An uncommon cause of acute abdomen in an acromegalic patient: colonic volvulus

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Acromegaly is a chronic endocrine disease, typically caused by a pituitary adenoma leading to increased circulating GH levels and increased IGF-I secretion by peripheral tissues. Gastrointestinal diseases related acromegaly such as colon cancers, adenomatous polyps, and dolichocolon are much less known. We aimed to present a case of complete colonic volvulus in a patient with acromegaly. A 60-year-old male patient presented with the complaints of inability to pass gas or stool, worsening distension, and pain for one week. The clinical exam showed significant tenderness and distended abdomen as well as a typical appearance of acromegaly. In his past medical history, he underwent an operation of pituitary adenoma with the diagnosis of acromegaly. He also underwent a sigmoid colon resection due to dolichocolon detected by abdominal computerized tomography (CT) and colonoscopy. Upright abdominal X-ray was compatible with volvulus, so endoscopic distortion was tried but failed. At exploration, entire colon was extremely dilated and had a necrotic appearance, and observed to be twisted 360 degrees around its mesenteric axis. The patient underwent total colectomy and end ileostomy. Pathology result was reported as necrotic and hemorrhagic colon with 150 cm in length and 20 cm in width. The patient was discharged on the 14th postoperative day without complications. GH and IGF-1 have an irreversible effect on colonic collagen synthesis in acromegaly and has been proposed to be correlated with the presence of dolichocolon. Dolichocolon often can lead to an abnormal rotation, volvulus, and development of Chilaiditi syndrome. The occurrence of volvulus including the whole colon, although the pituitary adenoma was treated and partial colectomy was done, refers to predisposing factor being the irreversible effect of acromegaly on the colon.

KEY WORDS: Acromegaly, Emergency, Volvulus

Introduction

Colonic volvulus refers to torsion around the bowel's own mesentery. This usually occurs in a redundant colonic segment with an elongated mesentery [1,2]. Colonic volvulus is the third most common cause of colon obstruction after cancer and diverticulitis. It is responsible for 5% of all intestinal obstructions and 10-15% of large intestinal obstructions. It is usually seen in the sigmoid colon (80%), but may also occur in lesser numbers in the cecum (15%), transverse colon (3%) and splenic flexure (2%). Volvulus, if untreated, causes ischemia, gangrene, perforation, and death with reduced blood flow in the associated segment [4]. In treatment, if appropriate, colonoscopic decompression followed by simultaneous or elective surgery is performed. Cecal volvulus is not suitable for endoscopic decompression, and most cases are treated with surgery [5].

Acromegaly is a chronic endocrinopathy characterized by excessive GH production and exposure of peripheral tissues to IGF-1, which is usually caused by a pituitary adenoma. The incidence is 5 cases in a million per year, and the prevalence is 60 in a million [6]. Although acromegaly has many different findings, acromegaly-associated gastrointestinal findings such as colon cancers, adenomatous polyps, and dolichocolon are less acknowl-

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edged [7]. In this study, we aimed to present a massive volvulus case that developed suddenly in an acromegalic patient, required surgery, and hardly ever seen in the literature.

Case Report

A 60-year-old male patient referred to the emergency room with complaints of inability to defecate and pass gas, stomachache, and excessive swelling of the abdomen. He had abdominal pain and bloating for the last one year, but stated that complaints had been exacerbating for the past one week. In 2009, trans sphenoidal pituitary adenoma excision was performed with acromegaly diagnosis secondary to pituitary adenoma. Colonoscopy and abdominal computerized tomography (CT) performed in 2012 revealed dolichocolon, and due to complains, and a sigmoid colon resection was performed. Subsequent incisional herniorrhaphy was performed due to an incisional hernia which occurred after the sigmoid colon resection. Physical examination of the patient revealed a typical acromegalic appearance (Fig. 1), as well as marked tenderness and excessive distension in all quadrants in the abdomen. There was no gaita transmission on the rectal examination. Laboratory tests results were as following; WBC: 11600, neutrophil: 87.4%, amylase: 2419, and LDH: 489. On the other hand, the patient had a volvulus-compatible appearance on the direct standing abdomen graphy (DSAG) (Fig. 2), and an endoscopic detorsion was performed without success. The patient's general condition worsened following the colonoscopy, and he developed a respiratory arrest. The patient was taken into intensive care unit and after the appropriate resuscitation patient was taken to an emergency operation. At the explorations, entire colon was observed to be extremely dilated (> 10 cm) and had a necrotic appearance more prominent at the distal, and

Fig. 1: Acromegalic appearance characterized with growth and prolongation of the lower sieve, coarsening of the facial lines, enlargement and growth of the nose, ear, lips and forehead.

Fig. 2: Pre-operative direct abdominal x-ray shows dilatation of entire colon , being more evident in the sigmoid colon.

mesoaxial planar torsion was seen in all of the colon, from the caecum to the rectum (Fig. 3a). After the detorsion of the torsional colon (Fig. 3b), warm soaked laparotomy pads were applied to the affected colon, but the patient underwent total colectomy + endoostomy operation due to lack of proximal colon reperfusion. The patient who needed mechanical ventilation for three days after surgery was discharged on the 14th postoperative day despite the development of acute renal failure. Pathology result was reported as necrotic and hemorrhagic colon specimens due to ischemia or venous thrombosis reaching 20 cm in the widest area and 150 cm in length (Fig. 4). The ileostomy was closed with an ileorectal anastomosis three months after, and the patient is still being followed without any problems.

Discussion

Acromegaly is a chronic and slowly progressive disease caused by an increase in GH and IGF-1 levels due to pituitary adenoma and exposure of peripheral tissues to these hormones. Acromegalic patients have been shown to develop colon cancer, adenomatous polyp, dolichocolon, and diverticulitis [8-10].

In acromegaly, GH and/or IGF-1 have irreversible effects on collagen synthesis in the colon. It has been suggested that there is a relationship between long-term and severe exposure to elevated GH and IGF-1 levels and development of dolichocolon and diverticula [7]. GH causes changes in extracellular matrix composition by increasing the expression of matrix metalloproteinase gene. This suggests

Fig. 3: A) intraoperative view, B) detorsed ischemic colon prior to resection.

Fig. 4: Pathological appearance after resection.

that the abnormal extracellular matrix predisposes the development of diverticula and dolichocolon [11].

The dolichocolon, which is characterized by the extension of the sigmoid colon, is more common in the elderly population. It may cause abnormal rotation and pseudo obstruction like Volvulus and Chilaiditi syndrome [12,13]. History of prior abdominal surgery (volvulus/hysterectomy) or dolichocolon may be present in patients with cecum and/or transverse colon volvulus. In our case, even though there was a history of sigmoid resection for a dolichocolon, a volvulus containing the entire column above the old resection level was formed. Klein and colleagues examined the colons of 17 acromegalic patients with a barium enema. As a result, the column was often found to be wider and longer, which made performing and interpreting the barium study to be difficult [14]. In the literature review of our case, Klein's study and an acute colonic pseudo obstruction mimicking colonic volvulus in an acromegalic patient with dolichocolon were the only literature found, and also pseudo obstruction cases secondary to hypothyroidism were reported. This also reveals the fact that our case is a very rare phenomenon. The sigmoid colon is the most common site of colon volvulus and accounts for 60-75% of cases. In order of frequency after the sigmoid colon; cecum, transverse colon, and splenic flexure volvulus are observed [15]. Sigmoid volvulus is the third leading cause of colon obstruction in adults [16]. Sigmoid volvulus is mostly seen in older males. Although it is often considered to be idiopathic, the main predisposing factor is the presence of a redundant sigmoid colon with a narrow mesenteric junction [17]. Additionally, it can develop due to secondary causes such as high-fiber foods, certain neurological disorders, congenital megacolon and Chagas' disease [16].

Cecal and sigmoid volvuluses have different demographic characteristics. Sigmoid volvulus is seen more frequently in elderly, and cecal volvulus is more common in younger ages and women[5] . This indicates the connection between cecal volvulus and pregnancy. Because the gravid uterus causes displacement in the cecum, it elongates the mesentery and makes the torsion easier [18]. Cecal volvulus seen in older ages may also be associated with previous surgery. Surgery may lead to a mobile examination or may lead to postoperative adhesions in which the cecum can rotate [19,20]. In this case, it can be considered that the previous operation due to sigmoid dolichocolon may have created the basis for the formation of complete colonic volvulus or may cause cecal volvulus by partially changing the natural position of the colon in the abdomen. However, it is undeniable that long-term acromegaly is the primary factor in the development of complete colonic volvulus due to irreversible colon damage.

The average age is between 56 and 77 years, and almost one-third of all colonic emergencies in elderly patients are caused by sigmoid volvulus [21]. Compared to women, there is a tendency towards men and a ratio of 2.1: 1. This shows that the tendency of men in acute sigmoid volvulus is significant. The diagnosis of acute sigmoid volvulus is made in the presence of clinical and radiological findings. In the majority of patients, a complete physical examination and abdominal radiography are sufficient for diagnosis. The most common findings are tenderness and asymmetric abdominal distension [21]. Plain abdominal graphies are diagnostic in 57-90% of patients [16]. The classic sign is the sign of the coffee bean.

Abdominal CT frequently reveals dilated column segment, twisted column, and mesentery with air/fluid level [22] .
Initial management includes fluid resuscitation, nasogastric suction, and analgesia. Conservative management involves using a flatus tube and/or sigmoidoscope decompression in uncomplicated patients [17]. If these fail or evidence of gangrenous bowel is present, then emergency laparotomy is required [15]. Conservative treatment was initially tried in this patient, but sudden emergence of respiratory arrest suggests that an ischemic event is underlining and the patient was treated with emergency laparotomy following a rapid resuscitation.
Emergency surgery is associated with considerable mortality and morbidity. Kassi and colleagues found that mortality was 12% in Hartman procedures and the most frequent complication is surgical site infection which occurs in 42.86% of the patients and intestinal continuity is achieved in 50% of the patients [23]. Bhatnagar and his friends state that; Age greater than 60, shock at the time of admission, and previous exposure to volvulus are risk factors associated with mortality. [24]. Given the risk factors mentioned above, maximum effort should be provided by the intensive care staff to closely monitor homeostatic disturbance to reduce mortality in elder patients presenting with shock findings. The presentation, diagnosis, treatment modalities and surgical approaches to different cases of colon volvulus are similar regarding localization in the alimentary system. The lack of mortality in our case can be attributed to the appropriate surgical technique as well as to the early stage of surgery and intensive care support due to the patient's respiratory arrest.
In conclusion, the hypophyseal adenoma treatment could not prevent the development of dolichocolon in the patient, suggesting that the effects of long-term GH elevation are irreversible. The occurrence of volvulus including the whole colon, although the pituitary adenoma was treated and partial colectomy was done, refers to predisposing factor being the irreversible effect of acromegaly on the colon. However, contributions of the prior surgery cannot be ruled out. Therefore, although it is rare, recurrent volvulus should be considered in acromegalic and elderly male patients presenting with obstruction findings.

Riassunto

L'acromegalia è una malattia endocrina cronica, tipicamente causata da un adenoma pituitario che porta ad un aumento dei livelli circolanti di GH e aumento della secrezione di IGF-I da parte dei tessuti periferici. Molto meno conosciute sono le acromegalia correlate a malattie gastrointestinali come tumori del colon, polipi adenomatosi e dolocolocolon.
Abbiamo mirato a presentare un caso di volvolo del colon completo in un paziente con acromegalia. Un paziente maschio di 60 anni presentava difficoltà del transito di gas o feci, con peggioramento della distensione e dolore per una settimana. L'esame clinico ha mostrato dolorabilità significativa e addome dilatato nonché un aspetto tipico dell'acromegalia. Nell'anamnesi patologica remota figurava l'asportazione di un adenoma pituitario con la diagnosi di acromegalia, ed una resezione del colon sigmoideo su diagnosi di dolocolocollo rilevato dalla tomografia computerizzata addominale (TC) e dalla colonscopia.
La radiografia addominale in ortostatismo si presentava compatibile con una diagnosi di volvolo, ma era fallito il tentativo endoscopico di detorsione.
Durante l'esplorazione chirurgica, l'intero colon si presentava estremamente dilatato e aveva un aspetto necrotico e ruotato di 360 gradi attorno al suo asse mesenterico.
Si è preceduto a colectomia totale e ileostomia finale. Il referto anatomo-patologico ha indicato colon necrotico ed emorragico con 150 cm di lunghezza e 20 cm di larghezza.
Il paziente è stato dimesso il 14 ° giorno postoperatorio senza complicazioni.
GH e IGF-1 hanno un effetto irreversibile sulla sintesi del collagene del colon in acromegalia ed è stato proposto di essere correlato con la presenza di dolocolocolon.
Il dolocolocollo spesso può portare ad una rotazione anormale, al volvolo e allo sviluppo della sindrome di Chilaiditi. La presenza di volvolo incluso l'intero colon, sebbene l'adenoma ipofisario sia stato trattato e la colectomia parziale sia stata eseguita, si riferisce al fattore predisponente che è l'effetto irreversibile dell'acromegalia sul colon.

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ERRATA-CORRIGE

Nel lavoro "*Percutaneous endoscopic gastronomy (PEG) in elderly patients with dementia and anorexia. Medical and ethical issues regarding placement"* di Sivero et al, pubblicato sul n. 4/2018 pagg 305-308 è stato inserito in modo errato il cognome di uno degli Autori che deve essere corretto in: GIOVANNI CESTARO invece di Giovanni Cestari.