The anaerobic Gram-positive bacteria *Actinomyces israeli* normally inhabit the oral cavity (especially the tonsils and teeth), gastrointestinal tract (usually the appendix and colon) and genital tract in humans, and rarely cause disease [1]. Actinomycosis requires the presence of many other bacteria, which destroy the over-vascularized regions and convert an aerobic microenvironment to an anaerobic one [2]. Actinomycosis occurs most frequently in the cervicofacial (50%), abdominal (20%) and thoracic (15%) regions [3,4].

Most cases result from perforated appendicitis, after an uncomplicated appendectomy, bowel perforations secondary to diverticulitis, foreign bodies and trauma [5,6]. Other, more sporadic reports describe pelvic disease in association with intrauterine devices [7,8]. The infection seems to occur after the mucosal surface is breached by disease and actinomyces may cause multiple abscess formation, draining sinuses, abundant granulation, dense fibrous tissue or mass lesions after the mucosal barrier is broken. Since the late 1970s, gynecologic sources have been the prevalent origin of abdominopelvic actinomycosis [9–14]. Here, we report a recently diagnosed case of multiple intra- and extra-abdominal actinomycosis mimicking urachal tumor that was diagnosed by computed tomography, which presented as two tender abdominal palpable masses following long-term intrauterine device use. The disease was confirmed by postoperative histopathologic examination, which revealed sulfur granules microscopically, and was successfully treated by complete surgical resection following a period of appropriate antibiotic treatment.
CASE PRESENTATION

A 48-year-old female patient complained of two palpable abdominal masses over the periumbilical area noted about 1 month previously. She had initially gone to the gynecologic outpatient department for help, and two abdominal soft and painful masses were palpable during physical examination.

Abdominal computed tomography (CT) revealed an abdominal wall tumor with heterogeneous enhancement that was suspected of being urachal carcinoma with invasion of the anterior and left abdominal walls (Figures 1A and 1B). Another nodule over the superior bladder wall, suspected of being a site of metastasis, was also noted (Figure 1C). Uterine and cervical soft mass lesions suspected of being leiomyoma, an intrauterine device, left side fallopian tube wall thickening and prominent ovary were also noted during the study of this image (Figure 1D).

She was referred to the urology ward for surgical intervention. Laboratory assessment and body temperature were all within normal limits prior to the operation, except for high fasting blood sugar. She denied any underlying disease except diabetes mellitus, which was controlled with medication. Exploratory laparotomy was performed and two nodules within the tissue of the rectus abdominal muscle with direct invasion of the peritoneum, omentum and ileum were noted. Two soft nodules were also noted over the superior wall of the urinary bladder and serosa of the rectum. The masses were separated from the abdominal wall, and those involved with the omentum were excised and sent for frozen section examination. The examination revealed no malignant cells. Finally, the nodules over the superior bladder wall and serosa of the rectum were completely excised. Resection of the involved omentum and segmental resection of the ileum, plus an end-to-end anastomosis were also performed. Finally, abdominal total hysterectomy and bilateral salpingo-oophorectomy were carried out due to possible uterine leiomyoma.

Grossly, the nodules were grayish and elastic with a smooth surface. Microscopically, the nodules of the abdominal wall, the ileum, the serosa of the rectum

Figure 1. (A, B) Two heterogeneously enhanced abdominal wall tumors located over the anterior and left abdominal walls, with invasion of the peritoneum and omentum. (C) One nodule over the superior bladder wall was suspected to represent a site of metastasis. (D) Uterine and cervical soft mass lesions suspected of being leiomyomata, with fluid collection within the uterine cavity and an intrauterine device, were noted. Left-side fallopian tube wall thickening and a prominent ovary and right-side cystic-like ovary were also noted.
and the bladder wall showed dense inflammatory infiltration with pus accumulation and fibrosis. The left fallopian tube also showed the same characteristics microscopically. There were some sulfur granules surrounded by inflammatory cells, suggesting actinomycosis (Figure 2).

After actinomycosis was confirmed, an intravenous penicillin injection was given, followed by oral penicillin administration after she was discharged. At the 3-month postoperative follow-up, the patient was completely asymptomatic with normal laboratory assessment, and was free of abdominal lesions.

DISCUSSION

Actinomycosis is an infectious disease caused by the anaerobic Gram-positive filamentous bacteria, *Actinomyces israeli*. Actinomycosis has a worldwide distribution, with equal frequency in urban and rural populations, but is seen predominantly in areas with poor standards of dental care [15]. The organisms produce a characteristic granulomatous inflammatory response, with pus production and abscess formation followed by necrosis and extensive reactive fibrosis. Most cases occur in adolescents and middle-aged individuals. The most common forms of actinomycosis are cervicofacial, abdominal and thoracic [16,17]. Abdominal actinomycosis is the second most commonly affected region, accounting for 20–25% of disease presentation [18,19]. In this form of the disease, the most common affected organs are the appendix and cecum (65%) [20,21]. Other reported sites in the abdomen include the colon, stomach, liver, gallbladder, pancreas, small bowel, anorectal region, pelvis, urinary tract, retroperitoneum and abdominal wall [22,23].

Bradshaw first described a patient with abdominal actinomycosis in 1846, as reported by Romeo Berardi.
Abdominal actinomycosis is a rare clinical entity and its various features usually make the diagnosis difficult. This organism was initially thought to be a fungus, but is actually a filamentous Gram-positive bacillus. The lack of a nuclear membrane, absence of chitin from cell walls, reproduction by fission, and most importantly, inhibition of growth by penicillin and insensitivity to amphotericin B, classifies them as bacteria and not fungi.

Clinically, there is no evidence of specific symptoms related to actinomycosis in the abdominal wall. Abdominal actinomycosis usually presents with painful palpable mass, body weight loss, anorexia, and constipation associated with fever and chills. In laboratory analysis, the dominating signs are anemia, leukocytosis and positive inflammatory markers. Immunocompromised status, such as diabetes mellitus, steroid therapy and neoplasma, is a significant predisposing factor for actinomycosis.

Accurate diagnosis is reached preoperatively in fewer than 10% of cases. Abdominal actinomycosis can be confused with malignancies, intestinal tuberculosis, chronic appendicitis, ameboma, diverticular disease, Crohn’s disease and pathologic states within the rectus abdominis muscle and its sheath. Most cases are detected during surgical exploration, after drainage of an abscess or during postmortem examination. Definitive diagnosis is achieved by culture, but even cultures are reported positive in less than 50% of cases, and false-negative results have been frequently reported. Imaging studies have not been very helpful in preoperative diagnosis. CT seems to be the most helpful modality, demonstrating a solid mass with focal areas of attenuation or a cystic mass with a thickened wall that enhances with infusion. Low signal intensity on magnetic resonance imaging, performed on T2-weighted sequences, may sustain the suspicion of actinomycosis. Because preoperative diagnosis is difficult, it is usually made during the operation, and confirmed later by pathologic examination. The identification of sulfur granules obtained from the pus in the lesion is the most reliable way of confirming the disease, but these have been reported in only half of all cases.

Medical treatment with sensitive antibiotics is extremely effective for this disease. The recommended antibiotics are crystalline penicillin, 10–20 million U/day for 4 weeks, followed by phenoxybenicillin, 2–4 g/day, for a year. Antibiotic pretreatment may facilitate and influence the outcome of the operation. Surgery is considered when malignancy cannot be excluded, or if removal of persistent sinuses, drainage of abscess and excision of necrotic tissue is necessary. Recently, combined surgical and antibiotic treatment has been outlined as the most efficient treatment modality; this treatment gives good results in more than 90% of cases.

In the past two decades, the popular use of intrauterine devices (IUDs) and inadequate gynecologic follow-up seems to have resulted in an increased incidence of pelvic actinomycosis. Actinomyces israeli can be isolated in 10% of asymptomatic IUD users and 25% of IUD users with symptoms during routine vaginal examination. Long-term use of IUDs could promote the overgrowth of actinomyces in the vagina. It is believed that prolonged use of an IUD increases the concentration of Actinomyces israeli in the endometrial cavity and its ultimate migration to the fallopian tubes and ovaries.

The incidence of pelvic actinomycosis in Taiwan has been reported to be 0.08% in admitted gynecologic patients. Multiple organ involvement of actinomycosis, such as our case, is infrequently seen. The rare case of multiple organ actinomycosis was suspected to be IUD-related, and was successfully treated with complete surgical resection and high-dose long-term penicillin therapy.

REFERENCES
Actinomycosis mimicking urachal tumor

子宮內避孕器的婦女身上被誤以為腎尿管腫瘤的腹部內及腹部外放射線菌瘤
— 病例報告

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放射線菌瘤是一種由格蘭氏陽性絲狀厭氧菌，放射線菌 israelii 所造成的慢性化膿性感染。因為其不典型的特徵，這種病通常在手術後才被診斷出來。腹部的放射線菌瘤是此疾病第二常發生的地方而且常會與腹部腫瘤、發炎性腸疾和憩室炎相混淆。腹部及骨盆的型式通常因為過久的使用子宮內避孕器所造成。本文報告一位罕見的合併腹部及腹腔外的放射線菌瘤患者一開始經電腦斷層診斷誤以為腎尿管腫瘤的長期使用子宮內避孕器的婦女，主訴為腹部出現兩疼痛性腫塊。此病最終由手術完全切除，併長期適當的抗生素治療而治癒。且由術後病理學檢查發現硫顆粒而確定診斷。

關鍵詞：放射線菌感染症，子宮內避孕器，腎尿管腫瘤
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