Sebaceous glands in the esophagus are very rare and have been reported mostly in autopsy studies. They have been considered to be of no clinical significance. We report a 50-year-old man with gastroesophageal reflux disease who had sebaceous glands in the esophagus on endoscopic biopsy. [Indian J Gastroenterol 2007;26:36]

Sebaceous glands arise in close association with hair follicles to form the pilosebaceous apparatus. Ectopic sebaceous glands have been reported in a variety of sites, including the genitals, eyes, orbits, nipples, palms, soles and parotid glands. Several hypotheses have been postulated to explain heterotopic sebaceous glands, including developmental defect and metaplasia. The absence of sebaceous glands in the esophagus of children and their presence in adults could be indicative of a metaplastic process.

A 50-year-old obese white man on treatment for hypertension, hypercholesterolemia, asthma, depression and colonic polyps complained of two-month history of epigastric burning and acid regurgitation. Results of physical examination and routine laboratory tests were unremarkable. Endoscopy revealed reflux esophagitis and hiatal hernia along with multiple nodules scattered throughout the esophagus. Biopsy revealed fragments of squamous esophageal mucosa showing sebaceous gland formation (Fig). These glands consisted of units of large polyhedral cells with clear vacuolated cytoplasm. Each unit was delineated by a small basement membrane. A small collection of inflammatory cells was present in the vicinity of the sebaceous glands.

The patient was started on proton-pump inhibitor therapy and is doing well.

Sebaceous glands in the esophagus were first described at autopsy by Dela Pava and Pickren in 4 of 200 subjects. Ramakrishnan and Brinker described the first case in 1976. Approximately 30 cases have been reported since then. Some of these cases have been reported to have gastroesophageal reflux disease. Merino et al concluded that the sebaceous glands were heterotopic and incidental, with no relationship to patients' symptoms.

Esophageal sebaceous glands should be differentiated from submucosal tumors and mucosal proliferative lesions. Our patient also had inflammatory reaction around the heterotrophic sebaceous glands.

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Correspondence to: Dr Tak, Department of Internal Medicine, Brooks Memorial Hospital, 529 Central Avenue, Dunkirk, NY 14048, USA. E-mail: momintak@yahoo.com
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Recurrent limb weakness as initial presentation of Wilson’s disease
Partha Pratim Chakraborty, Sanjay Kumar Mandal, Dipanjan Bandyopadhyay,* Ramtanu Bandyopadhyay, Subhasis Roy Chowdhury

A 28-year-old normotensive euthyroid man presented with recurrent lower motor neuron type of weakness without sensory or autonomic involvement, with preserved reflexes. Systemic examination was significant for mild hepatosplenomegaly. Investigations revealed persistent hypokalemia, hyperchloremic, normal-anion-gap metabolic acidosis with deranged liver functions. Urine pH was 6.0 even after oral ammonium-chloride loading test. Type I renal tubular acidosis was diagnosed. A search for the etiology revealed bilateral Kayser-Fleischer ring, with low serum ceru-
loplasmin levels and high urinary copper, confirming it to be Wilson’s disease. [Indian J Gastroenterol 2007;26:36-38]

Renal affection is part of the clinical picture of Wilson’s disease (WD) and a frequent abnormality is distal renal tubular acidosis (DRTA). Patients with DRTA may show hypokalemic paralysis.

A 28-year-old man was admitted with weakness of all four limbs. It involved the lower limbs first and then progressed rapidly to involve the upper limbs, making him bed-bound the next day. The weakness was not associated with sensory abnormalities or diurnal variation. There was no history suggestive of alteration of higher functions, cranial nerve or sphincter involvement and of radicular pain or girdle sensation. Weakness was not preceded by vaccination, respiratory tract infection or diarrhea, and was not associated with food intake, exercise or change of temperature. He denied unexplained sweating, tremors, heat intolerance, prolonged vomiting, diabetes, hypertension, or any drug intake over a prolonged period of time.

The patient had two such similar attacks in the past, but those were milder, subsided spontaneously and did not require hospitalization. His personal and family history was non-contributory.

He had normal vitals. Neurological examination revealed marked hypotonia and weakness in all four limbs, the power being 1/5 in lower and 2/5 in the upper limbs. There was no cranial nerve involvement, sensory impairment and no abnormality in any of the superficial or deep reflexes. Plantar reflexes were bilaterally flexor. Both the lobes of liver were palpable 1.5 cm below the costal margin, with firm consistency. He had mild splenomegaly with no venous prominence or clinical evidence of free fluid in the abdomen.

Investigations: complete blood count was normal but fasting plasma glucose was 182 mg/dL and the post-challenge plasma glucose was 214 mg/dL. Liver function tests revealed: bilirubin 1.4 mg/dL, AST 68 U/L, ALT 36 U/L, proteins 6.4 g/dL, albumin 2.9 g/dL. Serum sodium, potassium, chloride and bicarbonate were 140 mEq/L, 2.5 U/L, proteins 6.4 g/dL, albumin 2.9 g/dL. Serum sodium, chloride and bicarbonate were 140 mEq/L, 2.5 U/L and 20.4 mEq/L, respectively. Arterial blood gas analysis suggested mild metabolic acidosis with normal anion gap.

A provisional diagnosis of hypokalemic paralysis was considered and the electrolyte deficit was corrected with oral potassium supplementation. The patient improved significantly but repeat estimation after 4 days showed persistent hypokalemia (2.7 mEq/L) and metabolic acidosis. Measured calcium (9.1 mg/dL), phosphate (3.8 mg/dL) and magnesium (1.9 mg/dL) were within normal limits.

Thyroid profile, HIV serology, creatine kinase, aldolase, porphobilinogen, cerebrospinal fluid study and electrophysiological studies were normal. 24-hour urinary potassium excretion was 242.76 mEq (69.36 mEq/L). Plasma and urinary osmolality were 296.76 mOsm/Kg and 482.2 mOsm/Kg, respectively. Estimated transtubular potassium gradient (TTKG) was 15.8.

To proceed with the diagnosis of a case of hypokalemia, we followed a standard algorithm.1 Potassium excretion >15 mmol/day and TTKG more than 4 suggested a diagnosis of RTA; oral NH₄Cl test (0.1 g/Kg) revealed worsening of acidosis (7.377 to 7.289) with urinary pH of 6.

Systemic examination revealed a faint corneal ring (which was missed previously) and slit-lamp biomicroscopy confirmed Kayer-Fleischer ring. Ultrasonography showed coarse hepatic echotexture, mild splenomegaly, and minimal ascites. Liver biopsy revealed early cirrhotic changes. Diagnosis of WD was confirmed with low serum ceruloplasmin level (13.67 mg/dL; normal 18-35) and high 24-hour urinary copper excretion (105 microgram; normal 20-50). The quantitative hepatic copper assay revealed 436 mcg per gram of dry weight of liver; normal 20-50).

Hypokalemia with paralysis is a potentially reversible medical emergency. It is primarily the result of enhanced shift of potassium into cells, decreased intake or excessive loss. The urine K⁺ excretion and evaluation of blood acid-base status could be helpful in diagnosis and management.2 Episodic weakness with onset after age 25 years is almost never due to periodic paralysis with the exception of thyrotoxic periodic paralysis,3 which was ruled out in this case by a normal thyroid profile. High urinary potassium in the presence of hypokalemia suggests the kidneys are the problem. If in doubt, measurement of TTKG can help.4 This is a semi-quantitative index of the activity of potassium secretion from the distal convoluted tubule and the cortical collecting duct. Hypokalemia with TTKG >4 suggests renal potassium loss due to increased distal potassium secretion.1

DRTA results from ineffective addition of hydrogen ions to the lumen of the distal nephron. The syndrome is manifested by hyperchloremic metabolic acidosis often associated with hypokalemia with normal serum anion gap.5 Absence of bicarbonaturia and worsening of systemic acidosis with urine pH above 5.5 following oral ammonium-chloride loading test confirm type 1 RTA.

Renal affection is part of the clinical picture of WD. A frequent affection is DRTA, more frequently in its latent form, with normal systemic pH but inability to reduce urinary pH below 5.5 after acid load. DRTA is frequent when WD is diagnosed late.6 RTA can give rise to hypokalemic paralysis.7 Recurrent hypokalemic paralysis due to RTA as a rare initial presentation of WD has been reported before.8

References
Double-barrel common bile duct: a rare cause of extrahepatic biliary obstruction


Department of Pediatric Surgery, Indira Gandhi Institute of Child Health, Bangalore; and Departments of *Radiology and **Gastroenterology, Bangalore Hospital, Bangalore

Double-barrel common bile duct is rare. We report a 50-year-old woman with defective canalization of the common bile duct, presenting with extrahepatic biliary obstruction due to stones in one compartment. CT scan highlighted this anomaly. After failed attempts at stone extraction at ERCP, she was successfully operated on. [Indian J Gastroenterol 2007;26:38-39]

Congenital anomalies of the extrahepatic biliary tree are commonly encountered at surgery. In fact the normal arrangement is seen in less than 50% of cases.1 A majority of abnormalities are related to alterations in the original budding from the foregut, or to failure of vacuolization of the bile duct diverticulum.2 Incomplete vacuolization may lead to sepatation of the biliary tree.

A fifty-year-old woman presented with history of epigastric pain, chills and rigor for four days. She had experienced similar symptoms two years earlier. She was diagnosed then to have cholelithiasis with dilatation of the common bile duct (CBD). She underwent ERCP with sphincterotomy and laparoscopic cholecystectomy. Recovery was uneventful.

At admission she was febrile, with minimal epigastric tenderness. Blood investigations revealed elevated leukocyte counts with normal liver function tests. Ultrasonography revealed post-cholecystectomy status with a 2-cm dilatation of the CBD. ERCP confirmed previous sphincterotomy. There was dilatation of the CBD with two filling defects. Dormia basketting was unsuccessful. A biliary stent was placed. Her symptoms improved and the patient was discharged. After three weeks, repeat ERCP showed the presence of bile duct stones. Since extraction was once again unsuccessful, she was referred to us for open choledocholithotomy.

On review of the original ERCP there was a questionable spiraling of the lower CBD with clear delineation of the hepatic ducts. Review of the previous CT scan revealed a clear partitioning of the CBD from the origin to its entry into the duodenum, in the coronal plane (Fig). This was indicative of double-barrel CBD.

Laparotomy revealed a dilated CBD. On transaction of the CBD at the upper border of the duodenum, two compartments were revealed. The stent placed earlier was found in the anterior compartment and the stones were identified in the posterior compartment. On cannulation of the anterior compartment the catheter selectively entered the left hepatic duct; the posterior compartment led to the right hepatic duct. Both compartments were found to communicate with each other at the hilum. The distal end of the CBD was closed, and the proximal CBD containing the two compartments was anastomosed to the first part of duodenum. The anastomosis was splinted. Postoperative recovery was uneventful. On review endoscopy after three months both compartments were well-visualized and freely draining bile.

Complete duplication of the CBD has been reported.3 These drain the two lobes of the liver through separate openings into the duodenum. The associa-
tion of common hepatic duct diaphragms and biliary obstruction has been reported. However anomalies related to incomplete vacuolization of the extrahepatic duct leading to compartmentalization has not been reported so far. Though the septation was congenital in nature, it manifested in adulthood with features of obstructive biliary pathology. CT outlined the two compartments and the septum from the hilum to the pancreas in the coronal plane.

In unrecognized cases of double-barrel CBD, exploration may be negative because the stone-bearing channels may be completely missed. Further, unrecognized injuries to one of the channels can lead to fistula or stricture formation. Establishing a pre-operative diagnosis may facilitate total laparoscopic correction, i.e., cholecystectomy with choledocho-duodenostomy.

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Correspondence to: Dr Shankar, Indira Gandhi Institute of Child Health, South Hospital Complex, Dharmanam College Post, Bangalore 560 029. Fax: (90) 2665 41799. E-mail: bpgshankar@gmail.com

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Adult idiopathic ileosigmoid intussusception prolapsing per rectum

Arun W S David, Edwin Stephen, Nihar R Pradhan, Sukria Nayak, Benajmin Perakath

General Surgery and Colorectal Surgery Unit, Christian Medical College, Vellore 632 004

Ileosigmoid intussusception involves complete telescoping of the bowel from the terminal ileum, cecum and intervening large bowel into the sigmoid colon, producing two sites of obstruction, one at the invaginated loop of terminal ileum and another at the sigmoidorectal intussusception. This can lead to ischemia and necrosis of either the ileum or sigmoid colon. There is one report of ileosigmoid intussusception presenting as a mass prolapsing per rectum in children.1

A 50-year-old gentleman presented with abdominal distension, constipation and a large mass descending per rectum for three days. This was preceded by diarrhea for four days. He gave history suggestive of recurrent episodes of subacute intestinal obstruction in the past. Physical examination revealed an emaciated man with a silent, nonrigid, distended abdomen. There was a dusky 10 cm x 10 cm mass prolapsing per rectum, with a wide stalk. It was possible to pass a finger all around the mass into the rectum. No point of invagination was felt (Fig).

Abdominal X-ray was unremarkable; he had elevated white cell count. At laparotomy he was found to have intussusception of the distal 60 cm of ileum, cecum, and ascending, transverse and descending colon, and proximal sigmoid. This was then descending per rectum and was gangrenous. He underwent subtotal colectomy with ileosigmoid anastomosis and diversion loop ileostomy.

Histological examination of the surgical specimen revealed gangrene of the ileocecal region with patchy gangrene of the colon and proximal sigmoid. No leading point or underlying abnormality was found.

Intussusception causing intestinal obstruction in adults is rare, comprising 1%-5% of all causes of intestinal obstruction.2,3,4 Only 5%-16% of all intussusceptions are seen in adults.5 In adults, an underlying cause is present in 90% of cases whereas in children a precipitating lesion is found in only 10% of patients.2,3,4

Fig: A 10 cm x 10 cm mass is seen prolapsing per rectum. It was possible to pass a finger all around the mass into the rectum. No point of invagination was felt

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Optimal treatment of adult intussusception is controversial. The debate centers on the issue of reduction versus mandatory en-bloc primary resection. Weilbacher and associates emphasized resection without reduction whenever possible. This was based on a high incidence of underlying malignancy that could not be confirmed either preoperatively or intraoperatively. The downside of reduction includes the theoretical risk of intraluminal seeding or venous embolization in regions of ulcerated mucosa. Other concerns with reduction are possible perforation during manipulation or subjecting the patient to increased risk of anastomotic complications in the setting of edematous or weakened bowel. The only exception may be in cases of sigmoid-rectal intussusception secondary to carcinoma when reduction may allow a sphincter-sparing procedure to be performed.

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Correspondence to: Dr David, Surgery Unit-5. Fax: (416) 223 2054. E-mail: sur5@cmcvellore.ac.in
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Esophageal stricture following electrical injury
Syed Ibrahim Hassan, M Uma Devi, P Shravan Kumar, B Ramesh Kumar, K Venugopal Reddy, K Nageshwar
Department of Gastroenterology, Osmania General Hospital, Hyderabad

Esophageal injury resulting from electrical shock is rare. Stricture of esophagus following external electrical injury has not been reported yet. We report a 24-year-old electric lineman who developed esophageal stricture following external electrical shock. He responded to dilatation with Savary-Gilliard dilators.

Though few cases of esophageal injury due to electrical shock have been reported, there is no reported case of esophageal stricture due to electrical shock.

A 24-year-old electric lineman presented with sudden onset of absolute dysphagia of 1-day duration, following grade I dysphagia of 6 months’ duration. Six months back, he had sustained an electric shock while working on an electric pole and lost consciousness for a few hours. He had undergone amputation of the right upper limb due to electrical injury. He also had superficial electrical burns on the right side of the neck and right shoulder. He was hospitalized for 1 week. He noticed dysphagia while in hospital. There was no history of heartburn, intake of corrosives, or nasogastric tube insertion.

Physical examination revealed amputated right upper limb up to mid forearm with two large scars, one over the left neck and the other surrounding the acromion process of the left shoulder. Systemic examination was normal.

Upper GI endoscopy (Fig) revealed multiple linear non-circumferential ring-like trabeculations from 20 cm, with stricture at 24 cm with food bolus impaction; mucosa showed no ulceration or friability. Food bolus was removed with snare and the patient underwent three weekly sessions of esophageal dilatation with Savary-Gilliard dilators and was able to take solid food.

Though the exact entrance and exit wounds are not known in this patient, it appears that the electrical current passed from the right hand with which he held the electrical wire through the thorax, leading to esophageal injury with stricture formation.

Only anecdotal cases of esophageal injury due to electrical accidents are reported. Filsak and Berman reported a suicide attempt by a patient who swallowed the cut end of an electrical cord that was then plugged in. The patient developed a stricture at the site of contact; it was unresponsive to dilatation and required surgical resection. Rubin et al studied a patient following external electrical shock. Barium swallow and upper GI endoscopy were essentially normal; motility studies revealed abnormal esoph-
ageal motility. The patient recovered over time.

In summary, this patient developed external electrical shock-induced esophageal stricture due to passage of current through the thorax; it responded to dilatation.

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Correspondence to: Dr. Uma Devi, Assistant Professor. E-mail: umadevimalladi_66@yahoo.co.in

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**Coexistence of chronic calcific pancreatitis and celiac disease**

Ajit Sood, Vandana Midha, Neena Sood, Manu Bansal, Manpreet Kaur, Anuza Goyal, Nidhi Sharma

Department of Medicine, Dayanand Medical College and Hospital, Ludhiana, Punjab

Coexistence of celiac disease with chronic calcific pancreatitis is rare. We describe a 26-year-old woman with chronic calcific pancreatitis in whom non-response to treatment was due to celiac disease. [Indian J Gastroenterol 2007;26:41-42]

Mild exocrine pancreatic insufficiency has been frequently observed in both untreated and treated patients with celiac disease.\(^1\)\(^2\) However, the coexistence of chronic calcific pancreatitis with celiac disease is unusual and may be just a coincidence, but in view of the known pathogenesis of pancreatic calcification with intestinal dysfunction, this association is of interest.

A 36-year-old woman was first admitted for recurrent abdominal pain of 2 years’ duration. The pain was dull in character, localized in the epigastrium with occasional radiation to the back and left flank. In addition, there was history of diarrhea and steatorrhea, poor appetite, and weight loss of 12 Kg over a period of 1 year. She was recently detected to have diabetes mellitus. Family history was noncontributory.

**Investigations:** normal complete blood count, ESR, renal and liver functions, amylase, lipase, calcium, triglycerides and cholesterol. Random blood sugar was 336 mg/dL. Chest X-ray was normal. X-ray of the abdomen revealed abundant scattered calcifications throughout the pancreas. CT scan also showed pancreatic ductal calculi and parenchymal calcification. D-xylose was normal (0.8 g/5 g/5 h) and fecal fat was elevated at 16 g/day (normal ≤6 g/day).

A diagnosis of chronic calcific pancreatitis was made and she was placed on pancreatic enzymes, H2 blockers, insulin therapy and vitamin supplements. Two months later her symptoms had improved, with considerable reduction of pain and steatorrhea and her weight had increased by 2 Kg. Her diabetic status was controlled.

A year later she again suffered from recurrent severe pain in abdomen and increased bowel movements. Her weight started decreasing. No symptomatic relief was achieved with proton pump inhibitors, insulin, enzyme supplements, antimitoty drugs and antibiotics. ERCP was performed six months later and an attempt was made to clear pancreatic duct calculi. However, the dormia basket got impacted in the pancreatic duct as the calculi were large. Whipple’s procedure was then carried out. She tolerated the surgery well and recovered gradually.

Her condition again started worsening after 3 months, and she was referred to our institution with further weight loss of 6.8 Kg and persistent diarrhea. On examination, she was wasted, with significant pallor and mild pedal edema. Her weight was 39.5 Kg with BMI 15.5 Kg/m\(^2\). Chest and cardiovascular examination was normal. Abdominal examination showed scar of previous surgery. *Investigations:* hemoglobin 7.8 g/dL, total leukocyte count 5.8 x 10\(^9\)/L; MCV 78 fl, microcytic hypochromic peripheral blood film, serum ferritin low, mildly elevated transaminases, serum albumin 2.8 g/dL and calcium 6.8 g/dL. Blood sugar and renal functions were normal. CA 19-9 was normal.

In view of persistent iron-deficiency anemia and diarrhoea a possibility of celiac disease was considered. Stool examination was normal. Barium meal follow-through showed mucosal edema, flocculations and dilution of contrast; colonoscopy was normal. *Anti-tissue transglutaminase* was high (104 U/L) and duodenal biopsy showed total villous atrophy, Marsh III. She was put on gluten-restricted diet. At 3 months her symptoms improved, with significant resolution of diarrhea and gain in weight.

On last follow up over 2 years later she was asymptomatic; her weight had increased to 60 Kg.

Although exocrine pancreatic insufficiency has been observed in patients with intestinal malabsorption,\(^1\) chronic calcific pancreatitis has only rarely been reported to coexist with celiac disease. Inversely, the morphologic structure of the small bowel is reported normal in about 68% of patients with pancreatic insufficiency secondary to chronic pancreatitis and carcinoma pancreas.\(^2\) Discrepancies in the morphology and pancreatic function are reported in 12%-29% in some other studies.\(^3\) The etiology of pancreatitis in patients with celiac disease is believed to be related to protein malnutrition, which apart from decreasing pancreatic enzymes also decreases pancreatic stone protein, thereby initiating the pathway to development of chronic pancreatitis.\(^4\) Additionally, abnormalities in hormonal function and papillary stenosis have been implicated in devel-
opment of pancreatitis in patients with celiac disease.\textsuperscript{1,2,3}

In the previous cases reported the two conditions were diagnosed concomitantly but in our case the diagnosis of celiac disease followed that of chronic pancreatitis, raising the possibility of coincidental occurrence.

References

Landry-Guillaine-Barré syndrome as presentation of celiac disease

Vandana Midha, Narinder Pal Jain, Ajit Sood, Rajinder Bansal, Sandeep Puri, Vipin Kumar

Department of Medicine, Dayanand Medical College and Hospital, Ludhiana, Punjab

Celiac disease has been associated with a variety of neurological illnesses, most frequently cerebellar ataxia and peripheral neuropathy. We report presentation as Landry-Guillaine-Barré syndrome in a 28-year-old woman with previously unsuspected celiac disease.\textit{[Indian J Gastroenterol} 2007;26:42-43]

Neurological disorders are estimated to occur in 6\% to 10\% of patients with celiac disease (CD). In fact, a high proportion of patients with neurological symptoms of unknown origin have been found to have gluten sensitivity.\textsuperscript{1} The neurological symptoms said to be associated include cerebellar ataxia, epilepsy, neuropathy, myelopathy, myopathy, dementia, headache, autism, depression and multifocal leukoencephalopathy.\textsuperscript{1,2} These are usually chronic and progressive.

A 28-year-old woman was admitted to the emergency department with complaints of gradually increasing weakness of all four limbs over 2 days. She expressed difficulty in breathing few hours prior to admission. There was no history of fever, nausea, vomiting, diarrhea, urinary complaints, or intake of any indigenous medications. There was no history of palpitations, tremulous, excessive perspiration or marked anxiety. She was married and had one child. Family history was non-contributory. She had visited a doctor about a year ago for evaluation of marked weakness, bodyache, off-and-on diarrhea and weight loss of 10 Kg in 6 months. She was found to have severe osteoporosis on DEXA bone mineral density analysis. However, no diagnosis was offered for this. Her medications in the past included occasional intake of ibuprofen, acetaminophen, iron and calcium supplements.

On physical examination, the patient exhibited clear and cognitive consciousness. Her BP was 120/74 mmHg, pulse rate 98/min, respiratory rate 18/min, and she was afebrile. General examination was unremarkable except for pallor. Thyroid gland was not palpable. Cardiopulmonary and abdominal examination was unremarkable. Neurological examination revealed symmetrical flaccid paralysis and areflexia of all four limbs. No fasciculation, myoclonus, or muscular atrophy was observed. Cranial nerves were functionally intact, as were sensation and proprioception. Visual acuity and visual field examination were normal.

Investigations: hemoglobin 8.5 g/dL, hematocrit 32.4, white cell count 8,260/µL, platelet count 186,000/µL, ESR was 76 mm in 1st hour, Na\textsuperscript+ 140 mmol/L, K\textsuperscript+ 1.9 mmol/L, Cl\textsuperscript- 95 mmol/L and HCO\textsubscript{3} 24.0 mmol/L. Serum liver and kidney function tests were normal. She had positive ANA but negative dsDNA, positive rheumatoid factor, normal complement levels and normal CRP. Urinalysis was normal. Blood gases were also normal. Thyroid profile was normal and CPK was 115 U/dL (normal 26-190). K\textsuperscript+ replacement was commenced at 15 mmol/h, but her respiratory effort and sensorium worsened and she had to be put on ventilatory support. Potassium levels rose to 5.4 mmol/L and then normalized at 4.4 mmol/L.

Non-contrast CT head revealed mild diffuse cerebral edema. CSF examination revealed 6 cells/mm\textsuperscript{3} (all lymphocytes), glucose 152 mg/dL, protein 67 mg/dL and globulin 1+. Nerve conduction revealed motor-sensory polyradiculopathy. She was given intravenous immunoglobulins 0.4 mg/Kg for 5 days. By the end of this therapy there was improvement in muscle power and she could raise her arms against gravity and also adduct her lower limbs. However, there was difficulty in weaning her off the ventilator. Her electrolytes including calcium and magnesium were normal.

Review of her history of diarrhea in the past, presence of anemia and severe osteoporosis at a young age prompted us to think of malabsorption state. Anti-tissue transglutaminase was high (48 U/dL). We initiated her on gluten-free diet and she started showing gradual recovery and could be tapered off the ventilator subsequently. She remained hospitalized for 35 days. She gained weight, hemoglobin improved and renal functions remained normal. Duodenal biopsy 7 days later showed total villous atrophy (Marsh IIIa).

Celiac disease is associated with numerous neu-
neurological manifestations, peripheral neuropathy and cerebellar ataxia being the most common. Evidence for peripheral neuropathy has been found in up to 47% of these patients. The most commonly reported peripheral neuropathy is the chronic distal, symmetrical, predominantly sensory-motor neuropathy; however pure motor neuropathy, mononeuritis multiplex, Guillaine-Barré-like syndrome and autonomic neuropathy have also been described. Our case is exceptional for unusual initial presentation of CD as Landry-Guillaine-Barré syndrome. Hypokalemic periodic paralysis or acquired hypokalemia is one of the many disorders that mimic this condition.

Neurological abnormalities were formerly thought to be sequelae to the gut disease, but it is now recognized that they may not only precede CD but can also be its only manifestation. Genetic factors and humoral response have been proposed in the pathogenesis of gluten neuropathy. Anti-ganglioside antibodies have been reported in 65% of patients with CD and neuropathy. Cultured T lymphocytes from a patient with acute demyelinating polyneuropathy showed \( \gamma/\delta \) subpopulation, which is considered a marker for latent celiac disease. The neuropathy in CD has shown variable response to gluten restriction.

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Correspondence to: Dr Ajit Sood, 6-E Tagore Nagar, Ludhiana 141 001. E-mail: ajitsood10@sify.com

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Massive recurrent hemorrhage due to duodenal diverticulum – role of tattooing and suture plication

D Bandyopadhyay, A R Haque,* A Mahomed

Department of Surgery, Calderdale Royal Hospital, and *Department of Surgery, Huddersfield Royal Infirmary, West Yorkshire, UK

We report a 79-year-old lady with massive recurrent GI bleeding from a duodenal diverticulum arising at the mesenteric border. We used tattooing during endoscopy as a method for identifying the bleeding source at surgery. The diverticulum was treated with suture plication. [Indian J Gastroenterol 2007;26:43-44]

Duodenal diverticulum is the most common small bowel diverticulum. Most of these are asymptomatic. Complications can cause significant morbidity.

A 79-year-old lady was admitted with two-day history of passage of dark blood per rectum. There was no other GI symptom. Her weight was steady. She was known to suffer from hypothyroidism, hypertension, angina and transient ischemic attacks.

She had a similar episode of rectal bleed about 7 months ago. On that occasion her hemoglobin was 5.6 g/dL with raised urea and normal creatinine. She was resuscitated with blood transfusion. Upper GI endoscopy was normal. Colonoscopy showed scattered diverticulae and altered blood throughout the colon without a bleeding source. Mesenteric angiogram showed possible extravasations from the first branch of the superior mesenteric artery. It was technically not possible to embolize the bleeding vessel. At laparotomy underrunning of a bleeding vessel in a duodenal diverticulum was performed. The diverticulum was at the mesenteric border at the junction of the 3rd and 4th parts of the duodenum.

During her current admission she was pale. Abdominal examination was unremarkable. Rectal examination revealed dark blood and clots. Hemoglobin was 8 g/dL; she was resuscitated and transfused blood. At upper GI endoscopy a bleeding duodenal diverticulum was seen. It was not possible to achieve hemostasis with endoscopic measures; the diverticulum was tattooed with India ink.

At laparotomy the duodeno-jejunal flexure was difficult to identify due to dense adhesions from previous surgery. Following extensive adhesiolysis, the tattooed area was seen and a small diverticulum was identified (Fig)
on the mesenteric border. The patient was not fit for major bowel resection and reconstruction. A longitudinal duodenotomy of the 3rd part of duodenum was performed. The little finger was inserted into the diverticulum. This acted as a guide to aid suture plication of the diverticulum from the apex to the base.

The postoperative period was uneventful and the patient was discharged 2 weeks following surgery. She remained well at eight months following operation.

The incidence of symptoms from duodenal diverticulum increases with the age of the patient. Analysis of a case series of 100 duodenal diverticula from Taiwan revealed that 7% presented with upper GI hemorrhage. A majority are in the second part of the duodenum, unlike in our patient. Massive recurrent bleed from duodenal diverticulum is not frequent.

Treatment options include endoscopic hemostasis, embolization and surgery. Endoscopic therapy is usually not effective as they tend to rebleed. Successful superselective arterial embolization has been described, but it is technically demanding. Surgery remains the definitive treatment in such cases.

At surgery identification of the bleeding point can be a major problem; especially for patients like ours who have undergone previous operation involving the same location. Tattooing with India ink of the bleeding point at the time of endoscopy was a crucial diagnostic aid in our patient.

Duodenal diverticulectomy is an effective procedure; however, it is associated with considerable leak rate. Diverticulectomy is a reasonably straightforward operation for a diverticulum originating from the anti-mesenteric border. The diverticulum in our patient involved the mesenteric margin. Isolation of the diverticulum at this site bears risk of devascularization of a segment of duodenum. In our opinion, major surgical resection in such a case in an elderly unstable patient with co-morbidity is a fruitless exercise. A more conservative approach such as suture obliteration of the diverticulum is safe and effective.

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Correspondence to: Dr Bandyopadhyay, 75 Thornhill Road, Steeton, West Yorkshire UK, BD20 6RE. Fax: (44) 1422 222296. E-mail: dbanerjee@blueyonder.co.uk
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