CASE REPORT

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A Rare Cause of Secretory Diarrhea Causing Syncope Attack: Mckittrick-Wheelock Syndrome

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ABSTRACT McKittrick-Wheelock syndrome (MKWS) is characterised by electrolyte abnormalities such as secretory diarrhoea with mucus, hypokalemia, hyponatremia, dehydration from fluid loss, and/or prerenal acute renal failure. Depending on the severity of clinical symptoms, syncope may happen. This is caused on by a massive hypersecretory villous adenoma, which is typically seen in the rectum or rectosigmoid region. If untreated, this condition could worsen and result in death, so a careful evaluation is necessary. Depending on the size, location, and level of involvement in the colorectal wall, the villous adenoma causing the condition may be treated by endoscopic, laparoscopic, or open surgery. This article's goal is to demonstrate an assessed case of syncope that underwent examinations and was ultimately classified as MKWS.

Keywords: Hypokalemia; villous adenoma; diarrhea; syncope; McKittrick-Wheelock syndrome

Colon polyps are usually asymptomatic, raised formations of the mucosa. When located in the rectum, they can occasionally ulcerate and cause bleeding, tenesmus, and obstruction by obstructing the lumen. They can occasionally induce secretory diarrhea, dehydration, electrolyte imbalances, and acute renal failure. Adenomas are the most common type of neoplastic polyp found in the intestines. They can be cylindrical, villous, or tubulovillous in nature. Villous structures account for 5% of this group and bear a 15-40% risk of malignancy, depending on size.[1] Secretory activity is present in 3% of those larger than 3 cm.[2-4] The epidermis of secretory villous adenomas contains mucin-secreting goblet cells.[5-7]

For the first time in the literature, Garris defined "rectal polyp causing prerenal uremia" in 1941.[8] Later, in 1954, McKittrick and Wheelock characterized the syndrome that bears their names.[6] As a result, three components comprise this potentially fatal syndrome: (1) chronic mucus diarrhea; (2) renal dysfunction with hydro-electrolyte imbalance; and (3) a hypersecretory large villous adenoma situated in the rectum or rectosigmoid junction. The colon impacted by the large villous adenoma secretes water, sodium, and potassium, whereas the normal colonic mucosa reabsorbs sodium and water and secretes potassium.

This article describes a case of mucus-producing secretory diarrhoea caused by a villous adenoma encompassing the whole rectum, which resulted in hyponatremia, hypokalemia, and dehydration. Syncope and dizziness developed as the process progressed.

CASE REPORT

The patient provided informed consent for this case report.

A 68-year-old female patient was admitted to the emergency outpatient clinic with nausea, watery stools, dizziness, and hazy consciousness. There were no neurological or otological abnormalities. It was discovered that she had mucoid defecation 8-10 times

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a day on average during the last seven months. Rectal examination revealed a palpable large lesion. Biochemical tests revealed Na: 127 mmol/L, K: 2.43 mmol/L, Cl: 80.7 mmol/L, creatinine: 2.78 mg/dL and urea: 119 mg/dL. C-reactive protein and leukocyte counts were both normal. The rectum appeared to be dilated (78 mm in diameter) and dense, according to computed tomography. A mass lesion was detected as an intraluminal polypoid soft tissue density with a width of 30 mm, beginning about 2 cm distal from the anal canal entry and spreading along the 12 cm segment of the rectum (Figure 1). Except for similar results on magnetic resonance imaging, there was no invasion in the mesorectal fatty tissue and fascia, and the mass had malignant characteristics (Figure 2). A villous mass lesion covering 3/4 of the rectum

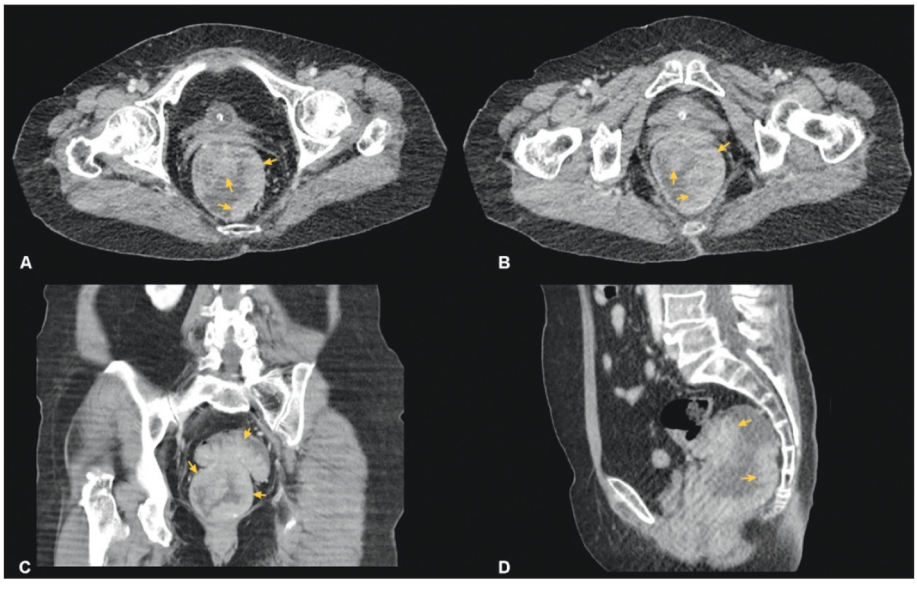

FIGURE 1: Axial **(A, B)**, coronal **(C)**, and sagittal **(D)** plan intravenous contrast-enhanced computed tomography images show an intraluminal polypoid lesion (arrows) with soft tissue density resulting in expansion of the rectum, beginning about 2 cm distal from the anal canal entry and spreading along the rectum.

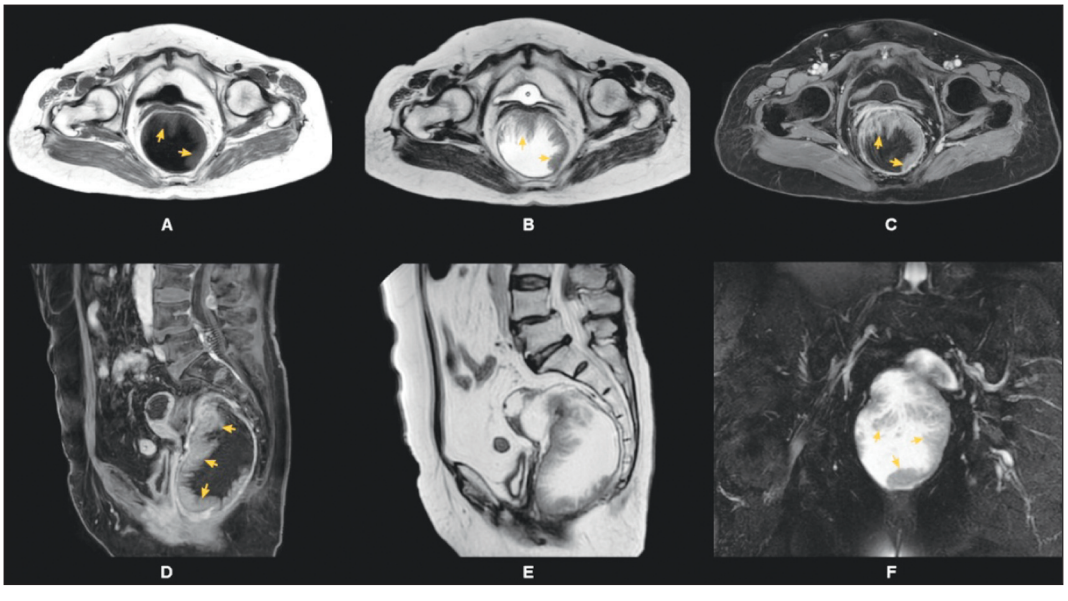

FIGURE 2: Contrast-enhanced pelvic magnetic resonance imaging demonstrates an intraluminal polypoid lesion (arrows) in the rectum without invasion in the mesorectal fatty tissue and fascia. **(A)** axial plan T1-WI; **(B, E)** axial and sagittal plan T2-WI; **(C, D)** axial and sagittal plan fat-saturated contrast-enhanced images; (F) coronal plan fat-saturated T2-WI. WI: Weighted imaging.

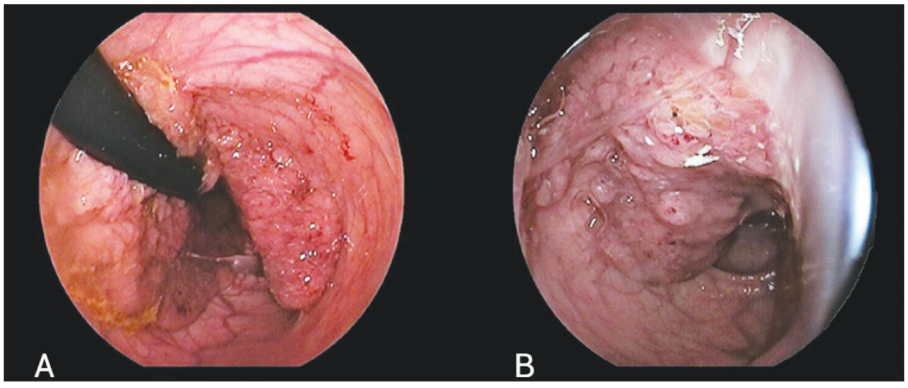

FIGURE 3: Colonoscopic view. **(A)** Villous polyp covering 3/4 of the rectal wall in retroversion. **(B)** View of the part of the polyp located in the ampulla recti.

wall, beginning at the proximal dentate line at the entrance to the anal canal and extending to the rectosigmoid junction, was discovered during a colonoscopic examination (Figure 3). The microscopic analysis of the tissue samples revealed no invasive malignancy and tubulovillous adenomas with areas of high-grade dysplasia. The present findings include ultra-low anterior resection, colo-anal anastomosis, and diverting ileostomy. A polypoid lesion in the rectum measuring 11x8.5x6 cm and positioned 0.2 cm from the distal surgical border was discovered during a macroscopic examination of the resection material. The polyp as a whole was sampled and studied. There was a complex architectural structure, loss of polarity, cellular stratification zones, and enhanced staining with p53 immunohistochemical staining in these areas, as well as a high proliferation index with Ki-67. The case was reported as a tubulovillous adenoma with high-grade dysplasia (Figure 4). Adjuvant treatment was not required, and the ileostomy was closed in the fourth week following surgery. The electrolyte imbalance resolved, and the frequency and regularity of defecation returned to normal.

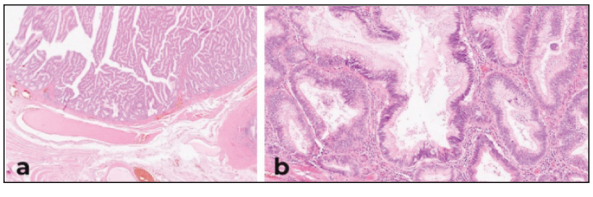

FIGURE 4: (A) Tubulovillous adenoma, H&E, x10. **(B)** Tubulovillous adenoma, H&E, x10, areas of dysplasia.

DISCUSSION

Diarrhea is defined as an increase in stool frequency, fluidity, and volume. It is determined by a variety of circumstances. It happens as a result of reduced water absorption, increased colon secretion, or a combination of the two.[9] It can be either osmotically or secretory mediated. Osmotic diarrhea is caused by substances that are not absorbed but are present in the intestinal lumen and draw fluid from the plasma into the intestinal lumen. Secretory diarrhea impairs electrolyte transfer from the colon mucosa, lowers water absorption, and increases secretion into the lumen. The fecal osmotic gap is used to differentiate between the two. In osmotic diarrhea, this number is greater than 125 mOsm/kg; in secretory diarrhea, it is less than 100 mOsm/kg.[10] Secretory diarrhea can be caused by a variety of conditions, such as colonic neoplastic tumors (including lymphomas, villous adenomas, and colon cancer) (Table 1).[9] Villous adenomas with secretory diarrhea are typically 4-18 cm in size and produce 500-3,000 cc of feces each day.[10] Prostaglandin E2 and cyclic adenosine monophosphate allow adenomas to release active ions into the lumen. They have a high number of goblet cells and significant COX 2 activity.[11] In the current case, the fecal anion gap level was 80 mOsm/kg, and the average daily stool volume was 2,800 mL, indicating secretory diarrhea.

According to the systematic review by Orchard et al., patients with McKittrick-Wheelock syndrome (MKWS) were 69 years old on average, had 92% diarrhea, an average of 10 defecations per day, and a

TABLE 1: Causes of secretory diarrhea.

1. Peptide-secreting endocrine tumors
• Gastrinoma
• Carcinoid
• VIPoma
• Somatostatinoma
• Glucagonoma
2. Infection
3. Bile acid malabsorption
4. Nonosmotic laxative use
5. Inflammatory bowel diseases
• Crohn's disease
• Ulcerative colit
• Microscopic colitis
6. Broken regulation
• Post-vagatomy syndrome
• Diabetic neuropathy
7. Neoplasia
• Lymphoma
• Villeus adenoma
• Colon cancer
8. Idiopathic and epidemic secretory diarrhea

stool volume of 1,900 mL. Furthermore, the average concentrations of sodium, potassium and chlorine are 122 mmol/L, 2.7 mmol/L, and 75 mmol/L, respectively. The tumour was found in the rectum in 60.5% of patients and at the rectosigmoid junction in 32.4% of cases. It is approximately 5 cm distant from the anal margin and has an average diameter of 12 cm. It completely ringed the lumen, and dysplasia was detected in 26.8% of the patients. A total of 30.4% of the cases underwent anterior resection, while 27.2% underwent abdominopelvic resection.[12] Our patient was diagnosed with an 11-cm tubulovillous adenoma with high-grade dysplasia that invaded the whole rectum. It surrounded three-quarters of the lumen, starting at the anal border. The mesorectal planes were preserved with an ultra-low anterior resection utilising oncological principles. An end-to-end coloanal anastomosis was performed using a circular stapler, and the opened loop ileostomy was closed after six weeks.

The review by Malik et al. highlighted the 23-month duration of symptoms prior to diagnosis.[13] This period gets shorter when the distribution is determined based on the years.[12] The duration between the onset of symptoms and the operation date in the present case was seven months, which is less than the average of the literature. As awareness grows, the transition period from diagnosis to therapy will be shorter.

In cases of severe fluid loss, Fernández-López and Paredes-Cotore suggested taking 400 mg of indomethacin daily to minimize PGE2 activation until the surgical surgery.[14] On the other hand, Caliskan et al. found that octreotide acetate was useful in individuals who refused surgery and had insufficient indomethacin tolerance.[15] In the current case, medical therapy for diarrhea was not attempted; instead, liquid electrolyte support was given. This option should be investigated to keep clinical findings under control before surgery. Furthermore, it should be noted that the risk of malignancy in MKWS is very low, and efforts should be made to ensure that the surgical intervention conducted is as minimally invasive as possible.

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: *Sami Açar;* ***Design:*** *Sami Açar;* ***Control/Supervision:*** *Nurten Türkel Küçükmetin;* ***Data Collection and/or Processing:*** *Sami Açar, Can Aydın, Sevil Karabağ;* ***Analysis and/or Interpretation:*** *Sami Açar, Sevil Karabağ;* ***Literature Review:*** *Hadi Sasani, Sami Açar;* ***Writing the Article:*** *Sami Açar, Sevil Karabağ;* ***Critical Review:*** *Hadi Sasani;* ***References and Fundings:*** *Sami Açar;* ***Materials:*** *Sami Açar.*

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