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Review Article

ANAL FISTULA SECONDARY TO ACTINOMYCOSIS, AN EXTREMELY RARE COMPLICATION

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ABSTRACT

Actinomycosis is the most misdiagnosed disease so far, though it is known to clinicians since 150 years. Primary perianal actinomycosis is extremly rare. The infection is caused by the bacterium Actinomyces, gram positive bacilli of the Actinomycetales genus, and A. israelii is responsible for the majority of human disease, which often is a saprophyte. Male gender and diabetes are risk factors. Patients, who have undergone appendicectomy, have had a missed perforated appendicitis or women with a history of intrauterine contraceptive device use are at an increased risk. The diagnosis is a challenge and often delayed, with a protracted history of masses and sinuses extending into the gluteal, urogenital and anorectal region. The diagnosis is suspected by the observation of draining sulfur granules and promptly confirmed by histology .These findings are contrasting with the classic picture of perianal actinomycosis. It is concluded that perianal actinomycosis can occur in the absence of risk factors and that early diagnosis requires a high degree of suspicion. An infection with Actinomyces should be suspected in the presence of lesions containing watery purulent material with sulfur granules. It gives unusual tumoral lesions with abcesses & fistulas. Cope in 1949 said: - "Actinomycosis occurs so seldom in the colon and the rectum that no surgeon, even if he be a proctologist is likely in a life time to see more than a few cases". An anorectal actinomycosis case can mimic rectal cancer .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 acute inflammatory pathologies or malignancy. The treatment, a combination of surgery and antibiotics [penicillin], is poorly standardized. Recurrences are possible. So the condition remains a challenge over the century, but it can be overcome by an integrated approach.

INTRODUCTION

Abdominal actinomycosis with anorectal manifestation is a rare condition encountered in surgical practise which needs high degree of suspicion along with biopsy for diagnosis. It is a infectious condition caused by Actinomyces species, gram positive bacteria, non-sporing absolute or facultative anaerobes. The predominant form in human disease is A. israelii. Actinomyces species secrete the polysaccharide that bind the bacilli's branched filaments forming characteristic 'sulphur granules'. These are typically yellow. They are commonly considered diagnostic of actinomyceal infection, but are present in only 50% of cases. Abdominal actinomycosis is approximately three times commoner in men than women. The condition is commonly seen in middle aged rural people.

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A israelii and other actinomyces species are endogenous inhabitants of most human mucous membranes, with the mouth, bronchi and gastrointestinal tract.They inhibit areas of stasis and the tonsillar crypts in the mouth and appendix, caecum and sigmoid are the predominant gastrointestinal sites. Preceding mucosal injury is required before actinomycosis supervenes. Perforated appendicitis is the commonest cause with neoplasm or trauma, such as fishbone perforation of the gastrointestinal tract. Occasionally there is no preceding mucosal injury. An initial stage of localised abscess formation is followed by extension into spreading peritoneal disease. Finally the disease progresses into a stage of fistulisation, both internally and cutaneously. The abscesses generally consist of a thick layer of granulation surrounding a central pool of pus containing the typical sulphur granules with liquefaction. Actinomycotic infection is typified by a progressive inflammatory response which is contiguous and insidious with multiple connecting abscesses.

Lymphatic or haematogenous spread is rare. Polymicrobial superinfection is common with synergetic organism. Human actinomyceal infection commonly affects three areas. Most (>50%) are cervicofacial, Thoracic actinomycosis represents 15–20% of cases .Abdominal actinomycosis accounts for approximately 20% of cases. The remainder are sporadic cases involving organs as diffuse as the brain, salivary glands and vertebra or occasionally systemic disease. As per as Anorectal clinical manifestations are concerned, they are usually secondary in the form mass/growth, abscesses, sinuses or fistulae with some atypical abdominal symptoms. These are recurrent in nature.

Apart from microbiological examination, no test is diagnostic. Malignancy is often the main differential diagnosis and investigation is usually directed at excluding it. Plain abdominal radiology is not so useful. Ultrasound scanning can identify an abdominal mass and if consistent with an abscess then percutaneous drainage. Barium enema examination may show a tapered luminal narrowing, mucosal fold thickening or mucosal nodularity. CT scan is the single most useful imaging modality. It reveals both the extent of inflammation and degree of organ involvement. MRI of the pelvis and anorectum to delineate fistula-in-ano in perianal actinomycosis. In short no radiological, haematological or endoscopic investigation demonstrates any specific features to allow it to be diagnostic for abdominal actinomycosis although the infiltrative nature of the fibrotic process across different organs

The recommended drug of choice is penicillin, 2.5–5 Megaunits, administered IV four times daily. In penicillin-allergic individuals erythromycin & tetracycline are suitable alternatives. If cases that remain symptomatic after a month of penicillin therapy, additional antibiotics such as aminoglycosids and linozalid should be used to prevent polymicrobial superinfection. Therapy may be need for up to a year. After all there are more chances of recurrences. Surgical intervention is necessary in case perianal abscesses, sinuses and fistulae, but it also doesn't give complete cure. Resection of the infected, necrotic tissue is generally recommended reducing the bacterial burden and lessening the chances of recurrence.

CASE STUDY

A 27 years old male individual from rural area farmer by occupation attended OPD, presented with perianal oedema having a sinus at 5 o'clock 7 cm away from anal opening, pus discharge, perianal and left lower abdominal pain, anorexia with low grade fever. Patient was relatively all right before one year and eight months, and then he gradually developed above symptoms, so he went to local family physician that treated him only symptomatically. But he got temporary relief. After one and half month, he had the same problem. Again he was seen by the same local physician and referred him to general surgeon who admitted him for further surgical management. Under general anaesthesia, incision and drainage was done followed by wound care for a week. The patient was discharged with satisfactory relief .He remained asymptomatic for ten months and again developed the same symptoms & sign with significant loss of weight. He again consulted the same surgeon who investigated completely mainly to rule out tubercular origin.

The patient again underwent I & D with good antibiotic cover and wound care. After ten days, he was discharged with complete relief. Unfortunately, the patient presented with the same clinical picture after 3 months. With lot of physical & mental disturbances, the patient was consulted us with previous reports. The patient was admitted in male surgical ward for further management. There was no significant past/drug/allergy history. Diet was regular, mostly vegetarian, but sometimes non vegetarian. Sleep was adequate, no addiction noted. Family history showed father was hypertensive. General examination- general condition moderate, mild febrile, cooperative, intelligent, conscious, oriented, pale, cachexic, anxious, gait normal, eyes sunken, muddy conjunctiva, tongue dry & coated, tachycardia, blood pressure normal, no skin lesion, no cyanosis, no jaundice, no clubbing, bilateral superficial inguinal lymphadenopathy. Systemic examinations- CVS, RS, CNS normal. Per abdomen, liver slightly palpable, there was mild tenderness in left iliac region.

Local examination revealed moderate oedema, redness, tenderness and raised temperature over left posterior aspect of anus with a discharging sinus at 5 o'clock position 7 cm away from anal opening. Overlying skin shiny. Sphincter spasm noted. Surprisingly discharge didn't contain sulphur granules. Routine investigations normal except low Hb%, leucocytosis, monocytosis, raised ESR. Chest X-ray normal, USG abdominal pelvis commented colitis picture, Fistulography & Endo rectal MRI showed transphincteric tract going high above levitor ani. So it was high anal pelvi rectal secondary fistula. Symptomatic appropriate treatment was started and the patient was posted for Ksharsutra therapy. It is a unique, time proven scientific treatment for anorectal disorders especially anal fistula. Ksharsutra is a medicated thread which causes chemical cauterisation of the fistular tract by proteolysis enzymes. It cuts and simultaneously heals the tract. Average cutting rate of track is 1 cm/week.

Under spinal anaesthesia, probing and threading done according to Goodsaal's rule as per fistulography. To confirm the diagnosis, a piece from the tract along with collected secretions [biopsy]was taken and sent for histopathological study. Broad spectrum higher IV antibiotics, analgesics, anti inflammatory drugs, multi vitamins & mineral given for two week. Wound care was taken properly. After 5 days, we got histopath report which showed surprisingly "ACTINOMYCOSIS OF RECTUM". On 10 th day, the patient was discharged with encouraging relief.

The same management was continued, Ksharsutra was changed weekly by rail road methods. Certain Ayurved drugs were prescribed- 1. Gandharva Haritaki powder 3 tsf HS with warm water [for laxative purpose], Triphala Guggulu, Saptang vishanti Guggul, [for proper healing of the tract],Arogya vardhini ,Aam pachak vati [for digestion & detoxification],Gandhak rasayan [antibiotic] each 1 tab BD after each meals, Abhayarishta 10 ml BD with equal amount of water after meals[for gastrointestinal kinesis]. Yashtimadhu ghrit[wound cleaner & healer] for local application. By six and half month, all symptoms & signs gradually disappeared and fistular tract cut & healed completely. The patient had follow up fortnightly till two years. Yet we don't find any complication, sequel, recurrence.

DISCUSSION AND CONCLUSION

Anorectal actinomycosis is an uncommon condition usually secondary to mostly abdominal actinomycosis.Perianal abscesses, sinuses and fistulae are the most common presentations. The possibility of actinomyceal infection should be borne in mind when dealing with atypical abdominal presentations, particularly if there is a previous history of appendix perforation or IUCD usage. Pre-operative diagnosis is often difficult and radiological imaging is unlikely to allow a definitive diagnosis. Surgical interference is frequently required with long-term antibiotics. In such cases, integrated approach is certainly fruitful.

Ksharsutra along with Ayurved therapy have been found to be very beneficial in primary as well as secondary fistulae.

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