CASE REPORT

A case of giant rectal villous tumor with severe fluid-electrolyte imbalance treated by laparoscopic low anterior resection

Won Ho Choi, Jongpil Ryuk, Hye Jin Kim, Soo Yeun Park, Jun Seok Park, Jong Gwang Kim, Gyu-Seog Choi

Colorectal Cancer Center, Kyungpook National University Medical Center, Kyungpook National University School of Medicine, Daegu, Korea

McKittrick-Wheelock syndrome is a disorder caused by fluid and electrolyte hypersecretion from a colorectal tumor. To present the case of a patient with a giant rectal villous tumor with McKittrick-Wheelock syndrome who was successfully treated with laparoscopic surgery. The case of a 59-year-old man who came to the emergency department with syncope, prerenal azotemia, and electrolyte disturbances with a background of chronic diarrhea is reported. His condition was the result of fluid and electrolyte hypersecretion caused by rectal villotubular adenomas. Laparoscopic low anterior resection and subsequent volume and electrolyte replacement therapy resulted in complete recovery. A microscopic examination revealed multiple, well-differentiated adenocarcinomas arising in villotubular adenomas. Laparoscopic surgical resection is a feasible therapeutic modality for McKittrick-Wheelock syndrome.

Key Words: Diarrhea, Renal insufficiency, Villous adenoma, Laparoscopy

INTRODUCTION

McKittrick-Wheelock syndrome, which is a disorder characterized by fluid and electrolyte depletion, is caused by a secretory colorectal tumor [1-3]. The majority of tumors are villous adenomas [1,3]. The main clinical features of McKittrick-Wheelock syndrome are dehydration, mucous diarrhea, and symptoms of hyponatremia (headache, nausea, weakness, muscle cramps, lethargy, and seizures), and hypokalemia (fatigue, paresthesias, cramps, ileus, vomiting, hypotension, cardiac arrhythmias, and electrocardiographic changes). Laboratory findings typically reveal prerenal azotemia, hyponatremia, and hypokalemia [1,3].

Endoscopic resection is frequently difficult owing to the large size, unfavorable location, extension, or malignant change of the tumor. Laparoscopic surgical resection is an effective, rapid, and safe alternative in such cases [4].

We describe the case of a 59-year-old man who suffered from diarrhea, acute renal failure, and electrolyte dis-
turbances caused by a giant rectal villous tumor.

CASE REPORT

A 59-year-old man arrived in our emergency department with the sudden onset of syncope. On arrival, he was clinically dehydrated, with reduced skin turgor and dry mucous membranes. Examination revealed that the patient had decreased blood pressure (70/50 mmHg) and an increased heart rate (94 beats/min). The patient is an entrepreneur, with no history of any medication use. Laboratory tests revealed a serum sodium (Na) level of 122 mmol/L, potassium (K) level of 1.7 mmol/L, blood urea nitrogen (BUN) level of 96 mg/dL, and creatinine (Cr) level of 2.6 mg/L. The peripheral blood leukocyte count was $8.05 \times 10^3$ leukocytes/μL, hemoglobin was 17.4 g/dL, and platelet count was $449 \times 10^3$ platelets/μL.

A central venous catheter and urinary catheter was inserted to monitor the central venous pressure and urine output. After hydration by intravenous isotonic saline and potassium chloride (20 mgEq/L) replacement, the patient’s blood pressure recovered (118/78 mmHg), but he remained potassium-depleted (2.0 mmol/L) in the emergency room.

He was admitted for further treatment of his dehydration and electrolyte imbalances. After subsequent resuscitation, all physical and serological parameters were normalized and maintained (Na, 138 mmol/L; K, 3.8 mmol/L; BUN, 16 mg/dL; Cr, 1.1 mg/dL).

A detailed review of his history revealed that the patient had been suffering from chronic diarrhea for approximately 10 years. Except for this condition, he was otherwise healthy. However, in the past month, the diarrhea had worsened (more than 20 times a day), and he felt progressive malaise and frequent cramps in his legs. He had also undergone marked weight loss (10 kg in a month). Digital rectal examination revealed a huge, firm tumor with a velvety surface on the whole circumference of the rectum, 7 cm from the anal verge. Abundant bloody mucous rectal discharge was also present. An abdominal computed tomography (CT) scan was performed to check the lesion; it revealed a huge mass throughout the rectum and distal sigmoid colon (Fig. 1). Full colonoscopy also demonstrated huge conglomerated polyps in the rectosigmoid colon; the other part of the colon was unremarkable (Fig. 2).

On the seventh day of hospitalization, a laparoscopic low anterior resection with high ligation of the inferior mesenteric artery was carried out. The laparoscopic technique was hampered by the distension of the rectal ampulla owing to the intrarectal huge tumor and mucus. Thus, the distal part of the lesion was resected twice to ensure a safe distal margin, as the distal resection margin (DRM)
The ARF treated with laparoscopic surgery

Fig. 2. Colonoscopy revealed multiple polyoid lesions throughout the distal sigmoid colon and rectum.

Fig. 3. Macroscopic appearance of multiple polypoid lesions, measuring 25 cm × 12 cm. Most of them were villotubular adenomas. In the central ulcerative area (*), adenocarcinoma with subserosal invasion was confirmed.

Fig. 4. Microscopic appearance of the well-differentiated adenocarcinoma arising in a villotubular adenoma (H&E, ×40).

was free from adenoma, and DRM length was 8 cm while proximal resection margin length was 39 cm in the pathological report; however, the other part of the specimen was not photographed (Fig. 3). However, the soft feature of the lesion did not preclude adequate visualization of the pelvic structures. Therefore, a standard low anterior resection with total mesorectal excision and high ligation of the inferior mesenteric artery was performed successfully. The
pathological diagnosis was multiple adenocarcinomas arising in villotubular adenomas (T3N0M0, stage IIA) (Fig. 4).

The patient recuperated favorably during the postoperative days. He began to take sips of water on the third day after the operation, and the next day, he ate a soft meal without any problems. He was discharged on the seventh day after the operation with a full recovery (Na, 141 mmol/L; K, 3.4 mmol/L; BUN, 20 mg/dL; Cr, 1.0 mg/dL). During a follow-up at 9 months, a positron emission tomography scan and an abdominal CT scan were checked. No evidence of abnormal lesion remained that suggested recurrence or distant metastasis. The last serum carcinoembryonal antigen and carbohydrate antigen 19-9 levels were 0.5 ng/mL and 5.6 U/mL. His family history did not have any specific details but we urged other family members to receive endoscopy due to the early onset of his symptoms.

**DISCUSSION**

Villous adenomas of the colon that cause secretory diarrhea were first described by McKittrick and Wheelock in 1954. It is a very rare but known complication of villous adenoma, which was described for the first time 50 years ago. Reaching the right diagnosis is not always as easy as in this case, and a complete colonoscopy is always mandatory. Therefore, in the case of a triad of prerenal failure, electrolyte disorder, and chronic diarrhea, the existence of an intestinal adenoma should always be considered.

About 3% of villous adenomas have secretory activity [4], and secretory villous adenomas associated with depletion syndrome are large, ranging from 7 to 18 cm at their greatest dimension. They are situated primarily in the rectum and occasionally in the sigmoid colon, as in the present case [1,4,5]. The large size allows more surface area for secretion, and the distal location of these lesions means a minimal area of normal colonic mucosa remains to allow fluid absorption, resulting in depletion syndrome [6]. It is unclear whether secretory villous adenomas occur in the proximal colon without depletion syndrome. It is certainly possible since goblet cells are equally distributed throughout the colon [1].

In a review of the literature on possible mechanisms of fluid and electrolyte imbalances, the electrolyte composition of mucin secreted by abnormal cells within the causative lesion is of interest. Whereas a normal bowel absorbs sodium and water and secretes potassium, segments of intestine affected by villous adenoma have been found to secrete water, sodium, and potassium. The absorptive capacity is largely unchanged. In both cases, the net movement of water is directly related to the net movement of sodium. No relationship between the net movement of water and potassium loss is known [6].

The cause of this abnormal secretory function has been postulated as secretagogue-mediated. Rectal effluent from a patient with villous adenoma of the rectum demonstrated prostaglandin E2 (PGE2) levels that were 3 to 6 times higher than normal [7]. Tissue from villous adenomas synthesizes more PGE2 than normal colonic mucosa. Mucosa adjacent to adenomatous polyps has been found to be unaffected by this, but adenoma-associated mucosa was demonstrated to synthesize larger amounts of PGE2 [6]. These secretagogues are active at sites containing the prostaglandin synthetic pathway. Nonreversible cyclooxygenase-inhibiting agents have been used to reduce PGE2 production and consequently the loss of sodium and water through the rectum. Cyclic nucleotides have also been implicated [3].

Fatal hyponatremia, hypokalemia, and dehydration due to adenocarcinoma are extremely rare [8]. However, the malignant potential of villous adenomas is well-recognized, with an incidence of carcinomatous change having been reported as high as 90% [2,9]. Therefore, surgical removal is the only satisfactory treatment both to stop fluid and electrolyte loss and to prevent carcinoma formation [4].

Endoscopic resection is presently considered the treatment of choice for adenoma. However, especially in McKittrick-Wheelock syndrome, it is not always effective owing to an unfavorable location or extension of the tumor. Moreover, large colonic polyps that are unresectable during colonoscopy are associated with a high rate of unsuspected cancer [10]. In a study by Pokala et al. [8], postoperative histopathology reports after laparoscopic
resection for endoscopically unresectable polyps revealed adenocarcinomas with an initial benign histology in up to 20% of cases [8]. Nusko et al. [9] reported a cancer risk for adenomas larger than 2.5 cm of the colon and rectum of 34% and 51%, respectively. In view of this high incidence of carcinoma in large polyps, a formal oncologic segmental colectomy is the most appropriate treatment of McKittrick-Wheelock syndrome, and laparoscopic approach could be a feasible option in such cases [10].

In conclusion, McKittrick-Wheelock syndrome should always be considered when severe hyponatremia and hypokalemia are accompanied by watery diarrhea. Early diagnosis is essential to commence the appropriate symptomatic therapy directed towards life-threatening electrolyte disturbances, and causal treatment can be undertaken. Definitive treatment of McKittrick-Wheelock syndrome is removal of the adenoma. Laparoscopic surgical resection is a feasible therapeutic modality for McKittrick-Wheelock syndrome.

CONFLICTS OF INTEREST

No potential conflict of interest relevant to this article was reported.

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