A rare case of hematemesis following gastro-duodenal strongyloides infection

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Abstract

We report the case of a 30-year old Black African man with a two-year history of nausea, abdominal discomfort and pruritus due to infection with Strongyloides stercoralis, which was successfully treated, but then complicated by the development of a bleeding pseudo-tumor in the duodenum. A review of the literature was performed. (Acta gastroenterol. belg., **2014**, 77, **383-385**).

Key words : pyogenic granuloma, upper GI bleeding, strongyloides infection, abdominal discomfort, lower GI tract.

Introduction

Strongyloides stercoralis (SS) is a parasite belonging to the helminth group, and infects humans, dogs, cats and primates. *Strongyloides stercoralis* is endemic in tropical and subtropical regions and occurs sporadically in temperate regions (1). It causes localized autoinfection in immunocompetent individuals but may cause disseminated disease in immunocompromised patients. *Strongyloides stercoralis* infection may be acute or chronic (1-3).

Pyogenic granuloma (PG) is a common, benign tumor that often presents as a rapidly growing, bleeding lump on the skin or in the oral cavity. It is most often seen on the head, the neck, the upper trunk, and the hands and feet (4). Less than 45 cases have been described in the gastrointestinal (GI) tract, mainly in the colon, esophagus, stomach and, rarely, the small intestine (4-6). Many of these cases have been from Asia, but this observation may be related to a publication bias. This is the first report, to our knowledge, which describes a duodenal PG in a Black African following gastro-duodenal SS infection.

Case report

A 30-year old Black African man consulted with a 2-year history of recurrent episodes of upper abdominal discomfort, nausea or vomiting, heartburn, and generalized itching. He had a history of malaria several years earlier. He had not taken any medication for two years and had stopped mild alcohol consumption six months earlier. He did not smoke. Physical examination was normal except for the presence of either urticarial or small scar lesions on the trunk, the back and the tops of his legs.

Blood tests revealed slight eosinophilia without any inflammation (Table 1). Liver function tests were normal. Examination of three stool samples showed no parasite larvae or pathogens. Direct examination and culture of the squamous skin lesions were negative. Upper abdominal ultrasonography was normal.

An upper gastrointestinal endoscopy (upper GIE) was performed and gastric biopsies were taken. The endoscopy showed atrophic gastropathy and inflammatory duodenopathy characterized by erythema, edema, and mild villus blunting. Pathological examination of an antrum specimen showed severe chronic gastritis. There were no *Helicobacter pylori* on immunohistological staining. Pathological examination of a duodenal biopsy specimen showed atrophic duodenal mucosa and infiltration of the mucosa by a mixture of mononuclear cells, polymorphonuclear neutrophils and eosinophils. The glands were hypocrine. Some glands in the antrum (Fig. 1A) and duodenum contained adult worms. ELISA serology and a second series of stool samples were strongly positive for SS. Serology tests for malaria, human T-cell leukemia virus type 1 (HTLV-1), and human immunodeficiency virus (HIV) were negative. A diagnosis of chronic strongyloides autoinfection was made.

The patient was treated orally with a single dose of 200 μg/kg ivermectin repeated two weeks later. His condition improved and he remained healthy for a 2-week period, but then presented with a 1-month history of episodes of early morning nausea and black-stained vomiting (visit 3). He was not taking any medication and clinical examination was normal. Ferritin level and mean corpuscular volume had decreased to the lower normal ranges (Table 1). A second upper GIE was performed, which showed normal mucosa of the esophago-gastric junction and the stomach, and a solitary pedunculated, bright red pseudo-tumor, 10 mm in size, on the flexure between the first and second parts of the duodenum. This lesion was endoscopically resected with a polypectomy snare. Pathological examination of the resected specimen (Fig. 1B) showed a lobular architecture with vascular and capillary proliferation and edema of the stroma, accompanied by acute and chronic severe inflammatory infiltrates with many eosinophils, features characteristic of a

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Table 1. — **Hematological and biochemical tests**

Variable	Reference Range	Visit 1	Visit 3	6 months
Hematology				
Hemoglobin (g/dL)	13-18	13.1	13.3	14.8
Hematocrit (%)	40-53	40.2	42.4	46.8
Mean corpuscular volume (μm^3)	80-100	85	79	84
White blood cell count (cells/L)	3.5-11 × 10^9	4.5	3.57	4.39
Eosinophils (%)	< 5	10.7	10	3.5
Eosinophils (cells/L)	< 400	480	360	150
Biochemistry				
Ferritin (ng/mL)	15-300	44	21.3	49
C- reactive protein (mg/L)	0.1-10	< 1	7.6	< 1
Urea (mg/dL)	< 45	23	–	26
Creatinine (mg/dL)	< 1.2	0.9	–	0.9
Sodium (mEq/L)	135-145	134	–	141
Potassium (mEq/L)	3.4-4.5	–	3.4	4.3
Chloride (mEq/L)	96-106	101	–	107
Bicarbonate (mEq/L)	22-28	30	–	25

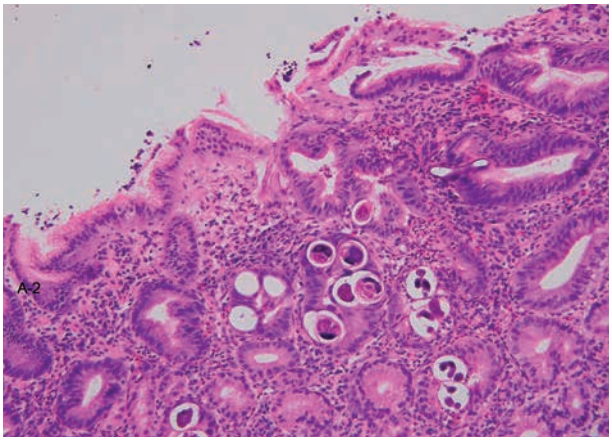

Fig. 1A. — Gastric (antrum) mucosa with chronic severe infiltration of the submucosa by inflammatory cells and eosinophils. Parasites in some gland lumens (HE stain ×100).

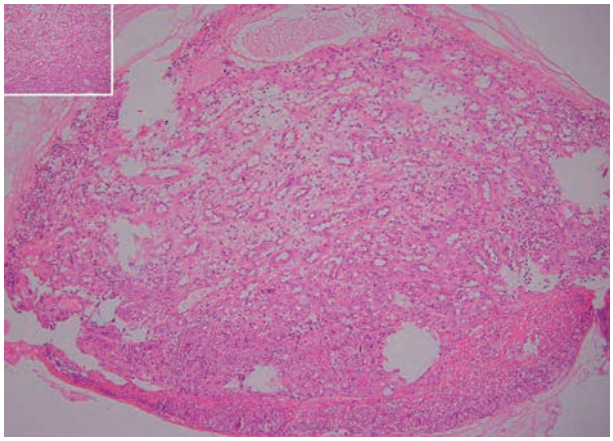

Fig. 1B. — Resected specimen of the duodenum (HE stain ×400) : vascular and capillary proliferation, edema of the stroma accompanied by acute and chronic severe inflammatory infiltrates with many eosinophils, characteristic of pyogenic granuloma. Inset : HE stain ×100.

PG. No malignant stigmata were observed, including absence of cytokeratin AE1 and AE3, and human herpes virus 8. There was no evidence of parasite larvae in the gastro-duodenal biopsies. During follow up at 3 and 6 months the patient remained asymptomatic and healthy. Control examinations (stool, anti-SS titer, and upper GIE with gastric and duodenal biopsies) were consistently normal.

The most likely diagnosis was, therefore, PG of the duodenum following resolution of chronic gastro-duodenal SS infection.

Discussion

Chronic SS infection is, in the majority of people, either asymptomatic or presents with mild, non-specific symptoms. This condition is also called strongyloides autoinfection because continuous re-infection occurs with low volumes of parasites. In contrast, disseminated strongyloidiasis is a catastrophic event associated with decreased immunity, which, if untreated, invariably results in death (hyperinfection). In this condition, the volume of parasites is very high and parasites may leave

the usual route (intestine → venous circulation → lungs → trachea → intestine) and can even cause meningitis (1-3,7).

Manifestations in "benign" autoinfection are mainly GI (i.e., nausea, vomiting) and dermatologic (i.e., urticaria, pruritus) ; these were both present in our patient. Risk factors for dissemination, notably immunodepression, were not present, and our patient thus had benign autoinfection and not hyperinfection (1-3).

The diagnosis of SS is based on the presence of larvae in stool or biopsies from the upper GI tract and/or serology. In 25% of patients, stool examination may be negative; because the larvae are excreted intermittently and chronic autoinfection is associated with a low parasitic output. Endoscopic duodenal features are non-specific, and include edema, discoloration of the mucosa, and megaduodenum (8). Gastric involvement has been described rarely.

The treatment of choice in uncomplicated cases is ivermectin, single doses of 200 μg/kg administered two weeks apart. In the case of treatment failure in immunocompromised patients, the disease can progress to hyperinfection, which carries a worse prognosis (10% mortality) (7,9). During follow-up, stool examination and blood tests (eosinophilia and serologic antibody titer) should be performed (1) ; endoscopy should be conducted only in selected cases.

The pathogenesis and etiology of PG is not well understood. It has been suggested that PG could be a hyperplastic, neovascular response to an angiogenic stimulus with imbalance of promoters and inhibitors (10,11). Trauma may have a trigger role and PG can develop after a wound to the mucosa, when there is a discrepancy between the time taken for the epithelium to heal (which is slowed) and that **for** the connective tissue, so that granulation tissue becomes excessive and bulges from the wound (12,13). Our patient had not been exposed to direct gastro-duodenal trauma, although the recurrent vomiting could be considered a possible mechanism. The biopsies could not have been involved because they were taken distally from the 2nd part of the duodenum. Other causes that have been described in the literature include hormonal influences (pregnancy) and medications (cyclosporine, capecitabine, etc), which were not relevant in this case. The role of infection is controversial, although PG has been reported following recovery from terminal ileum campylobacter infection and after recovery from erosive esophagitis (4-6,11,14-15).

Clinically, patients with a GI tract PG can be asymptomatic or symptomatic, depending on the GI tract segment involved, with symptoms ranging from dysphagia to overt hematemesis or bile duct obstruction (4-6).

At endoscopy, PG is seen as a small red, oozing and bleeding nodule, which often grows rapidly over a period of a few weeks as we observed in our patient. The endoscopic differential diagnoses for PG are Kaposi's sarcoma (KS), bacillary angiomatosis, non-Hodgkin's lymphoma (NHL) or cancer metastasis. All these conditions can present as a single red, flat, sessile, or polypoid lesion. Although KS and bacillary angiomatosis are often acquired immunodeficiency syndrome (AIDS)-related, all of these lesions can also be encountered in patients who do not have AIDS (4). Pathological examination is essential to differentiate the above diagnoses.

A GI tract PG can be completely resected by polypectomy snare because the tumor develops from the mucosa or the submucosa. Other therapeutic modalities include argon plasma coagulation, laser, embolization, or surgical resection, and treatment of skin lesions involves excision or laser. GI tract PGs seem to have a benign course, without recurrence, as observed in our patient at 6-month follow-up. In contrast, skin PGs are characterized by a tendency to recur. Malignant transformation has not yet been described (4-6,14).

In conclusion, SS auto-infection can cause mild, chronic complaints and can involve the stomach as well as the duodenum. The major concern with GI tract PGs is that they may be a rare cause of acute GI bleeding, and of occult bleeding, leading eventually to iron deficiency and microcytic anemia.

References

1. SEGARRA-NEWNHAM M. Manifestations, diagnosis, and treatment of strongyloides stercoralis infection. *Ann. Pharmacother*., 2007, **41** : 1992-2001.
2. KEISER P.B., NUTMAN T.B. Strongyloides stercoralis in the immunocompromised population. *Clin. Microbiol. Rev*., 2004, **17** : 208-217.
3. THOMPSON B.F., FRY L.C., WELLS C.D., OLMOS M., LEE D.H., LAZENBY A.J. *et al*. The spectrum of GI strongyloidiasis : an endoscopic-pathologic study. *Gastrointest. Endosc*., 2004, **509** : 90910.
4. VAN EEDEN S., OFFERHAUS G.J., MORSINK F.H., VAN REES B.P., BUSCH O.R., VAN NOESEL C.J. Pyogenic granuloma : an unrecognized cause of gastrointestinal bleeding. *Virchows Archiv*., 2004, **444** : 590-593.
5. KUSAKABE A., KATO H., HAYASHI K., IGAMI T., HASEGAWA H., TSUZUKI T. *et al*. Pyogenic granuloma of the stomach successfully treated by endoscopic resection after transarterial embolization of the feeding artery. *J. Gastroenterol*., 2005, **40** : 530-535.
6. SEOUNG H.G., KIM G.H., SONG G.A. *et al*. Esophageal pyogenic granuloma : endosonographic findings and endoscopic treatments. *Clin. Endosc*., 2013, **46** : 81-84.
7. VANDEBOSCH S., MANA F., GOOSSENS A., URBAIN D. Strongyloides stercoralis infection associated with repetitive bacterial meningitis and SIADH : a case eport. *Acta Gastroenterol. Belg*., 2008, **71** : 413-417.
8. KIM J., JOO H.S., KIM D.H., LIM H., KANG Y.H., KIM M.S. A case of gastric strongyloidiasis in a Korean patient. *Korean J. Parasitol*., 2003, **41** : 63-67.
9. CARVALHO B.S., LOUIS T.A., IRIZARRY R.A. Quantifying uncertainty in genotype calls. *Bioinformatics*, 2010, **26** : 242-249.
10. GIBLIN A.V., CLOVER A.J., ATHANASSOPOULOS A., BUDNY P.G. Pyogenic granuloma-the quest for optimum treatment : audit of treatment of 408 cases. *J. Plast. Reconstr. Aesthet. Surg*., 2007, **60** : 1030.
11. PIGUET V., BORRADORI L. Pyogenic granuloma-like lesions during capecitabine therapy. *Br. J. Dermatol*., 2002, **147** : 1270.
12. PIRACCINI B.M., BELLAVISTA S., MISCIALI C., TOSTI A., DE BERKER D., RICHERT B. Periungual and subungual pyogenic granuloma. *Br. J. Dermatol*., 2010, **163** : 941.
13. PAGLIAI K.A., COHEN B.A. Pyogenic granuloma in children. *Pediatr. Dermatol*., 2004, **21** : 10.
14. MEUWISSEN S.G., WILLIG A.P., HAUSMAN R., STARINK T.M., MATHUS-VLIEGEN E.M. Multiple angiomatous proliferations of ileal stoma following Campylobacter enteritis. Effect of laser photocoagulation. *Dig. Dis. Sci*., 1986, **31** : 327-332.
15. TAJIKA M., NAKAMURA T., KAWAI H., SAWAKI A., MIZUNO N., TAKAHASHI K. *et al*. Short-term development of esophageal pyogenic granuloma observed on endoscopy. *Gastrointest. Endosc*., 2006, **64** : 269-270.