

A Peculiar Three-way Connection: Colo-urachal-vesical Fistula with Cutaneous Extension

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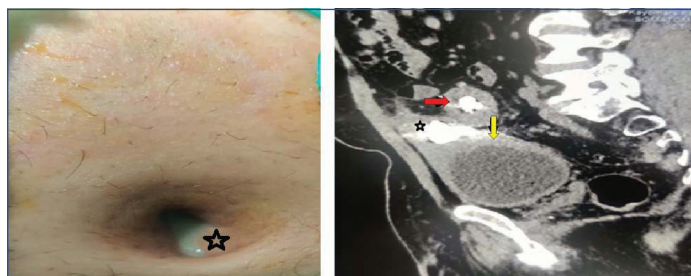
ABSTRACT

Urachal anomalies are quite a rare phenomenon in adults. This report is about a patient of a colo-urachal-vesical fistula with cutaneous extension. A 68-year-old diabetic male patient presented with purulent umbilical discharge since 15 days. Investigations revealed a colo-urachal-vesical fistula communicating with the umbilicus. He underwent a colo-urachal fistula excision with sigmoid-colectomy and bladder-cuff-excision. A thorough literature search revealed only nine reported cases so-far. While the most common symptom was faeculent umbilical discharge, this patient had a slightly varied-presentation of purulent-discharge. This is also the first study to conclusively demonstrate a vesical connection along with colo-urachal and cutaneous extensions.

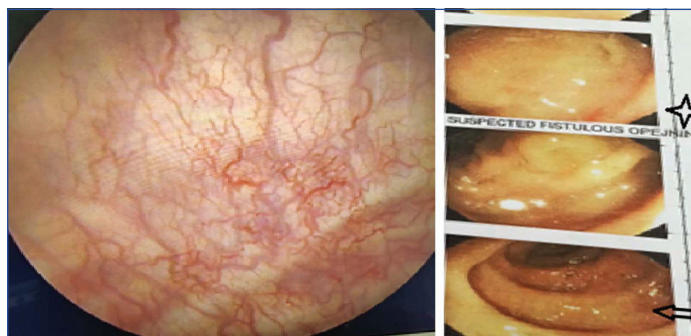
Keywords: Cystoscopy, Umbilical discharge, Urachus

CASE REPORT

A 68-year-old diabetic male patient presented to the Urology Out Patient Department (OPD) with complaints of umbilical discharge, since last 15 days, which was also observed to increase during straining. He had no associated symptoms or complaints. On examination purulent umbilical discharge was noticed [Table/Fig-1]. A contrast was injected from the fistulous opening and Computed Tomography (CT) scan of the abdomen with pelvis was obtained, which showed contrast opacification of the sigmoid colon with its adherence to dome of the bladder. It also revealed significant bladder wall thickening [Table/Fig-2]. Cystoscopy was also performed wherein the area at the junction of the bladder dome and left anterolateral wall was found to be oedematous and inflamed [Table/Fig-3]. Colonoscopy revealed a fistulous opening in the sigmoid colon and the presence of a diverticulum [Table/Fig-4].

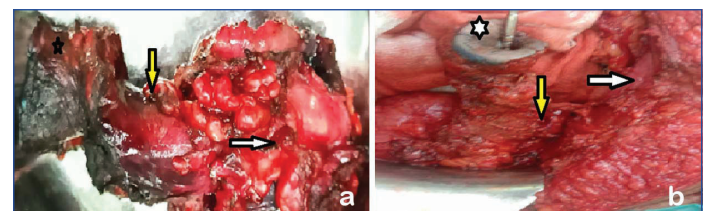


[Table/Fig-1]: Clinical picture of patient with colo-urachal-vesico-cutaneous fistula presenting with a purulent umbilical discharge. **[Table/Fig-2]:** CT scan showing contrast instilled through the umbilical opening of the fistula (star), opacification of the sigmoid colon (red arrow) with adherence to the dome of the bladder and bladder wall thickening (yellow arrow). (Images from left to right)



[Table/Fig-3]: Cystoscopy showing inflamed and oedematous mucosal area at the junction of the dome and left anterolateral bladder wall. **[Table/Fig-4]:** Colonoscopy images showing a suspected fistulous opening (star) in the sigmoid colon along with presence of a diverticulum (arrow). (Images from left to right)

The patient underwent an exploratory laparotomy. The fistulous tract was traced and found to be communicating with umbilicus, reaching till sigmoid colon and also involving the urinary bladder. The sigmoid colon was also found to be adherent to the dome of bladder. Communication with bladder was further confirmed by instillation of methylene blue dye. Complete resection of the fistulous tract, umbilicus and bladder cuff was performed along with a sigmoid colectomy with primary end to end colorectal anastomosis [Table/Fig-5]. Preoperatively, the patient was started on piperacillin-tazobactam and metronidazole antibiotics, which was continued for 15 days after the surgery. Postoperative course in ward was uneventful and he was discharged after one week. Histopathology revealed an inflamed colon with a patent remnant of urachus. Upon follow-up after one month, he was asymptomatic, without any bowel or bladder complaints and had normal serum parameters.



[Table/Fig-5]: Excised en bloc specimen containing the umbilicus (star), fistulous track (yellow arrow-a and b) and sigmoid colon with bladder cuff (white arrow-a and b).

DISCUSSION

Cases with persistent urachal remnants are uncommon and predominantly seen in the paediatric population. Colo-urachal-fistulae are even more uncommon; very few cases have been reported, while the one conclusively demonstrating a patent vesical communication hasn't been reported so far. Although cases of enterourachal-fistula have been reported secondary to Crohn's disease primarily in the small intestine, a colonic involvement is unusual [1,2]. In 1945, Sawyer was the first person to diagnose, secondary to diverticulitis, a fistulous connection between the sigmoid colon and a large urachal cyst [2].

A thorough search of all the available literature revealed only nine similar cases which have been published so far [Table/Fig-6] [1-9]. The age group of presentation in these cases was 55-65 years, [3] all but one being male patients. Only one study had a female patient who presented at 26 years of age [1]. The most common symptom was a prolonged history of faeculent umbilical discharge

Author	Age (years), Sex	Clinical presentation	Procedure performed
Dickoff C et al., [3] 2008	56, M	Purulent umbilical discharge	CUF excision, sigmoid colectomy with double loop ileostomy f/b colostomy closure later
Yang D et al., [5] 2020	56, M	Facial swelling with emphysema with pneumo-mediastinum	CT guided drainage
Gargouri MM et al., [2] 2013	51, M	Faeculent umbilical discharge	Urachal cyst with CUF excision, sigmoid colectomy and Hartman's procedure.
Rapoport D et al., [6] 2007	68, M	Greenish discharge with gas	CUF excision with sigmoid colectomy and Hartman's procedure
Mohan HM and Winter DC [8] 2010	91, M	Faeculent umbilical discharge	Insertion of percutaneous drain
Coons BJ et al., [9] 2009	44, M	Rupture of infected urachal cyst with purulent discharge	CUF excision with sigmoid colectomy with bladder cuff excision with end to end colonic anastomosis
Sakata S et al., [4] 2016	54, M	Faeculent umbilical discharge	Laparoscopic CUF with urachal cyst excision with removal of involved bladder with high anterior resection with primary anastomosis
Soyster M et al., [7] 2018	64, M	Faeculent umbilical discharge with umbilical mass	CUF excision with end to end colonic anastomosis
Quek ML et al., [1] 2003	26, F	Faeculent umbilical discharge	CUF excision with sigmoid colectomy with bladder cuff excision with end to end colonic anastomosis
Current study, 2022	68, M	Purulent umbilical discharge	CUF with vesico urachal fistula excision with bladder cuff removal with sigmoid colectomy with end to end primary colonic anastomosis.

[Table/Fig-6]: Summary of all colo-urachal fistula cases diagnosed till date as available in literature [1-9].

CUF: Colo-urachal fistula

indicating communication with gastrointestinal tract [3]. However, in present study patient presented with purulent umbilical discharge over a short duration. All patients had Colo-urachal fistulae on intraoperative assessment; 50-60% of patients also had cutaneous communication. Bladder wall thickening and adherence were found in two cases, whereas, this was the only study to have a definitive vesical extension also which was clearly demonstrated on methylene blue dye injection. All studies including the present one were operated by open approach except Sakata S et al., which used a laparoscopic method [4]. Bowel continuity was established immediately in four cases (including the present study) whereas in 3 cases, it was done at a later date [5]. Surprisingly, none of the study reported malignancy upon histopathological examination.

Most of the studies didn't report any long-term sequelae. In order to evaluate the relationship with surrounding organs, Magnetic Resonance Imaging (MRI) is a useful modality which also establishes continuity between the urachal cyst and the umbilicus when the fistula cannot be recognised by a CT scan [2].

Diverticulitis may also lead to formation of colo-urachal fistulae (seen in four cases including the present case; 50% incidence) which irritates a pre-existing urachal cyst [6]. While in rest of the cases, fistulae may develop as a result of fistulisation of an infected urachal cyst into colon following a prolonged period of irritation. A pre-existing urachal-umbilical sinus or its reopening after postnatal regression can lead to development of a communication between the colo-urachal cyst and umbilicus [2]. Blichert-Toft M and Nielsen OV observed a high rate of recurrence (upto 31%) in cases of incision and drainage of infected urachal cysts [10]. Hence, to avoid recurrence or carcinogenesis, the current treatment practice for such disorders includes an en bloc surgical excision of the urachus along with removal of the involved portion of the colon, umbilicus and bladder cuff [2]. Depending on the preoperative conditions, colonocolic anastomosis could be established primarily or it could be delayed [7].

CONCLUSION(S)

Colo-urachal fistulae can be diagnosed radiologically; CT reconstruction helps to establish communication among the urachal cyst, colon and umbilicus. En bloc colo-urachal-fistula excision with a sigmoid-colectomy and bladder-cuff-excision is imperative to prevent recurrence and carcinogenesis.

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