Actinomycosis is a chronic suppurative infection caused by the gram-positive anaerobic filamentous bacterium, *Actinomyces Israelii* (*A. Israelii*). Three common regions of involvement are cervicofacial, abdominopelvic and thoracic in descending order of frequency. Isolated involvement of abdominal wall is an extremely rare clinical presentation of this disease. Only 7 well documented cases could be found in the English literature in an extensive Medline search. The disease presents either as an indolent indurated mass or more commonly as an acute or chronic suppurative infection. In the absence of any demonstrable predisposing traumatic, abdomino-pelvic or distant lesion, immunocompromised states may be considered as significant contributing factors in the etiopathogenesis of the disease. In contrast to other locations, the primary abdominal wall disease has been characterized by a female preponderance, more elevated mean age, predilection for abdominal lower left quadrant, and a shorter duration of symptoms. Preoperative diagnosis for indolent lesions is sometimes possible with computerized tomography (CT) scan and fine needle aspiration cytology (FNAC). For suppurative disease, the diagnosis is usually established by histopathological examination after surgical drainage. Prolonged course of antibiotic after confirmation of the diagnosis, by FNAC or surgical drainage, usually results in a cure. This report describes a case of isolated actinomycosis of the abdominal wall. Review of the literature is included to present the clinicopathological spectrum of actinomycosis and to describe the characteristic features, diagnostic tools and therapeutic consideration for isolated actinomycosis of the abdominal wall.

**Case Report.** A 32-year-old, Saudi diabetic male presented with 2 weeks history of a painful swelling in the left lower quadrant of the abdomen associated with low grade fever. The swelling initially appeared as a slightly painful nodule and increased gradually in size up to the presenting limits. The patient denied any history of trauma, previous abdominal surgery, accidental pricks, injection or any previous abscess at the site. He was an insulin dependent diabetic and had been on irregular insulin therapy for the last 12 years. He denied having any insulin injection at the site. He was a non-addict and an occasional smoker. There

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**ABSTRACT**

Primary actinomycosis of the abdominal wall is a rare clinical entity. Only 7 adequately described cases have been reported in the English literature. We report a case of isolated abdominal wall actinomycosis involving the left lower quadrant of the abdominal wall in a 32-year-old diabetic male. The diagnosis was confirmed by histopathological examination. Surgical drainage of the abscess followed by long-term administration of penicillin resulted in cure. The clinicopathological spectrum of actinomycosis is reviewed and isolated involvement of the abdominal wall is characterized in light of the knowledge acquired from the available literature on this rare clinical presentation. The significance of obtaining tissues for culture and histopathology in all inflammatory lesions is emphasized.

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From the Department of General Surgery (Khalid, Qazi, Al-Saleh), Department of Histopathology (Joarder), Riyadh Medical Complex, Riyadh, Kingdom of Saudi Arabia.

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Address correspondence and reprint request to: Dr. Kamran Khalid, PO Box 331267, Riyadh 11373, Kingdom of Saudi Arabia. Tel: +966 (1) 4069420. Fax: +966 (1) 4352206. E-mail: kkkhawaja@hotmail.com
was no history suggestive of bowel or urinary tract pathology. Past medical history was unremarkable. Physical examination showed a regular pulse of 90/minute, normal blood pressure and temperature of 37.8°C. An 8 x 11 cm, red, hot, tender, and relatively fixed swelling with smooth surface and ill-defined margins was present in relation to the left lower quadrant of the anterior abdominal wall. The swelling was partly fluctuant with peripheral induration and was non-pulsatile. No bruit could be heard. There was no associated lymphadenopathy, and the distal neurovascular status of the left lower limb was normal. The rest of the systemic examination including rectal examination was unremarkable. The laboratory investigations revealed hemoglobin of 11.6 gm/dL, total white cell count of 15,300 with 75% neutrophils and athrocyte sedimentation rate (ESR) of 20 mm/hour. His serum glucose was 23.7 mmol/L with normal electrolytes and renal functions. The urine examination showed glycosuria, no ketones or pus cells. The chest and plain abdominal x-rays were unremarkable. Abdominal ultrasound (Figure 1) demonstrated a large well-defined, heterogenous, hypoechoic lesion with multiple hyperechoic shadows and septations inside, in relation to the abdominal wall and pushing the urinary bladder medially but distinctly separate from it. Doppler ultrasound obtained at the same time did not reveal any vascular involvement. A CT scan of abdomen with triple contrast (Figure 2) confirmed the findings of ultrasound, with no associated intra-peritoneal extension or pathology. The clinical diagnosis of an abscess of the anterior abdominal wall was made. After the control of diabetes, the incision and drainage resulted in the evacuation of 60-70 ml of thick creamy pus involving the musculo-fascial plane of the external and internal oblique muscles with multiple loculations and granular calcific material in the deeper pockets. The pus and granules were obtained for culture and curettings of the wall of the abscess cavity were taken for histopathology. The wound was left open with daily dressings. The postoperative course was smooth. The gram stain and culture were negative for any bacterial growth including acid fast bacilli. The tissue histopathology revealed an actinomycotic abscess surrounded by acute inflammatory cells, foamy macrophages and granulation tissue (Figure 3). Following the histopathology result, the patient was put on parenteral crystalline penicillin and an active search for possible primary focus was initiated. The orodental examination was normal. The colonoscopy, a repeat CT of the chest, abdomen and pelvis with contrast, and an intravenous urogram did not reveal any bowel or other organ involvement. The wound was closed secondarily by the end of second week and patient was discharged on oral Phenoxy Methyl Penicillin for further 6 weeks along with an advice regarding good control of diabetes.
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(Figure 4). The 2-years follow up has shown a well healed wound and no features of local or distant recurrence.

Discussion. Actinomycosis is a chronic, progressive, suppurative infection characterized by formation of multiple abscesses, draining sinuses, abundant granulation tissue and dense fibrosis. The presence of sulfur granules in the lesions, sinus walls or discharges of involved tissues is characteristic. According to Idem, Bradshaw keeps the credit of first description of a patient with abdominal actinomycosis in 1846. The disease entity, however, remained unknown during his days. Branching fungus was the primary infective agent in development of hard masses in the jaw bones of cattle. In the following year, the term Actinomyces bovis was described to designate the causative organism, which was believed to be a fungus, due to typical appearance of the filaments radiating from a central tangled mass. Israel identified the same mycelia in the granules obtained from human autopsies specimens in 1878. He succeeded to culture the organism and described some of its bacteriological properties, including anaerobic growth. Spilsbury and Johnstone discussed the first diagnosis of actinomyces in a living man. Wright in 1905 substantiated the endogenous theory of infection initially put forward by previous studies. Berardi reviewed that this was Erickson who clearly differentiated the human and animal strains of the organism on morphological, biochemical and serological basis and the human pathogen was named as Actinomyces Israelii. Rudin in 1976 demonstrated that Actinomyces fulfills all the criteria of a bacterium and classified it in the order Actinomycetales, which also includes the genera Streptomyces and Mycobactrium. Some of these criteria include: lack of a nuclear membrane, presence of chitin from the cell wall, reproduction by fission, and inhibition of growth by penicillin and insensitivity to amphotericin B. Actinomycosis has a worldwide distribution, with equal frequency in the urban and rural population, but is seen predominantly in areas with poor standards for dental care. The disease is most commonly reported in young males, with no racial or climatic difference. The reported overall annual incidence varies between 1/119,000 in the Netherlands and 1/40,000 in Cologne. Twenty to forty cases are reported yearly by the Public Health Laboratory Service in the United Kingdom and Republic of Ireland. The characteristic actinomycosis is an indurated or hardened zone with multiple abscesses communicating with surrounding granulation tissue and fibrosis. Draining sinuses and fistulas eventually result with progression of the disease, and have been reported in approximately one-third of the abdominal actinomycotic lesions. The disease spreads by direct extension and hematogenous and lymphatic spread is uncommon and disseminated infection is extremely rare. Microscopic features of a typical actinomycotic lesion comprise an outer zone of granulation around central purulent loculations containing variable number of granules. Granules are generally round or oval, basophilic colonies with a radiating fringe of eosinophilic clubs. The clubs and matrix of the granules are gram-negative. All granules contain numerous gram positive bacilli exhibiting branching. Cervicofacial actinomycosis remains the most commonly reported clinical form, comprising 33-58% of all cases. The various etiological factors implicated with this form of the disease include dental extraction, carious teeth and maxillofacial trauma. Thoracic actinomycosis (15%) is reported as the third most common affliction in most of series and usually results from aspiration of the infected material in the oropharynx. In majority of the reports, abdominal actinomycosis is the second most commonly involved region, accounting for 20-25% of the disease presentation. In this form of the disease; the most commonly affected organs have been appendix and cecum (65%). Other reported sites in the abdomen include the colon, stomach, liver, gall bladder, pancreas, small bowel, anorectal region, pelvis, urinary tract, retroperitoneum and abdominal wall. The disease usually follows inflammation or perforation of a viscus, abdominal surgery or use of an intrauterine contraceptive device. Hematogenous dissemination has been described as possible explanation in rare locations such as kidneys and retroperitoneum. Berardi reported an overall decreasing incidence of actinomycosis in general and of the abdominal form in particular. Others have observed that the abdominal form has apparently become more frequent during the last 20 years. Whereas the development of abdominal actinomycosis after acute appendicitis has decreased as of early diagnosis, a lower incidence of perforated appendices, and improved antibiotic therapy, an increased incidence of pelvic actinomycosis has been observed over the past 2 decades. This is partly explained by a more frequent and long-term use of intrauterine device (IUCD) and inadequate gynecological follow up. Actinomyces Israelii can be isolated in 10% of asymptomatic IUCD users and 25% of IUCD users with symptoms during routine vaginal examination, and the organism may be found in the genital track in 3-4% of women, whether with or without an IUCD. Isolated involvement of the abdominal wall is an extremely rare presentation of actinomycosis. The diagnosis can be established when no other associated intra-abdominal organ involvement can be documented by all the possible diagnostic aids. The infection exclusively involves the musculo-aponeurotic layers of the abdominal wall.
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In a recent review, Garcia and associates described a total of 20 patients of primary actinomycosis of the abdominal wall in the world literature. Only 7 adequately described cases could be searched reported in the English literature. In one other brief report on CT findings of actinomycosis in 6 patients, one primary abdominal wall abscess has been described. Most of the cases have been reported in the French, Spanish, German or Russian literature. In contrast to other common locations, a female preponderance and elevated mean age was noted for this form of the disease. However, the patient described in this report was a young male. Left lower quadrant of the abdomen has been reported as the most frequently involved site. Pathogenesis of the isolated abdominal wall actinomycosis remains to be elucidated. In this unusual form of the disease, reduced host immunity is put forward as the possible contributing factor for individual susceptibility. Diabetes mellitus, use of steroids, autoimmune diseases, neoplasms, immunosuppressive medications and human immuno-deficiency infection have all been reported to predispose to actinomycosis at unusual locations. Likewise, the patient described in this case report was an insulin dependent diabetic. Most of the time the disease takes a subacute course, but the duration of symptomatology is usually shorter as compared to the other more frequent forms of the disease. This could partly be explained by the more obvious location of the disease and partly by the more frequent presentation as suppurative rather than indolent indurated lesions. Palpable mass or visible sinus tracts are the most characteristic physical signs of abdominal wall actinomycosis. Low grade fever, leukocytosis and an elevated ESR, though not specific but are supportive of an inflammatory lesion. The high index of clinical diagnosis of actinomycosis can be kept in a patient with the above mentioned clinical features, along with a previous history of gastrointestinal disease, diabetes, abdominal surgery or prolonged use of IUCD. Definitive preoperative diagnosis is usually difficult since the clinical and laboratory findings of the disease are nonspecific and there is no confirmatory serologic test. A preoperative diagnosis is reported in only less than 10% of the cases in the world literature. The identification of sulfur granules in the discharge is considered as the most conclusive diagnostic finding, but they have been reported in only half of the cases and even in most of these cases in permanent sections only. Microscopic demonstration of sulfur granules or A. Israeliii from biopsy or smear materials obtained from the sinus tracts is necessary for the confirmation of the diagnosis. Definitive diagnosis is achieved by culture, but even cultures are reported positive in less than 50% cases and false negative results have been frequently reported. In addition to the upper and lower gastrointestinal endoscopy, imaging studies including plain radiology, ultrasonography, contrast studies and CT scan have been employed to exclude other organ involvement and to establish the diagnosis and extent of involvement. A CT has been advocated as imaging investigation of first choice and percutaneous needle aspiration has been recommended for definitive diagnosis. On CT, the usual lesion is a solid heterogeneous mass with focal areas of attenuation or a cystic lesion with a thickened wall and internal septations which demonstrate contrast enhancement. The mass usually described as "infiltrative" and may be associated with marked desmoplastic reaction. In addition, ultrasonographic or CT guided fine needle aspiration cytology followed by percutaneous drainage of the abscess have been reported with variable success rates. Surgical or percutaneous drainage of the abscess, debridement of necrotic tissues or excision of inflammatory mass have been various reported surgical options depending upon the nature and extent of the lesion. Obtaining the discharge for bacteriological and microscopic examination, pus and tissue cultures and histopathology are invaluable in establishing the definitive diagnosis. Medical treatment is an essential adjunct to eradicate the disease. However, on account of excessive fibrosis and relative low vascularity of the lesion, long-term therapy is required. Currently, high-dose penicillin is the recommended drug to penetrate the fibrotic wall and reach the colonies of the organism in the core of the sulfur granules. Penicillin is put forward as the possible contributing factor for individual susceptibility. Immunocompromised states are reported as possible predisposing factors. Short duration of symptoms, elevated mean age, female preponderance, prevalence for lower left quadrant of the abdomen are some of the distinct characteristic features for this form of the disease. A CT scan is the imaging investigation of choice. Clinical and radiological correlation is important to achieve the preoperative diagnosis. Percutaneous biopsy has been recommended for definitive diagnosis in clinically suspected lesions. Percutaneous drainage, followed by long term antibiotics, is considered as first line treatment where preoperative diagnosis is established. Surgical drainage and debridement are recommended for undiagnosed lesions and for those that fail to respond to more conservative methods.
The importance of obtaining tissues for culture and histopathology in all inflammatory lesions can not be overemphasized.

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