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Abdominal actinomycosis in the last 10 years and risk factors for appendiceal actinomycosis: review of the literature

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Background/aim: Actinomycosis is a granulomatous disease caused by filamentous, gram-positive, anaerobic bacteria. Actinomycetes are commensal inhabitants of the oral cavity and intestinal tract but acquire pathogenicity through invasion of breached or necrotic tissue. In abdominal actinomycosis (AA), the appendix and ileocecal region are usually involved. The aim of this study was to characterize patients diagnosed with AA and to establish the risk factors for appendiceal actinomycosis, including a short review of the literature.

Materials and methods: We conducted a prospective cohort study of all patients diagnosed with AA in the University Hospital Marques de Valdecilla (Santander-Cantabria) from January 2003 to October 2013. We also conducted a short review of the literature.

Results: We characterized the epidemiological features of patients diagnosed with AA and risk factors for the involvement of the appendix, as most of the cases were misdiagnosed as appendicitis. Risk factors for appendiceal actinomycosis are no antecedents of carcinoma (P = 0.034) and previous incorrect diagnosis (P = 0.006).

Conclusion: AA is a rare and chronic infection. It is only reported in case reports in the literature. We present the largest series of patients diagnosed with AA. Half of the patients had morbidities, mostly carcinoma, and penicillin was the preferred treatment. More studies are needed to characterize patients affected with AA and to establish the correct treatment.

Key words: Appendiceal actinomycosis, abdominal actinomycosis, actinomycosis

1. Introduction

Actinomycosis is a rare pathology. It is a chronic granulomatous disease caused by gram-positive anaerobic bacteria from the genus *Actinomyces*. Humans are the natural reservoir and under physiological conditions these bacteria colonize the mouth, colon, and urogenital tract. Infection usually occurs only after disruption of the mucous membranes, causing invasion of adjacent tissue and forming masses with characteristic sulfur granules. Actinomycosis involves three areas mainly. The most common is the cervicofacial area (40%–50%), followed by the abdominopelvic (20%) and finally the thoracic area (15%) (1–3).

Abdominal actinomycosis (AA) is usually insidious, can affect all organs, and simulates pathologies such as diverticulitis, appendicitis, inflammatory bowel disease, and malignant tumors, among others (4).

The preoperative diagnosis is difficult to achieve, and patients are most frequently diagnosed after surgery. Antibiotic treatment with penicillin is the preferred choice and the duration is very variable (5).

The aim of this study was to characterize patients diagnosed with AA and to establish the risk factors for appendiceal actinomycosis (APA) due to the high frequency of appendiceal involvement in our series, including a short review of the literature.

2. Materials and methods

A descriptive cohort study of all patients diagnosed with AA at the University Hospital Marqués de Valdecilla (Santander, Cantabria), was performed between 1 January 2003 and 31 December 2013 to provide a current characterization of the syndrome. The University Hospital Marqués de Valdecilla is a tertiary hospital with 900 inpatient beds that provides general and emergency services to a health area of 300,000 patients.

Epidemiologic data as age, sex, and reason for admission were recorded. Antecedent risk factors of

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dental manipulation, recent abdominal surgery, presence of intrauterine device (IUD), perforation of hollow viscera, solid organ cancer, positive HIV serology, and immunosuppression as recipient of transplant or corticosteroid therapy were noted. Clinical data and the location of the AA, as well as the analytical parameters (C-reactive protein, procalcitonin, white blood cells, erythrocyte sedimentation rate, and hemoglobin), were also collected. Finally, the diagnostic method, if it was done in the preoperative or postoperative period, and the treatment received, expressed as type and duration of antibiotherapy, were also recorded.

3. Results

A total of 13 patients diagnosed with AA were treated between 1 January 2003 and 31 December 2013. Mean age at diagnosis was 48.6 ± 22.9 years and 8 (61.5%) were men. Ten patients came from the Departments of General Surgery, two from General Medicine, and one from Nephrology. All data are provided in Table 1.

No patient needed admission to the Intensive Care Unit. Stays in the hospital were 14.85 ± 17.1 days. Patients had the following antecedents: solid organ cancer (n = 3), hollow viscera perforation (n = 1), solid transplant (n = 1), abdominal surgery in the last month (cholecystectomy) (n = 1), and HIV infection (n = 1). No dental manipulations were found. In the previous 3 months three patients had received antibiotherapy and one patient also corticosteroids. The most frequent abdominal site of AA was the appendix (38.5%). The descending colon and sigma were affected in 30.7% of the patients. The ascending colon and ileum, spleen, subcutaneous fat, and mesentery were affected in one patient each. Mean values of C-reactive protein, hemoglobin, and hematocrit at the moment of diagnosis were 7.4 ± 7.2 mg/dL, 11.9 ± 2.3 g/dL, and 35.4 ± 6.2%, respectively.

The final diagnosis was reached through histological methods in 11 patients based on histologic identification of actinomycotic sulfur granules, with gram-positive bacilli radiating from these granules. Diagnosis confirmation by sample culture was reached in two of them. The remaining cases were negative.

Surgery was performed for all patients. Antibiotherapy was recorded in 5 patients receiving antibiotics (penicillins) for 11 days, 14 days, 12 months, and 18 months for each patient respectively. No reinterventions or deaths were recorded during follow-up. Risk factors for APA were no antecedents of cancer (OR 4.9, P = 0.034) and a previous wrong diagnosis of appendicitis (OR 7.6, P = 0.006), as shown in Table 2.

4. Discussion

AA is an infrequent invasive bacterial disease caused by bacteria from the genus *Actinomyces*, mostly frequently *Actinomyces israelii*. Isolated cases of abdominal actinomycosis are usually reported in the literature (6–12). However, as it is an uncommon abdominal infection, there are not any large series of patients with this diagnosis reported. We present the largest series of patients diagnosed with AA in the last 20 years.

AA has a very nonspecific and variable presentation, mimicking acute appendicitis, diverticulitis, malignant tumor, tubo-ovarian abscesses, and bowel obstruction most commonly (13). The appendix, cecum, and transverse colon are the most prevalent abdominal sites of actinomycosis, which can occur weeks to years after gastrointestinal mucosa disruption (14).

Fever, leukocytosis, and abdominal pain are usually observed in these patients, as in our series. Due to the nonspecific symptoms and the similarity with other frequent pathologies, AA is often forgotten in the first presumptive diagnosis (15).

Previous surgery, inflammatory and neoplastic processes, and presence of a longstanding IUD are reported risk factors that physicians have to be aware of when suspecting actinomycosis (16). The most prevalent morbidity that we found was neoplastic processes. Even if the number of patients was small, the previous incorrect diagnosis and the absence of carcinoma was associated with developing involvement of the appendix.

Radiologic findings have been reported to suggest the diagnosis, like large solid infiltrative masses with focal areas of attenuation invading the adjacent tissues revealed by CT scan with contrast enhancement. Particularly for appendiceal actinomycosis, wall thickening and periappendiceal inflammation has been described, and, although it can reveal acute appendicitis, actinomycosis should be included in the differential diagnosis (17,18). Therefore, CT must be considered an important diagnostic tool as far anatomical location, extent of the disease, and effectiveness of treatment is concerned.

Preoperative diagnosis is difficult to achieve because bacterial cultures and pathology are the key to diagnosis. Maybe through colonoscopy and histological examination of endoscopically acquired specimens or through CT-guided aspiration diagnosis can be achieved, but in most cases this is not enough (19). Prolonged bacterial cultures in anaerobic conditions are necessary for identification of the bacterium, but this still remains sterile in more than 50% of cases. In our series, the majority of cases were finally diagnosed histologically. The presence of yellow color formations 0.1–1 mm in size, called sulfur granules, is often pathognomonic (20).

Consequently, surgery is usually performed. In all of our patients the definitive diagnosis was made postoperative by histological examination in the majority of cases. That could be interpreted as a lack of effectiveness,

Table 1. Epidemiological data of patients diagnosed with abdominal actinomycosis.

	Abdominal actinomycosis (n = 13)
Age (mean)	48.6 ± 22.9 years
Sex (M/F)	8/5
Department	
General surgery General medicine Nephrology	10 (76.9%) 2 (15.4%) 1 (7.7%)
Admission in ICU	None
Risk factors	
Solid organ cancer Perforation hollow viscera Recent abdominal surgery HIV infection Dental manipulation Corticosteroids/solid transplant Previous antibiotherapy IUD	3 (23.1%) 1 (7.7%) 1 (7.7%) 1 (7.7%) 0 2 (15.4%) 3 (23.1%) 0
Symptoms	
Abdominal pain Mass Weight loss and asthenia Asymptomatic	9 (69.3%) 1 (7.7%) 1 (7.7%) 2 (15.4%)
Presurgical diagnosis	
Appendicitis Diverticulitis Malignant tumor Others (bowel obstruction, bacterascites)	5 (38.5%) 1 (7.7%) 4 (30.8%) 3 (23.1%)
Abdominal site	
Appendix Ascending colon /ileum Descending colon/sigma Subcutaneous fat/mesentery Peritoneal bacterascites Spleen	5 (38.5%) 1 (7.7%) 4 (30.8%) 1 (7.7%) 1 (7.7%) 1 (7.7%)
Diagnostic method	
Histological Culture	11 (84.6%) 2 (15.4%)
Blood analysis	
C-reactive protein Hemoglobin Hematocrit	7.4 ± 7.2 mg/dL 11.9 ± 2.3 g/dL 35.4 ± 6.2%

Table 2. Risk factor for appendiceal actinomycosis.

Variable	Prevalence (%)	OR	P
Men	3 (23.1%)	0.627	0.427
≥75 years old	2 (15.4%)	0.666	0.415
Hollow viscera perforation	5 (38.5%)	1.644	0.200
Solid transplant	6 (46.2%)	1.309	0.253
Previous surgical intervention	6 (46.2%)	1.309	0.253
HIV	6 (46.2%)	1.309	0.253
Cholecystectomy 15 days before	5 (38.6%)	1.644	0.462
Malignancy	6 (46.2%)	4.485	0.034
Corticosteroids intake	6 (46.2%)	1.309	0.253
X-ray diagnosis	4 (30.8%)	1.931	0.165
Histologic diagnosis	6 (46.2%)	1.309	0.253
Abdominal pain	5 (38.6%)	1.081	0.299
Previous incorrect diagnosis	2 (16.7%)	7.638	0.006
Spread	4 (30.8%)	1.931	0.165
Leukocytes 8000–12000 × 10^9/L	1 (7.7%)	1.081	0.299
Allergy	6 (46.2%)	2.787	0.095
Appendectomy	4 (33.3%)	7.638	0.006

but considering that surgery enables the debridement of necrotic tissue, the removal of persistent sinuses, and a definitive diagnosis, it can also be valuable as a therapeutic adjunct. Recent studies also have shown that surgery allows for shorter time of antibiotic therapy (21).

Penicillin G or amoxicillin are the preferred choices, with low resistance of *Actinomyces*. In patients who are allergic to penicillin, other options include tetracycline, erythromycin, doxycycline, and clindamycin. Usually high doses for a prolonged time are required, from 6 to 12 months, unless optimal surgery with resection of infected tissues has been performed (22).

No deaths were found in our series. It seems not to be an aggressive disease, but physicians have to be suspicious of this entity in order to provide a correct treatment and to avoid unnecessary surgical intervention. More studies are needed to characterize the patients affected by AA and to establish the correct treatment.

References

1. Wong VK, Turmezei TD, Weston VC. Actinomycosis. BMJ 2011; 343: d6099.
2. Ferrari TC, Couto CA, Murta-Oliveira C, Conceicao SA, Silva RG. Actinomycosis of the colon: a rare form of presentation. Scand J Gastroenterol 2000; 35: 108-109.
3. Milach J, Ziókowski P, Orze W. A case of actinomycosis of the sigmoid in a 41-year-old woman with a clinical appearance of cancer. Wiad Lek 1989; 42: 895-898.
4. Smego RA Jr, Foglia G. Actinomycosis. Clin Infect Dis 1998; 26: 1255-1261.
5. Cintron JR, Del Pino A, Duarte B, Wood D. Abdominal actinomycosis. Dis Colon Rectum 1996; 39: 105-108.
6. Acevedo F, Baudrand R, Letelier LM, Gaete P. Actinomycosis: a great pretender. Case reports of unusual presentations and a review of the literature. Int J Infect Dis 2008; 12: 358-362.
7. Karakus E, Mambet E, Azılı MN, Gülhan B, Tiryaki T, Tezer H. Actinomycosis of the appendix in childhood- an unusual cause of appendicitis. APSP J Case Rep 2014; 26: 1-5.
8. Simsek A, Perek A, Ethem Cakcak IE, Durgun AV. Pelvic actinomycosis presenting as a malignant pelvic mass: a case report. Journal of Medical Case Reports 2011; 5: 40.
9. Karagulle E, Turan H, Turk E, Kiyici H, Yildirim E, Moray G. Abdominal actinomycosis mimicking acute appendicitis. Can J Surg 2008; 51: 109-110.
10. Lee SY, Kwon HJ, Cho JH, Oh JY, Nam KJ, Lee JH, Yoon SK, Kang MJ, Jeong JS. Actinomycosis of the appendix mimicking appendiceal tumor: a case report. World J Gastroenterol 2010; 21: 395-397.
11. Filippou D, Psimitis I, Zizi D, Rizos S. A rare case of ascending colon actinomycosis mimicking cancer. BMC Gastroenterol 2005; 4: 1.

12. Valour F, Sénéchal A, Dupieux C, Karsenty J, Lustig S, Breton P, Gleizal A, Boussel L, Laurent F, Braun E et al. Actinomycosis: etiology, clinical features, diagnosis, treatment, and management. Infect Drug Resist 2014; 5: 183-197.

13. Choi MM, Baek JH, Lee JN, Park S, Lee WS. Clinical features of abdominopelvic actinomycosis: report of twenty cases and literature review. Yonsei Med J 2009; 50: 555-559.

14. Ha HK, Lee HJ, Kim H, Ro HJ, Park YH, Cha SJ, Shinn KS. Abdominal actinomycosis: CT findings in 10 patients. Am J Roentgenol 1993; 161: 791.

15. Harris LF, Kakani PR, Selah CE. Actinomycosis. Surgical aspects. Am Surg 1985; 51: 262-264.

16. Fowler RC, Simpkins KC. Abdominal actinomycosis: a report of three cases. Clin Radiol 1983; 34: 301-307.

17. Litt HI, Levine MS, Maki DD, Sachdeva RM, Einhorn E. Ileal actinomycosis in a patient with AIDS. Am J Roentgenol 1999; 172: 1297-1299.

18. Harris LA, DeCosse JJ, Dannenberg A. Abdominal actinomycosis: evaluation by computed tomography. Am J Gastroenterol 1989; 84: 198-200.

19. Kim JB, Han DS, Lee HL, Kim JP, Sohn JH, Hahm JS. Diagnosis and partial treatment of actinomycosis by colonoscopic biopsy. Gastrointest Endosc 2004; 60: 162-164.

20. Bennhoff DF. Actinomycosis: diagnosis and therapeutic considerations and a review of 32 cases. Laryngoscope 1984; 94: 1198-1217.

21. Koren R, Dekel Y, Ramadan E, Veltman V, Dreznik Z. Periappendiceal actinomycosis mimicking malignancy. Report of a case. Pathol Res Pract 2002; 198: 441-443.

22. Smith AJ, Hall V, Thakker B, Gemmell CG. Antimicrobial susceptibility testing of Actinomyces species with 12 antimicrobial agents. J Antimicrob Chemoth 2005; 56: 407-409.