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Research Article

ILEOURACHAL FISTULA: UNUSUAL INITIAL PRESENTATION OF CROHN'S DISEASE

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ABSTRACT

Crohn's disease is a chronic inflammatory disease with several clinical presentations. Complications usually occur after average of ten years of evolution. An enterovesical fistula complicating a Crohn's disease is a rare situation but enterourachal fistula is even rarer. It's a diagnosis challenging especially as an initial presentation of the disease. Few cases of entero-urachal fistula were published in literature. We present a case of 32-year-old female presented with three months history of diarrhea and had recently developed dysuria. She was admitted to our institution with fever, lower abdominal pain and sub umbilical swelling. Ultrasonography and computed tomography revealed parietal thickening of the distal ileum continuing with an infected urachal cyst which is consistent with Crohn's disease. Ileourachal fistula was highly suspected. After medical treatment, an urachal resection, partial cystectomy and ileal resections were performed. Primary ileostomy was used and re-establishing continuity was programmed 8 weeks later. After surgery, symptoms improved significantly. The aim of our report is to describe this rare complication with a review of literature and to assess the importance of medical and surgical management.

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INTRODUCTION

A 32-year-old female patient present to the emergency for fever, lower abdominal pain, vomiting, dysuria and subumbilical swelling.The patient had been experiencing one year of recurrent episodes of diarrhea (without blood or mucus), abdominal pain, decreased appetite and weigh loss. ileocolonoscopy with biopsy performed in another institution was normal.

Patient's symptoms started two weeks ago with lower abdominal pain and dysuria. She was diagnosed with a urinary tract infection (Escherichia Coli) and antibiotic therapy was administered. Symptoms worsening by fever, vomiting and sensation of abdominal swelling witch motivating a consultation in the emergency department.In the admission, patient was noted to be pale, febrile and panful. Other vital signs were within normal limits. Tenderness in the lower abdomen and a palpable infraumbilical mass were detected during abdominal examination. No umbilical discharge was noted.Laboratory examination showed elevated inflammatory markers, a deep hypochromic microcytic anemia and hyperleukocytosis. Ultrasonongraphy performed on emergency showed fluid collection immediately above the bladder extended up to umbilicus (Figure 1). There was a thickened small bowel adjacent to the collection (Figure 2).

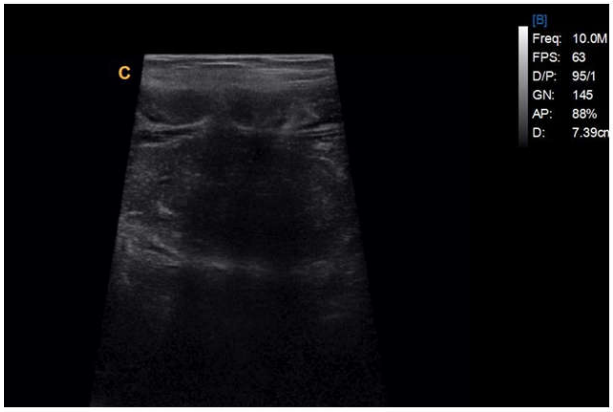

Figure 1 Echographic view of medline collection on the abdominal wall

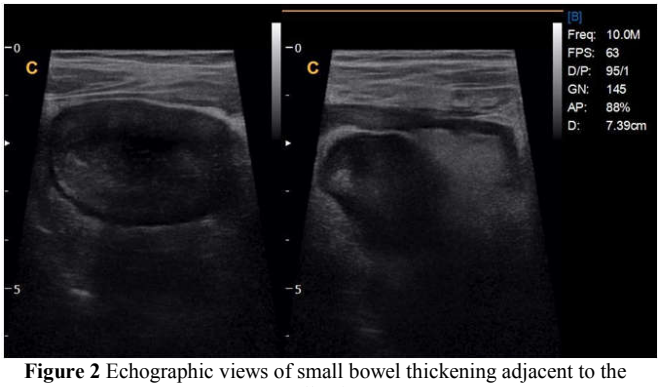

Figure 2 Echographic views of small bowel thickening adjacent to the collection

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Computed tomography (CT) scan confirmed ultrasonography finding. It showed an ovoid collection measuring 70x50mm with rim enhancing extending from umbilicus to the vesical dome consisting with an infected urachal cyst (Figure 3,4)

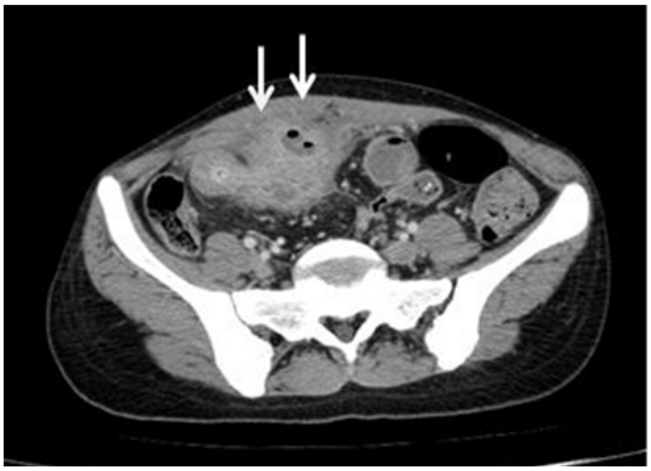

Figure 3 Axial computed tomography view of urachal infected cyst with small foci and gaz tracking (white arrows)

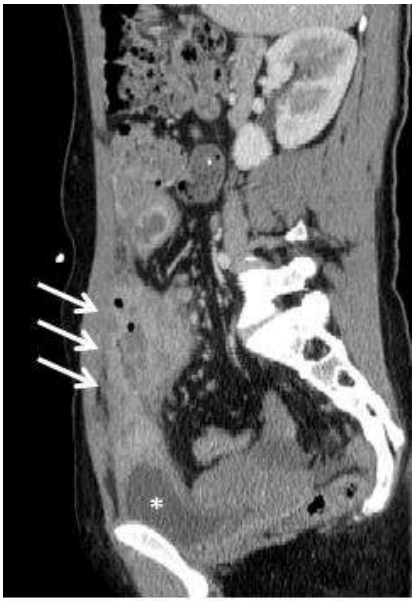

Figure 4 Sagittal computed tomography view of urachal abscess (white arrows) connects to the bladder (*)

Based on clinical history, laboratory tests and imaging findings we suspected a Crohn's disease complicated with an enterourachal fistula. After antibiotic, low dose steroids and anemia therapy, surgical resection was planned to control abscess and fistula. The diagnosis was confirmed by intraoperative findings. Abscesses cavity resections with urachal curettage were performed. The dome of the bladder was removed and ileal resection (280mm) including the fistula was performed to avoid recurrence (Figure5,6).

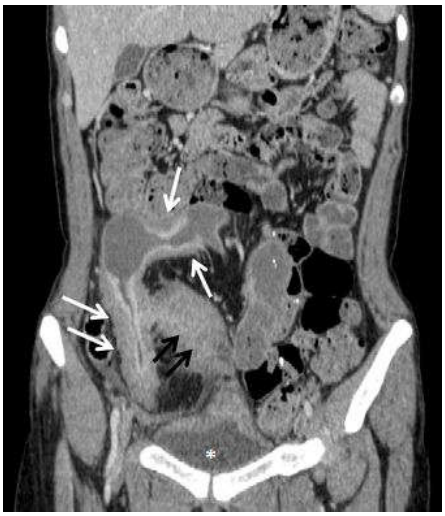

Figure 5 Coronal computed tomography images demonstrating the urachal abscess (black arrows) immediately in contact of ileitis (white arrows).

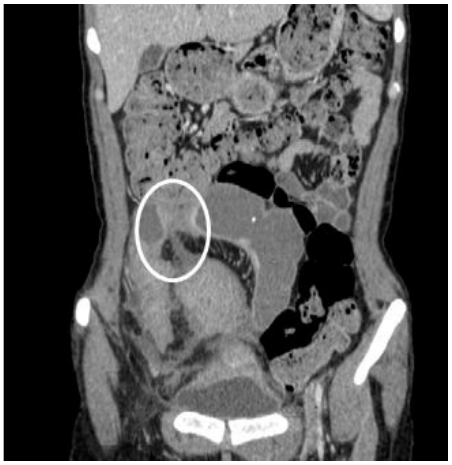

Figure 6 Coronal computed tomography images of the contact between the urachal abscess with ileitis highly suggestive of enterourachal fistula.

Because of severe local inflammation, ileostomy was made at first with a re-establishing of continuity in a second surgery two months later (Figure 7)

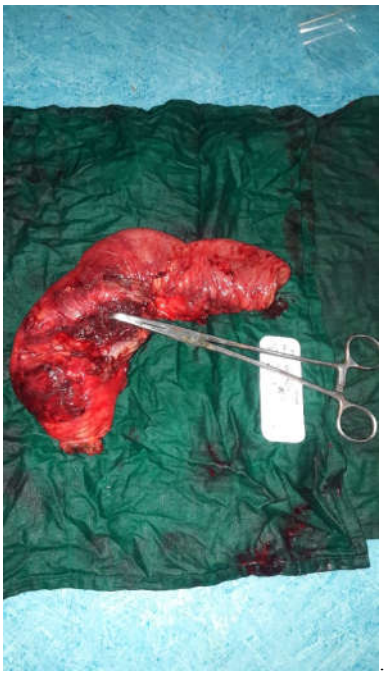

Figure 7 Partial ileal resection including fistula

References	age	sex	CD status	Symptoms	Investigations	Urachus abnormality	BowelInvolved	Treatment
Davidson. 1980[6]	28	M	Unknown CD	Abdominal pain, fever,Umbilicaldischarge	Sinogram Cystoscopy	Enterourachovesico-cutaneous fistula	Ileum and cecum	Urachalresection Partial cystectomy Ileocecalresection
Artigas et al., 1998[7]	20	F	Known CD	Abdominal pain, Umbilicaldischarge,	Sinogram Ultrasound CT	Enterouracho-cutaneous fistula	Ileum	Urachalresection Partial cystectomy Ileocecalresection
Klineberg et al., 2002[8]	19	M	Known CD	Abdominal pain, Dysuria	CT Colonoscopy Cystoscopy	Enterourachalfistula	Ileum	Urachalresection Partial cystectomy Ileocecalresection
Solem 2002[3]	n/d	n/d	Known CD	n/d	n/d	Urachalabscess	Ileum	n/d
	n/d	n/d	Known CD	n/d	n/d	Urachalabscess	Ileum	n/d
Sugiyama et al., 2003[9]	19	F	Unknown CD	Hypogastric pain, fecaluria	CT Cystoscopy Contrast x ray	Urachal abscess and secondary enterovesical fistula	Small bowel	Urachalresection Partial cystectomyileumresection
Keir et al., 2004[10]	16	F	Unknown CD	Umbilical discharge granulomatous lesion in the umbilicus	CT MRI Sinogram Colonoscopy	Infected patent urachus	Appendix, terminal ileum and cecum	Resection of urachus Appendectomy Right hemicolectomy
Bergman and Sloots, 2005[11]	19	M	Uknown CD	Fecaldischarge at umbilicus	CT withcontrast	Enterourachalfistula	Ileocecal	Urachal resetion Partial cystectomy Ileoceal resection
Weitten et al., 2005[12]	21	M	Unkown CD	Chronicfever	CT	Urachalcystabscess	ileum	Urachalcyst ablation
Ishii et al., 2007[13]	n/d	n/d	Known CD	n/d	n/d	Urachalabscess Enterourachocutaneousfitulas	n/d	Resection of the urachus,partial bowel resection partial cystectomy.
Hollander et al., 2012[14]	15	M	Unkown CD	Abdominal pain, infraumbilical mass, perianal fistula	CT MRI Colonoscopy Laparoscopy	Enterourachal Fistula obliterated by medical treatment	ileum	Urachal resection Partial cystectomy No bowel resection
Yheulon et al., 2013[15]	18	F	Known CD	Abdominal pain,	CT Cystoscopy	Enterouracho-vesical fistula. A retained video capsule in the fistuloustrack	Terminal ileum	Urachalresection Partial cystectomy Partial ileal resection
O'Brien et al., 2013[16]	26	M	Known CD	Abdominal pain, fever umbilicaldischarge	CT Laparoscopy	Infectedurachalcyst	cecum	Urachal cyst excision Right hemicolectomy
MadoR and Blair, 2014[17]	11	F	Known CD	umbilicaldischarge	US MRI Laparoscopy	Enterouracho-cutaneous fistula	ileum	Urachalcystresection Partial cystectomy Ileocecalresection
Tsukui et al., 2017[18]	31	F	Unkown CD	Abdominal pain Dysuria Umbilicaldischarge	CT Colonoscopy Cystoscopy	Ceco-urachalfistula	cecum	Urachalresection Partial cystectomy Ileocecalresection
Kuroki et al., 2018[19]	29	M	Known CD	Abdominal pain	CT	Urachalabscess	Ileum	Urachalresection Partial cystectomy
	43	M	Known CD	Umbilicaldischarge Pneumaturia Fecaluria	CT, Bowel contrasted xray	Enterourachalfistula	Ileum	Urachalresection Urachal curettage Partial cystectomy
SenthilkumarSankararaman, et al., 2019[20]	17	M	Unkown CD	Abdominal pain, dysuria, anterior abdominal wall mass	CT MRI	Enterourachalfistula	Ileum	Ileocecal resection Resection of adjacent abscess cavity
Current case	24	F	Unkown CD	Abdominal pain Dysuria Abdominal wall mass	US CT	Enterourachalfistula	Ileum	Urachalresection Partial cystectomy Partial ileal resection

Histopathological examination showed perforated ileal fistulas with adenitis. Granulomatous lesions without necrosis were seen in the ileum which was consistent with Crohn's disease.

Patient was sent home after surgery with antibiotic and steroid treatment. No abscess recurrence was noted. A follow-up at gastroenterology department is planned for maintenance therapy.

DISCUSSION

Crohn's disease (CD) is a chronic inflammatory condition of the gastrointestinal tract. It is characterized by transmural granulomatous inflammation leading to complications like fistula, bowel stricture or abscess. Fistula may form between bowel loops and any adjacent structure. Majority of patient are experiencing surgical resection within 10 years of diagnosis[1] and 41% of them are developing a fistula within 30 years of evolution[2]. The urachus is an embryologic tract resulting from the involution of the allantoic canal. It's a fibrous cord connecting the bladder dome to the posterior umbilicus. Rarely, uncomplete obliteration of foetal urachus can occur leading to four types of urachal abnormalities: Patent urachus, urachal cyst, urachal sinus and vesicourachaldiverticulum [4]. These entities are usually seen in childhood, and a late onset in adulthood is a rare situation.

Internal fistulization is a frequent complication of Crohn's disease. Enterovesical fistulas occur in 5,6 % of patient with Crohn's disease[5], but enterourachal fistula is extremely rare[3].

A systematic search in Medline and Embase Databases using terms Crohn's disease, urachal and fistula identified only eighteen cases published since 1980, date of the first case reported. Including our case, the total number of patient with Crohn's disease and urachal complication is nineteen[3,6–20] (Table 1)

The cases consisted on nine men and six female, ranging in age from 11 to 31 years (mean 21years).Abdominal pain was the most frequent clinical symptom. Umbilical discharge and urinary signs were also described. These nonspecific clinical signs can cause diagnosis delay.

Including our patient, the enterourachal fistula as initial presentation of Crohn's disease was described in nine cases

[6,9–12,14,18,20]. In these situations, correct preoperative diagnosis was a challenging. Past history of digestive symptoms like recurrent diarrhea, weight loss and anemia helped suspecting inflammation bowel disease.

Imaging investigations such ultrasound, CT, MRI or barium enema study plays an essential role in the preoperative diagnosis. In all cases urachal abnormalities was successfully diagnosed by the radiologist and the presence of adjacent inflammatory bowel wall thickening was helpful in the suspicion of enterourachal complication of CD.

As mentioned in the literature review, the initial diagnosis was confirmed during exploratory surgery. The enterourachal fistula could be obliterated with the medical management using steroids and antibiotics, which could be very helpful in reducing bowel inflammation and limiting surgical resection [14]. In our case, despite the use of medical treatment, single step surgery wasn't possible because of severe inflammation and ileostomy was first performed before a re-establishing continuity two months later.

Surgical resection is the standard treatment of enterourachal fistulas. It's consisted on urachal resection, partial cystectomy with intestinal resection. The medical management alone described in previous study usually results in recurrence [21]. Tsukui *et al*.[18] suggested that complete resection of bowel involved in fistula formation is necessary to treat entero-urinary tract fistulas in patient with Crohn's disease.

Urachal resection is also recommended because of potential of malignant transformation in primary urachal adenocarcinoma [22]. However management of asymptomatic urachal remnants remains controversial.

CONCLUSION

We report a case of enterourechal fistula as initial presentation of Crohn disease with nonspecific symptoms and without umbilical discharge. The preoperative diagnosis was successfully made by imaging findings avoiding diagnosis delay. Medical and surgical management as described by prior studies is recommended for complete cure.

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