;12:269-71.
21. Kavanagh D, Neary P, Dodd JD, et al. Diagnosis and treatment of enterovesical fistulae. Colorectal Dis 2005;7:286-91.
22. Daniels IR, Bekdash B, Scott HJ, et al. Diagnostic lessons learnt from a series of enterovesical fistulae. Colorectal Dis 2002;4:459-62.
23. Larsen A, Johansen TEB, Solheim BM, et al. Diagnosis and treatment of enterovesical fistula. Eur Urol 1996;29:318-21.
24. Ravichandran S, Ahmed HU, Matanhelia SS, et al. Is there a role for magnetic resonance imaging in diagnosing colovesical fistulas? Urology 2008;72:832-7.
25. McBeath RB, Schiff M Jr, Allen V, et al. 12-Year experience with enterovesical fistulas. Urology 1994;44:661-5.
26. Munoz M, Nelson H, Harrington J, et al. Management of acquired rectourinary fistulas. Dis Colon Rectum 1998;41: 1230-8.
27. Law WL, Chu SM. An unusual enterovesical fistula. Am J Surg 2008;195:814-5.
28. Kirsh GM, Hampel N, Shuck JM, et al. Diagnosis and management of vesicoenteric fistulas. Surg Gynecol Obstet 1991;173:91-7.
29. Liu CH, Chuang CK, Chu SH, et al. Enterovesical fistula: experiences with 41 cases in 12 years. Changgeng Yi Xue Za Zhi 1999;22:598-603.
30. Moss RL, Ryan JA. Management of enterovesical fistulas. Am J Surg 1990;159:514-8.
31. Amin M, Nallingerr R, Polk HC. Conservative treatment of selected patients due to diverticulitis. Surg Gynecol Obstet 1984;159:442-4.
32. Hall V. Actinomyces-Gathering evidence of human colonization and infection. Anaerobe 2008;14:1-7.
33. Huang CJ, Huang TJ, Hsieh JS. Pseudo-colonic carcinoma caused by abdominal actinomycosis:report of two cases. Int J Colorectal Dis 2004;19:283-6.
34. Alam MK, Khayat FA, Al-Kayali A, et al. Abdominal actinomycosis:case reports. Saudi J Gastroenterol 2001;7:37-9.
35. Garner JP, Macdonald M, Kumar PK. Abdominal actinomycosis. Int J Surg 2007;5:441-8.
36. Lee YM, Law WL, Chu KW. Abdominal actinomycosis. ANZ J Surg 2001;71:261-3.
37. Sumer Y, Yilmaz B, Emre B, et al. Abdominal mass secondary to actinomyces infection: an unusual presentation and its treatment. J Postgrad Med 2004;50:115-7.
38. Lee SH, Kim HJ, Kim HJ et al. Primary gastric actinomycosis diagnosed by endoscopic biopsy:case report. Gastrointest Endosc 2004;59:586-9.
39. Henderson SR. Pelvic actinomycosis associated with an intrauterine device. Obstet Gynecol 1973;41:726-32.
40. Fiorino AS. Intrauterine contraceptive device-associated actinomycotic abscess and actinomyces detection on cervical smear. Obstet Gynecol 1996;87:142-9.
41. Yeguez JF, Martinez SA, Sands LR, et al. Pelvic actinomycosis presenting as malignant large bowel obstruction:a case report and a review of the literature. Am Surg 2000;66:85-90.
42. Atad J, Hallak M, Sharon A, et al. Pelvic actinomycosis. Is long-term antibiotic therapy necessary? J Reprod Med 1999;44:939-44.
43. Piper JV, Stoner BA, Mitra SK, et al. Ileo-vesical fistula associated with pelvic actinomycosis. Br J Clin Pract 1969;23: 341-3.