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BRIEF ARTICLE

Left-sided appendicitis: Review of 95 published cases and a case report

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Abstract

AIM: To give an overview of the literature on left-sided acute appendicitis (LSAA) associated with situs inversus totalis (SIT) and midgut malrotation (MM).

METHODS: We present a new case of LSAA with SIT and a literature review of studies published in the English language on LSAA, accessed *via* PubMed and Google Scholar databases.

RESULTS: Ninety-five published cases of LSAA were evaluated and a 25-year-old female, who presented to our clinic with left lower abdominal pain caused by LSAA, is reported. In the reviewed literature, fifty-seven patients were male and 38 were female with an age range of 8 to 82 years and a median age of 29.1 ± 15.9 years. Sixty-six patients had SIT, 23 had MM, three had cecal malrotation, and two had a previously unnoted congenital abnormality. Fifty-nine patients had presented to the hospital with left lower, 14 with right lower and seven with bilateral lower quadrant pain, and seven subjects complained of left upper quadrant pain. The diagnosis was established preoperatively in 49 patients, intraoperatively in 19, and during the postoperative period in five; 14 patients were aware of having this anomaly. The data of eight patients were not unavailable. Eleven patients underwent laparoscopic appendectomy, which was combined with cholecystectomy in two cases. Histopathological examination of the appendix specimens revealed adenocarcinoma in only two of 95 patients.

CONCLUSION: The diagnosis of left lower quadrant pain is based on well-established clinical symptoms, physical examination and physician's experience.

Key words: Diagnostic dilemma; Left lower quadrant pain; Left-sided appendicitis; Midgut malrotation; Situs inversus totalis

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INTRODUCTION

Acute appendicitis is probably the most common intraabdominal condition requiring emergency surgery. The diagnosis is based on well-established clinical symptoms, basic radiologic findings and surgeon experience[1,2]. Approxi-

mately one third of patients with acute appendicitis have pain localized outside of the right lower quadrant because of the various positions of the appendix vermiformis, i.e. retrocecal, pelvic, subcecal, preileal and postileal, while subhepatic, meso-celiac, mid-inguinal and left-sided are seen more rarely[1,2].

Appendicitis causing pain in the left lower quadrant is extremely rare and can occur with congenital abnormalities that include true left-sided appendix or as an atypical presentation of right-sided, but long appendix, which projects into the left lower quadrant[2]. Left-sided acute appendicitis (LSAA) develops in association with two types of congenital anomalies: situs inversus totalis (SIT) and midgut malrotation (MM)[1-65]. Herein, we report an unusual case of SIT with acute appendicitis presenting as left lower quadrant abdominal pain. We also reviewed 95 published cases of LSAA with congenital anomalies retrieved from the Google Scholar and PubMed databases.

MATERIALS AND METHODS

We report a new case of LSAA with SIT. Additionally, for the review, a search of the English medical language literature in PubMed and Google Scholar was conducted for every case report, series, letter to the editor, original article and literature review relating to left-sided appendicitis. In addition, reference lists of the articles obtained and previous reviews were examined. Key words used were left lower quadrant pain, LSAA, appendicitis with left lower quadrant pain, SIT and appendicitis, acute left-sided appendicitis, and MM and appendicitis. The search included all articles from 1893 to July 2010. The articles containing adequate information such as patient age, sex, localization of the symptoms, time of the diagnosis, type of congenital anomalies, choice of incision and surgery were included in the study, while studies and comment articles with insufficient clinical and demographic data were excluded.

RESULTS

Case report

A 25-year-old female presented to the emergency unit on June 18, 2010 with severe abdominal pain, which started the previous night. The patient stated that the pain has begun first in the epigastric area and later expanded through the left lower quadrant. Besides the pain, she complained of inappetence and mild nausea. Patient history revealed no previous illness or surgery. On physical examination, rebound tenderness was observed in the left lower quadrant. Laboratory tests, including leukocyte count (9100 K/UL), were normal. Based on the patient's clinical status and our previous experience, we considered LSAA in the differential diagnosis; accordingly, relevant analyses were performed. Chest X-ray revealed dextrocardia (Figure 1) and abdominal ultrasonography (USG) demonstrated SIT and blind intestinal loop consistent with acute appendicitis in the left lower quadrant. The patient was immediately taken to surgery and as the laparoscopic device was out of order, a left McBurney's incision was performed. She was discharged on the second postoperative day with no complications (Figure 2). Contrast-enhanced thoraco-abdominal computed tomography (CT) in the postoperative period confirmed the diagnosis of SIT (Figure 3).

Figure 1 Chest X-ray showing dextrocardia.

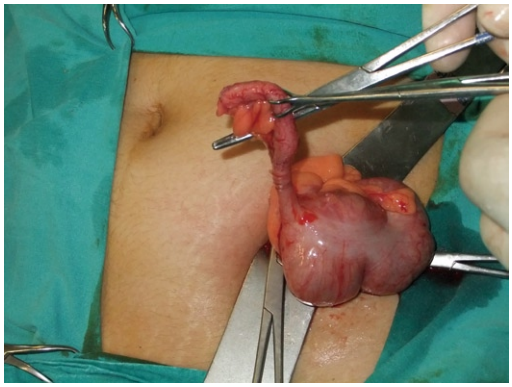

Figure 2 Intraoperative photograph showing an appendix together with cecum in the left lower quadrant.

Figure 3 Coronal contrast-enhanced multidetector computed tomography of the thorax, abdomen and pelvis. Computed tomography showed situs inversus totalis including dextrocardia, right-sided gastric bubble and reversed spleen and liver (postoperative view).

Analysis of literature about left-sided appendicitis

The English medical literature published to July 2010 in PubMed and Google Scholar databases was reviewed, and 64 reports concerning 95 cases of LSAA meeting the above-

Table 1 Summary of 95 reported cases of left-sided acute appendicitis with situs inversus totalis and midgut malrotation *n* (%)

Patient characteristics	Results
Mean age (yr, range)	29.1 ± 15.9 (8-82)
Sex	
Male	57 (60)
Female	38 (40)
Pain location	
Left-lower quadrant	59 (62.1)
Right-lower quadrant	14 (14.7)
Bilateral lower quadrant	7 (7.3)
Pelvic	2 (2)
Left-upper quadrant	7 (7.3)
Peri-umbilical	6 (6.3)
Congenital anomaly	
Situs inversus totalis	66 (69.4)
Midgut malrotation	23 (24.2)
Cecal malrotation	3 (3)
Unnoted	2 (2)
Other	1 (1)
Time of diagnosis	
Preoperative	49 (51.5)
Intraoperative	19 (20)
Known	14 (14.7)
Postop	5 (5.2)
Unnoted	8 (8.4)

mentioned criteria were included in this review[1-64]. The article types were as follows: case report - 48, case report and a review of the literature - 7, imaging for surgeon - 5, letter to the editor - 2, and abstract - 2. The clinicopathologic characteristics of the 95 patients are summarized in Table 1. The patients were aged from 8 to 82 years (mean: 29.1 ± 15.9 years). Fifty-seven were male (mean: 30.9 ± 15.2 years, range: 9-82 years) and 38 were female (mean: 26.5 ± 16.4 years, range: 8-76 years). Sixty-six patients had SIT, 23 had MM, 3 had cecal malrotation, in two the anomaly was previously unnoted and in one case, the end of the appendix running along the anterior side of sacrum was found in the left side. According to localization of the symptoms, 59 patients presented with left and 14 with right lower quadrant pain, 7 with bilateral lower quadrant pain, 7 with left upper quadrant pain, 6 with peri-umbilical, and two presented with pelvic pain. With regard to the diagnosis, 49 patients were diagnosed with appendicitis during the pre-operative period, in 19 patients, the diagnosis was established intraoperatively and in 5 postoperatively; 14 patients were previously known to have SIT and/or MM. No information was available in eight patients. Of 95 patients included in this literature review, 13.6% of patients underwent laparoscopic surgery. Laparoscopic appendectomy was performed in eight cases[6,10,12,13,15,21,24,58], and was combined with cholecystectomy in two[3,41] and with ablation of endometriosis implants in one patient[7]. In the remaining two patients, the intervention was switched to laparotomy due to technical reasons[8,25].

DISCUSSION

There are two anatomic abnormalities which result in LSAA - the first being SI, and the second, less common abnormality, is MM[6,7]. MM is the term used to describe a spectrum of congenital positional anomalies of the intestine caused by nonrotation or incomplete rotation of the primitive loop around the axis of the superior mesenteric artery (SMA) during fetal life. Although about 80% of cases are diagnosed in patients younger than 1 mo, malrotation has also been reported in older children and adults[8]. The incidence of MM cited in the literature varies from 0.03% to 0.5% in live births[1,3,9,10]. SI is an uncommon condition, which is caused by a single autosomal recessive gene with incomplete penetrance and occurs in 1 per 5000 to 1 per 10000 live births[7]. This condition may be complete (SIT), when both thoracic and abdominal organs are transposed, or partial, when only one of those cavities is affected[1]. The incidence of SIT reported in the literature varies from 0.001% to 0.01% in the general population[12-14], whereas the incidence of acute appendicitis associated with SIT is reported to be between 0.016% and 0.024%[5,13,14].

According to published reports in the literature, LSAA occurs between the age of 8 and 63 years and is 1.5-fold more frequent in men than in women[3,13]. In our review, we determined the mean age of the patients as 29.3 ± 16.1 (range: 8-82) years and the male/female sex ratio as 3/2[1].

The differential diagnosis of left lower quadrant abdominal pain includes: diverticulitis, renal colic, ruptured ovarian cyst, Meckel's diverticulitis, epididymitis, incarcerated or strangulated hernia, bowel obstruction, regional enteritis, psoas abscess, and right- and left-sided appendicitis (LSAA)[1,4].

LSAA is a diagnostic dilemma, because the appendix is located in an abnormal position. The differential diagnosis of LSAA may not be promptly established in the emergency setting and is often delayed due to lack of uniformity in the clinical signs[11,58]. It is assumed that even though the viscera are transposed, the nervous system may not show the corresponding transposition, which may result in confusing symptoms and signs. In about 18.4%-31% of patients with SIT and MM, the pain caused by LSAA has been reported in the right lower quadrant[1,5,11-13]. In this literature review, it was observed that 14.7% of patients had pain localized in the right lower quadrant, which indicates the importance of accurate preoperative diagnosis in order to avoid incorrect incision.

The diagnosis of acute appendicitis in patients with SIT or MM can be based on physical examination, electrocardiogram, chest X-ray, barium studies, USG, CT scan and diagnostic laparoscopy[1,2].

Plain radiographs are usually not helpful for establishing the diagnosis of appendicitis. However, the detection of dextrocardia on chest X-ray and right-sided gastric bubble on abdominal plane X-ray is of considerable value in establishing the diagnosis of SIT. Barium enema with gastrografin can reveal MM or SIT, when there are difficulties in making the diagnosis of acute left lower quadrant pain[11]. Over the last two decades, there has been an increasing use of imaging modalities, such as USG and

CT, in the diagnosis of acute appendicitis. USG is widely used in cases of appendicitis, however, it has significant limitations: it is operator-dependent, and examination of the lower quadrant can be compromised in patients with large body habitus or by overlying bowel gas. The value of CT in the diagnosis of acute appendicitis has been well-documented, with a reported accuracy of 90%-98%[2,13]. USG and CT may also be helpful in the detection of SIT and MM. Of the patients included in this literature review, CT has been used in the diagnosis of 28 patients and USG in 22 patients since 2000[1,6,8,17-24,26,27].

After establishing the diagnosis of SIT or MM, the surgical options are the same as for normal patients[1]. According to the reviewed literature, we observed that many open and a few laparoscopic procedures have been performed[1,6,8,15]. Laparoscopic appendectomy was first carried out in 1998 by Contini *et al*[58] in a 34-year-old male patient with SIT. Since then, laparoscopic appendectomy has been performed in a total of 20 cases (12 with MM and 8 with SIT), of which two have undergone cholecystectomy at the same surgical session[3,6,7,10,12,13,15,21,24,41,58,65]. We believe that laparoscopy may be very useful both in establishing the differential diagnosis and in performing the definitive surgery[1].

As in patients with normally localized appendix, appendectomy specimens in LSAA should be sent for pathological evaluation. To our knowledge, in the literature, only two of 95 patients (59 male, 76 female), who underwent appendectomy due to LSAA, were pathologically diagnosed with malignancy. Ascendent hemicolectomy was performed in both patients after pathological evaluation, which revealed mucinous adenocarcinoma and mucinous cystadenocarcinoma[19,26].

In conclusion, LSAA should be considered in the differential diagnosis of young patients presenting with pain localized in the left lower quadrant. Chest X-ray, abdominal USG and CT provide quite useful information. Diagnostic laparoscopy is the gold standard in cases with complicated differential diagnosis.

COMMENTS

Background

Acute appendicitis is a surgical condition, which manifests itself as pain usually localized in the lower right quadrant and can be diagnosed easily due to its clinical findings. Besides the surgeon experience, blood tests, radiological investigations, and in some circumstances, diagnostic laparoscopic techniques, should be used in the diagnosis of atypically localized appendicitis.

Research frontiers

Left-sided appendix is a rarely seen condition, which is frequently associated with situs inversus totalis (SIT) and midgut malrotation (MM). In this study, the authors performed an overall evaluation of management approaches and diagnostic processes in cases of left-sided acute appendicitis (LSAA) related to SIT and MM published in the English language literature to date, and presented the experience.

Innovations and breakthroughs

This study is the largest literature screening on LSAA to date.

Applications

This brief literature review demonstrates the importance of radiologic assessment and diagnostic laparoscopy along with surgical experience in the differential diagnosis of LSAA.

Peer review

The authors described a case of left sided appendicitis in the setting of situs inversus and a corresponding literature review. Over all, this paper is well written, concise and information.

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