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CASE REPORT

Tongue and esophageal involvement in Henoch-Schonlein purpura: A case report

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Abstract:
Henoch-Schonlein purpura (HSP) is the most common form of systemic vasculitis that primarily affects children. It is characteristically involving skin, joints, gastrointestinal tract, kidneys, central nervous system and scrotum. Gastrointestinal involvement is occur in 51-58% of children and manifesting as abdominal pain, nausea, vomiting, diarrhea and gastrointestinal hemorrhage; but the involvement of esophagus is rarely reported. Here a case of HSP with involvement of tongue, esophagus and stomach is reported in an adolescent patient.

Keywords: Henoch-Schnlein, Gastrointestinal tract, Abdominal pain
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Introduction

Henoch-Schonlein purpura (HSP) is the most common form of systemic vasculitis of unknown etiology in children. It primary affects children less than 15 years of age, with mean age of 6 year. HSP can involve skin, gastrointestinal tract, joints, kidneys, central nervous system and scrotum. In general, HSP is a benign and self-limited disorder (1-2).

Gastrointestinal involvement in HSP is manifesting as abdominal pain, nausea, vomiting, diarrhea and gastrointestinal hemorrhage; but the involvement of esophagus is rarely reported (2-4).

Case Report

The patient is a 16 year old boy who present with epigastric abdominal pain, nausea and vomiting for six days duration. Due to progression of abdominal

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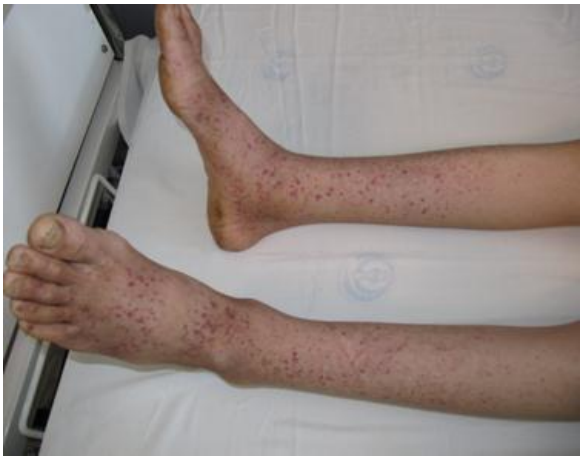

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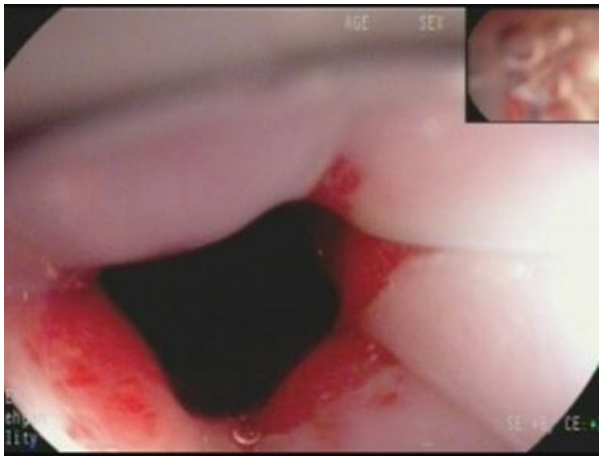

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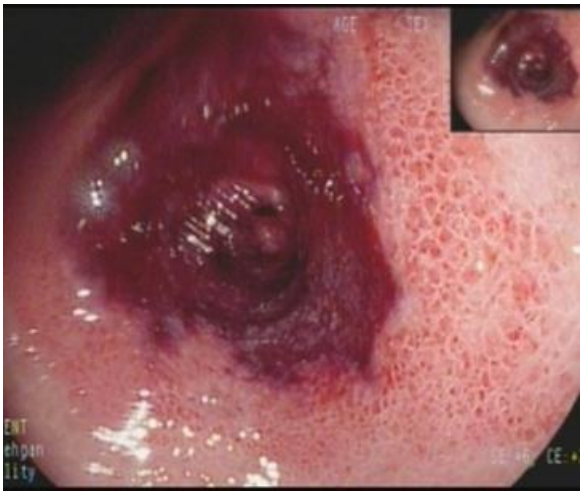

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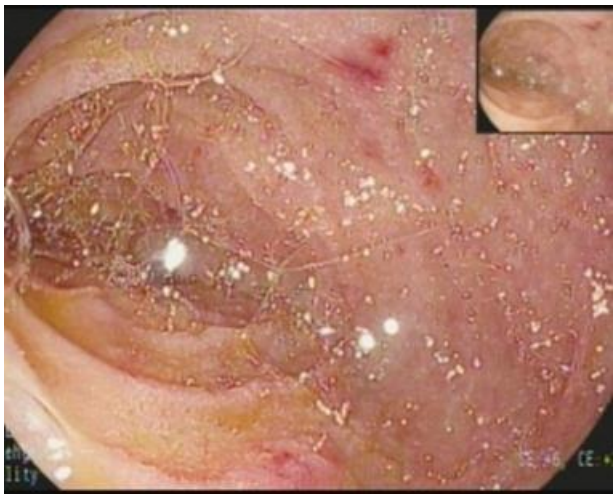

D

Figure I. A: Palpable purpura on both lower extremities of patient with Henoch Schonlein purpura; B: Severe hemorrhage and mucosal edema of lower esophagus; C: Intense hemorrhage and petechia on body and antrum of stomach; D: Petechia and mucosal edema on second part of duodenum

pain he was underwent appendectomy with impression of appendicitis. Two weeks later in addition to continue epigastric abdominal pain, nausea and vomiting; erupted maculopapular with some confluent and diffuse purpuric, hemorrhagic skin rash on both upper and lower extremities appeared. He referred to our hospital due to gastrointestinal hemorrhage that presented with coffee ground vomiting.

When he was admitted to our hospital, he had a normal temperature of 37°C, a pulse rate of 120/min regular and a blood pressure of 85/60 mmHg with orthostatic hypotension. Physical examination revealed pale conjunctiva, red tongue with purpuric lesions and abdominal tenderness at the epigastric and periumbilical region without rebound tenderness, but bowel sounds were normal.

There were also diffuse and confluent maculopapular and palpable purpuric skin lesions that involving both upper and lower extremities (Figure 1A). He had no evidence of central nervous system, joint or scrotal involvement.

Initial laboratory findings showed that the white blood cell count was 12900/mm3, platelet count was 595000/mm3, elevated erythrocyte sedimentation rate (ESR) of 36 mm/hour and C-reactive protein of 48 mg/dL, C3 was normal and C4 was slightly elevated, ANA and dsDNA were not significant. Serum lipase was normal but serum amylase was slightly elevated.

His urine analysis showed massive hemoglubinuria. Esophagogastroduodenoscopy revealed multiple petechia and hemorrhagic lesions and ulcers in lower third of esophagus, mucosal hemorrhage and congestion of antrum and prepyloric region and multiple petechia and purpuric lesions in bulb and second part of duodenum (Figure 1B-D).

Biopsies revealed a leukocytoclastic vasculitis in the skin.

He was treated with intravenous fluid and kept nothing by mouth for 2 days. He was started on methylprednisolon 20mg/kg/day and pantoprazole 1mg/kg/day. After 3 days, intravenous methylprednisolon changed to oral prednisolone 1mg/kg daily, with resolution of his symptoms within six days. The purpura disappeared gradually and was completely resolved three weeks later. There was not recurrence of disease during 6 months follow up.

Discussion

In susceptible individuals, several factors have been implicated as triggers of HSP such as; β-hemolytic streptococci, mycoplasma, yersinia, helicobacter pylori, hepatitis A and B viruses or medications including penicillin and ciprofloxacin, vaccination, foods, insect bites and neoplastic disorder (5-10).

Gastrointestinal manifestations of HSP are common that occur in 51-58% of children. They usually result from edema and ulceration of the bowel wall, leading to nausea, vomiting, abdominal pain and hemorrhage.

Gastrointestinal symptoms may also precede the skin rash in some cases. Gastrointestinal bleeding, either overt melena, hematochezia or hematemesis or with positive stool gauiac test occurs in 18% of children (5,11,12). Massive gastrointestinal bleeding has been reported, but is uncommon (12).

Sakagami et al, summarized gastrointestinal lesions in 32 Japanese patients with HSP and reported that the positive finding of purpuric lesions, erosions or ulcers were observed in 59% of patients in the stomach, 78% in the duodenum, 100% in the small intestine, and 88% in the colon. However, only one patient had esophageal ulcer, and extensive search of published reports revealed only 5 cases of HSP with esophageal involvement. One patient was complicated by esophageal perforation that required surgery (3).

Our patient had esophageal (mucosal erythema, petechia, ulcer and hemorrhage), gastric (intense mucosal edema, hemorrhage, erythema) and duodenal (mucosal hemorrhage and ulcer) involvement that presented with gastrointestinal bleeding during the course of HSP. Histologic evaluation of esophageal ulcer demonstrated nonspecific inflammatory changes. These findings appeared and resolved parallel with the other clinical manifestations, such as purpura of extremities. We think the esophageal involvements in the HSP may be due to underlying vasculitis, but the real pathophysiology of the esophageal involvement during course of HSP is unknown (13). In our patient gastrointestinal symptoms and endoscopic findings resolved after treatment with corticosteroid and acid suppressant medication.

Conclusion

Although esophageal involvement may rare, it is important to be aware that this can occur during the course of HSP.

In patients who don't visible purpura but compliant of acute onset of abdominal pain, HSP should be considered in the differential diagnosis and upper endoscopic examination may demonstrate raised erythematous mucosal lesion, diffuse patchy redness , multiple ulcer or erosive / ulcerative lesions, mainly in the stomach and duodenum.

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