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Case Report

A Rare Cause of Intestinal Obstruction

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Abstract: Angio neuronal dysplasia is relatively rare condition of stomach and intestine which is combination of two conditions angio and neuronal dysplastic changes followed by abdominal pain and intestinal obstruction. The current case presented sub acute abdominal obstruction. We present a case of angio- neuronal dysplasia of terminal ileum and cecum with an unusual mechanism of intestinal obstruction.

Key Words: Angio dysplasia, Neuronal dysplasia, GI Tract.

Introduction

Intestinal pseudo-obstruction is a rare motility disorder affecting segmental or entire gastrointestinal tract. It gives symptoms and clinical signs of mechanical intestinal obstruction despite negative results of all imaging disorders. The disease may occur in acute or chronic form [1]. The typical manifestation is characterized by recurrent episodes of abdominal pain, abdominal distension, with or without vomiting and altered bowel habits. Diarrhea often occurs as a consequence to small bowel bacterial overgrowth [2].

Intestinal neuronal dysplasia (IND) is a complex alteration of the enteric nervous system (ENS), mostly involving rectum and colon, or the whole intestine. This disorder is a frequent cause of intestinal dysmotility and pseudo-obstruction [3].

Intestinal neuronal dysplasia (IND), an abnormality of intestinal innervations associated with pseudo-obstructive dysmotility, was described in 1971 by Meier- Ruge [4].

Gastrointestinal angiodysplasia is described as gastrointestinal mucosal ectasias. Mostly are located in the cecum and ascending colon. Fifteen percent are located in the jejunum and ileum [5].

Case Report: A 12 years old female patient presented with history of abdominal pain on &off from last 2 years, exacerbated for last 2 months. Pain was colicky, non radiating in nature, and not associated with any aggravating or relieving factor. She was initially treated by general practitioners but her condition did not improve. Her past and family history was not significant.

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On examination she was a pale looking child and on abdominal palpation, in lower right quadrant there was 5 cm mobile mass, round in shape, firm in consistency and non tender. Sluggish bowel sounds were audible. Her baseline investigations were normal and hepatitis profile was negative for both hepatitis B & C. Urine R/E was normal.

The ultrasound abdomen showed multiple dilated loops in the abdomen showing to and fro movements. Finding was suggestive of sub-acute intestinal obstruction. X-Ray abdomen showed multiple air fluid levels. CT scan showed markedly distended small bowel filled with fluid and air, collapsed loops of colon and thickened bowel wall.

Gross Examination: An exploratory, laprotomy was performed. A mass measuring 7cm x 5 cm was found at terminal ileum and cecum. It showed no obvious ulcer or tumor. Mass was resected with clear margins (Fig 1, 2).

Microscopic Examination: The sections revealed essentially normal ileal mucosa. The muscularis mucosa was essentially normal. The submucosa contained frequent prominent blood vessels. Meisner's plexuses were not well developed. There were rather prominent Auerbach's intermuscular plexuses but contained mostly immature neuronal cells. Those cells were quite increased in numbers. The serosa contained large, malformed vessels and many scattered thickened nerves along with significant fibrosis (adhesions). There were also quite hyperplastic serosal lymphoid follicles with prominent germinal centres. Final diagnosis of focal angioneuronal dysplasia with fibrosis along with reactively hyperplastic serosal lymph nodes was made. (Fig 3, 4 &5).

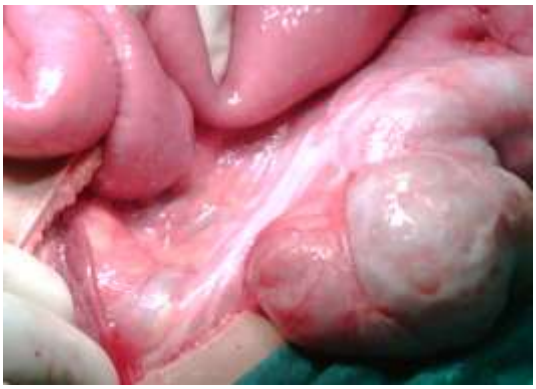

Fig.1

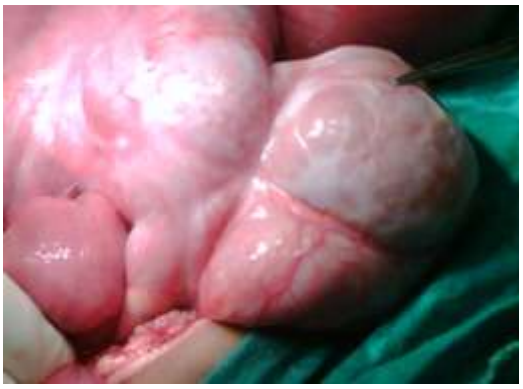

Fig.2

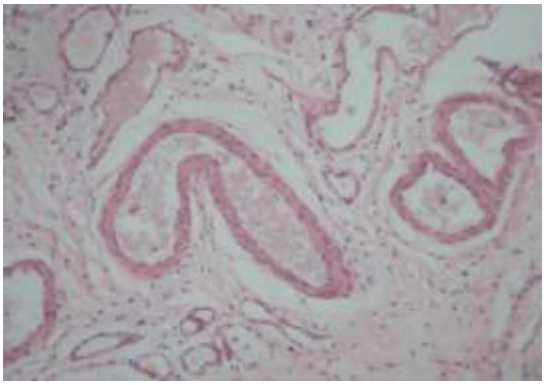

Fig.3 (H&E x 400)

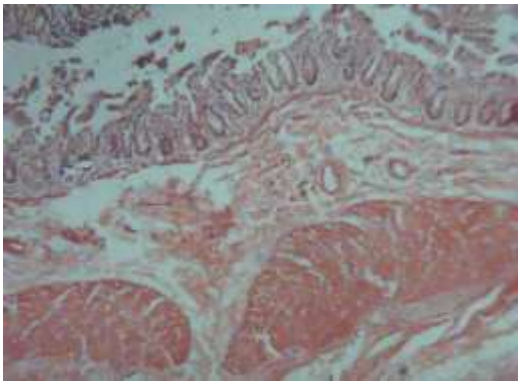

Fig.4 (H&E x 100)

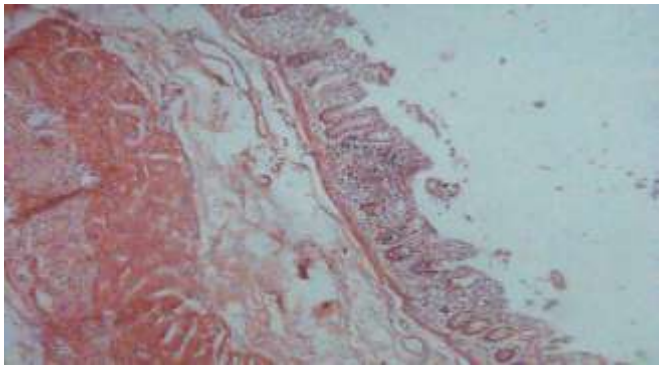

Fig. 5 (H&E x 100)

Discussion

Intestinal neuronal dysplasia (IND) is a complex alteration of the enteric nervous system (ENS), mostly involving rectum and colon, or the whole intestine. This disorder is a frequent cause of intestinal dysmotility and pseudo-obstruction. Although IND may exist as an isolated condition [6], more commonly, it occurs in association with aganglionosis[7-9]. Angiodysplasia is probably an acquired degenerative lesion associated with the aging process, in which malformed vessels are found in submucosa of the gastrointestinal (GI) tract [10]. They appear due to abnormalities of the arteriolar- capillary sphincter and subsequent large increase in the blood outflow, which results in dilatation of veins, venules and capillaries [11]. These lesion may be located anywhere in the GI tract (stomach, jejunum, duodenum), but most of them are found in the caecum and the ascending colon [12]. In our case, we report the histopathology findings of simultaneous intestinal neuronal dysplasia along with angiodysplasia. angiodysplastic lesions of small bowel with no significant gastrointestinal bleeding and intestinalneuronal dysplasia of small bowel as a rare reason of intestinal pseudo-obstruction. Our case illustrates another rare cause of intestinal obstruction.

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