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Tuberculous abdominal cocoon: original article

Tüberküloz abdominal koza: Orijinal makale

Imtiaz WANI,[1] Mohamad OMMID,[2] Arfat WAHEED,[2] Mehraj ASIF[3]

BACKGROUND
Tuberculous abdominal cocoon is a rare disease, and diagnosis is seldom made preoperatively. The bowel is encased in a membrane in a cocoon-like fashion. Histopathology is confirmatory.

METHODS
This prospective case note review was a study of patients diagnosed with tuberculous abdominal cocoon from April 2005 - April 2008. There were 8 females and 3 males.

RESULTS
All patients had features of small bowel obstruction. All had laparotomy and the characteristic finding of absence of the greater omentum from the involved area and the absence of any stigmata of gut tuberculosis. Peeling of membrane is all that is required, and patients received anti-tubercular therapy postoperatively. In each case, evidence of tuberculosis on histopathology of membrane was present.

CONCLUSION
Tuberculous abdominal cocoon is a rare entity. Females are commonly affected. Surgery is the preferred treatment.

Key Words: Antitubercular therapy; greater omentum; tuberculous abdominal cocoon.

AMAÇ
Tüberküloz abdominal koza, ameliyat öncesi dönemde nadir olarak tanı konulan ender bir hastalıktır. Bağırsak, bir membran içinde, koza gibi bir şekilde kılıfla kaplanır. Histopatoloji doğrulayıcıdır.

GEREÇ VE YÖNTEM
Bu prospektif olgu incelemesinde, Nisan 2005 ile Nisan 2008 tarihine kadar olan dönemde tüberküloz abdominal koza tanısı alan hastalar (8 kadın, 3 erkek) değerlendirildi.

BULGULAR
Tüm hastalarda ince bağırsak tıkanıklığı özellikleri vardı. Bütün hastalara laparotomi uygulandı; etkilenen bölgede büyük omentumun bulunmaması ile bağırsak tüberkülozu belirtisi bulunmaması karakteristik bulgu idi. Gerekli olan işlem membranın soyulmasıydı ve hastaların hepsi ameliyat sonrası antitüberküloz tedavisi aldı. Her olguda, membran histopatolojisinde tüberküloz kanıtı bulundu.

SONUÇ
Tüberküloz abdominal koza nadir görülür. Kadınlar yaygın olarak etkilenir. Cerrahi tercih edilen tedavidir.

Anahtar Sözcükler: Antitüberküloz tedavisi; büyük omentum; tüberküloz abdominal koza.

Abdominal cocoon (AC) is a rare disease in which intestinal obstruction results from the encasement of variable lengths of the bowel by a dense fibrocollagenous membrane that gives the appearance of a cocoon. Tuberculosis as a cause of AC is rarely seen. This is commonly seen in the tropics and subtropical areas. Females show a predilection for tuberculous AC. A presentation of small bowel obstruction (SBO) seen in most of the cases often diverts attention from this rare disease. Diagnosis is seldom made preoperatively. Surgical exploration with release of gut from the encasement membrane is the treatment of choice.

The aim of this study was to investigate the clinical features, intraoperative findings as well as response to antitubercular treatment (ATT) in all patients who were diagnosed with tuberculous AC.

MATERIALS AND METHODS

This study was conducted in S.M.H.S. Hospital, Srinagar, from April 2005 to April 2008 for a period of three years. All patients diagnosed with AC by preoperative imaging or by intraoperative findings and the respective histopathological findings were included in the study. Patients with history of previous surgery or

[1]Department of Surgery, SMHS Hospital; [2]Department of Anaesthesia, SKIMS; [3]Department of Surgery, SKIMS, Srinagar, Kashmir, India.

[1]SMHS Hastanesi, Cerrahi Bölümü; [2]SKIMS, Anesteziyoloji Bölümü; [3]SKIMS, Cerrahi Bölümü, Srinagar, Keşmir, Hindistan.

Correspondence *(İletişim):* Imtiaz Wani, M.D. Amira Kadal, Srinagar 190009 Srinagar, India. e-mail *(e-posta):* imtazwani@gmail.com

with previous adhesion/band obstruction were excluded from the study. All patients who were diagnosed with tubercular AC who underwent surgery were given ATT postoperatively in the follow-up.

RESULTS

The total number of patients with AC was 11 (8 females, 3 males). The median age was 18.3 years (range: 16 months - 58 years). Only one patient was from an urban area; the remainder of the patients were from rural areas. Preoperative diagnosis was made in only one patient on computed tomography (CT) scan; the rest were diagnosed intraoperatively.

All had features of recurrent attacks of SBO. A characteristic intraoperative feature seen in all our cases was absence of the greater omentum in the involved area of the gut. Gut loops were encased in a cocoon membrane. No other stigmata of any other gut tuberculosis were present. Two strictures in the distal ileum were seen in one case, with adhered omentum. A transparent membrane originating from the mesentry encompassing a single loop initially or multiple loops was seen. This flimsy membrane stacks together and adjacent membranes fuse to form an encasement membrane for the gut, which progressively becomes fibrotic. Only one case had membrane encircling the gut from the duodenojejunal flexure to the transverse colon; the rest had only jejunum and ileum involvement. In our series, the gut was simply piled upon itself in the sac with no adhesions. In one case (12/F), the dense and tough nature of the membrane precluded peeling. All patients with tubercular etiology were managed by ATT postoperatively for six months. None of our patients had any evidence of pulmonary tuberculosis.

Histopathology of the peeled membranes confirmed tuberculosis in all cases showing caseating granulomas. After a median follow-up period of 13.5 months, no recurrence of the condition was seen in any patient.

Diagnosis of tuberculosis in each case was based on clinical history, family history of tuberculosis, sputum analysis, ELISA (enzyme linked immunosorbent assay), polymerase chain reaction (PCR) of body fluids, and the documentation of histopathological evidence of caseating granulomas of the peeled membrane. No patient in our series had recurrence; only one had an episode of subacute intestinal obstruction for which she was managed conservatively.

DISCUSSION

Abdominal cocoon (AC) is a rare cause of intestinal obstruction. The condition has been classified as primary or secondary based on whether it is idiopathic or has a cause. Many causes are documented in the literature for the secondary type, of which chronic ambulatory dialysis, abdominal tuberculosis, prolonged beta-blocker practolol intake, ventriculoperitoneal shunts, and peritoneovenous shunts are the commonest.[1]

Tuberculous AC involves dense fibrous encasement of abdominal viscera, and predominantly affects the small bowel with a tendency to involve the terminal ileum and rarely leading to bowel perforation.[2] Occasionally, the large bowel, stomach, or other abdominal organs may be involved; encapsulation may be total or partial. It has been hypothesized that increased deposition of fibrin on the peritoneum, after the release of fibrogenic cytokines, converts fibrinous adhesions to generalized peritoneal fibrosis. Patients with tuberculous AC usually complain of recurrent attacks of SBO or abdominal lump, and some patients may be asymptomatic.[3] An accurate diagnosis is difficult to make preoperatively. The clinical features, ultrasonography barium X-ray and contrast computed axial tomography scan can increase preoperative diagnosis, with the intraoperative and histopathology findings confirming the final diagnosis.[4]

On conventional abdominal radiographs, there may be evidence of SBO, with dilated bowel loops and air-fluid levels. The characteristic features of an AC depicts encasement of most of the small-bowel loops in a thick fibrous membrane and arrangement of the loops in a concertina shape with a narrow posterior base, having the overall appearance of a cauliflower on ultrasonography. Concertina pattern or a cauliflower sign on barium small-bowel series provides a clue to the diagnosis of AC. CT scan shows conglomerated intestinal loops with mild dilatation at the center of the abdomen, which are encased with a membrane represented by a thickened peritoneum.[5] Diagnostic laparoscopy, in this era, has a major role in the management of this rarer cause of intestinal obstruction. An accurate diagnosis preoperatively is difficult. Suspicion of the diagnosis of tuberculous AC preoperatively based on abdominal CT findings allows for surgical intervention that helps in the early definitive diagnosis and improves the prognosis.[6]

Surgery is the preferred treatment. Peeling of the membrane is all that is required. The typical finding at surgery is a conglomeration of small bowel loops encased in a dense white membrane. Rarely, associated inter-loop adhesions can be seen. It should be stressed that interloop adhesions in the gut may sometimes simulate cocoon, but this is a different entity from AC. In true AC, interloop adhesions are scarcely seen, as in our series, in which the gut was simply piled upon itself in the sac with no adhesions. In one case (12/F), the dense and tough nature of the membrane precluded peeling. All patients with tubercular etiology were managed by ATT postoperatively for six months.

A characteristic feature seen in all our cases was the absence of the greater omentum in the involved area of the gut. Gut loops were stacked in the membrane. In all our cases of tubercular cocoon, no other stigmata of tuberculosis including enlarged nodes with caseation were seen.

Regarding membrane formation, a transparent membrane encompassing a single loop initially or multiple loops was seen at first, and the membrane was seen to originate from the mesentery, forming a shield-like membrane encasing gut loops. The membrane extended up to the lateral abdominal wall. Only one case had a membrane encircling the gut from the duodenojejunal flexure to the transverse colon; the rest had only jejunum and ileum involvement. This flimsy membrane originating from the mesentery involves a single stacked loop and adjacent membranes fuse to form the encasement membrane, which progressively becomes fibrotic.

Histopathological examination of the encapsulating membrane persistently shows thickened vascular fibrocollagenous tissue, with chronic inflammatory reaction evidenced by lymphocytic and plasma cell infiltrates. Mesenteric lymph nodes may demonstrate nonspecific reactive hyperplasia and may reveal the etiology of tuberculosis. In cases in which peeling is not possible, ATT is an option.

In conclusion, tuberculous AC is an unusual cause of SBO that is rarely diagnosed preoperatively. Surgery involves peeling away the fibrous material encasing the bowel. The procedure is usually uncomplicated and possible without perforating the underlying gut. Patients should be prescribed ATT, once the diagnosis has been confirmed histologically, and the condition is unlikely to recur.

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