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Congenital Jejunal Diverticulum

— A Case Report —

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INTRODUCTION

Jejunal diverticulum is an uncommon condition which was first described by Cooper in a 65 year old man in 1807[1]. Rosedale and Lawrence[2] reported an incidence of 0.06% in 5000 routine autopsies. Rankin and Martin[3] recorded three cases of jejunal diverticulosis in 966 small bowel resections (0.31%), Khatri et al[4] reported 4 cases among 40431 hospital admission (0.01%). Edward[5] found 9 cases of diverticulosis of jejunum in 2820 autopsies (0.32%).

Recently, authors experienced a case of congenital jejunal diverticulum in a 4 year old male child complicated by perforation. Herein we report this rare intestinal malformation with brief literature review.

A CASE REPORT

A 4 year old male was admitted to the hospital with chief complaints of severe abdominal and both shoulder pain. He had intermittent abdominal pain one day ago which was aggrevated progressively. On the physical examination, the patient showed a acutely ill appearance, but general conditions were good. Vital signs were; blood pressure 110/70 mmHg, pulse rate 100/min., respiration rate 25/min., body temperature 36.8°C and body weight 29 kg. Abdomen was slightly distended and tender without rebound tenderness. Incision scar due to previous herniorraphy at the right inguinal area one year ago was noted. CBC findings were; Hb. 12.3 gm%, Hct 37%, and WBC 14,500/mm with neutrophilic predominancy. Urinalysis was within normal limits. Simple abdomen revealed free air in both subdiaphragmatic areas and dilated small bowel lumen by gas. Chest PA was nonspecific. Family history was not contributary. Emergency operation was done under the impression of upper gut perforation.

At laparotomy a diverticulum was found at the 70 cm distal to the treitz ligament with perforated hole about 0.5 cm in diameter surrounded by fibrinous exudates. Segmental resection and end to end anastomosis were done. Other intestinal loops were within normal limits. Postoperative course was uneventful. Postoperative X-ray of small bowel series showed no other diverticulum.

Grossly the segment of jejunum attached with large diverticulum at the antemesenteric border, measuring 8 cm and 4.5 cm in longitudinal length, respectively, was submitted. Serosal surface of diverticulum showed grayish white fibrinous exudates. Cut surface exhibited a large opening of diverticulum to the lumen of jejunum, measuring 3 cm in diameter, and hemorrhagic necrosis at the mid portion of diverticulum where it was perforated. The wall of the diverticulum was thin but had muscular layers. Serosal surface also showed focal hemorrhage and fibrinous exudates (Fig. 1).

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Fig. 1. Resected segment of jejunum showing large diverticulum at the antemesenteric border with hemorrhagic necrosis and perforation in the mid portion.

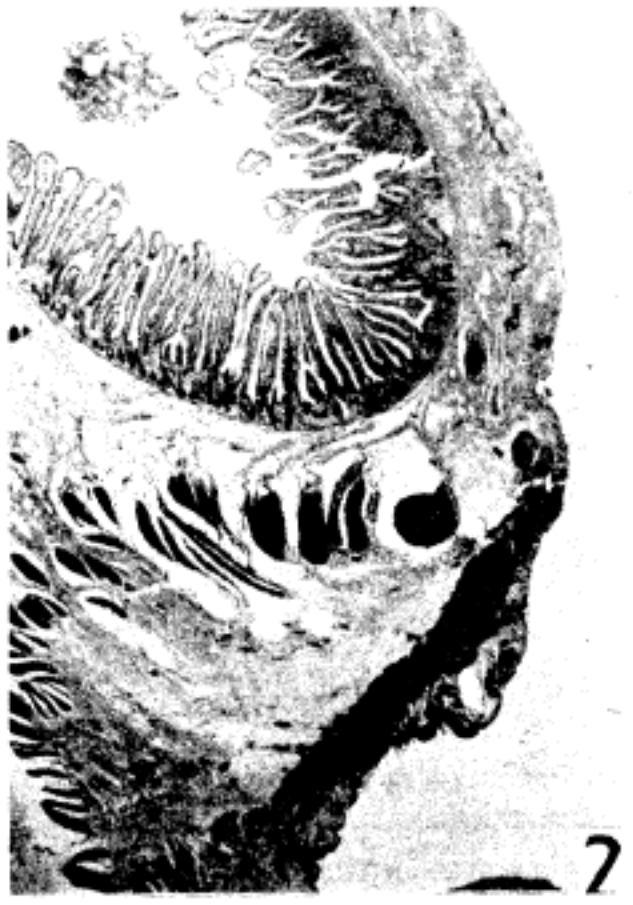

Fig. 2. Marked thinning of diverticular wall with underdeveloped muscular layers. (H&E, × 30)

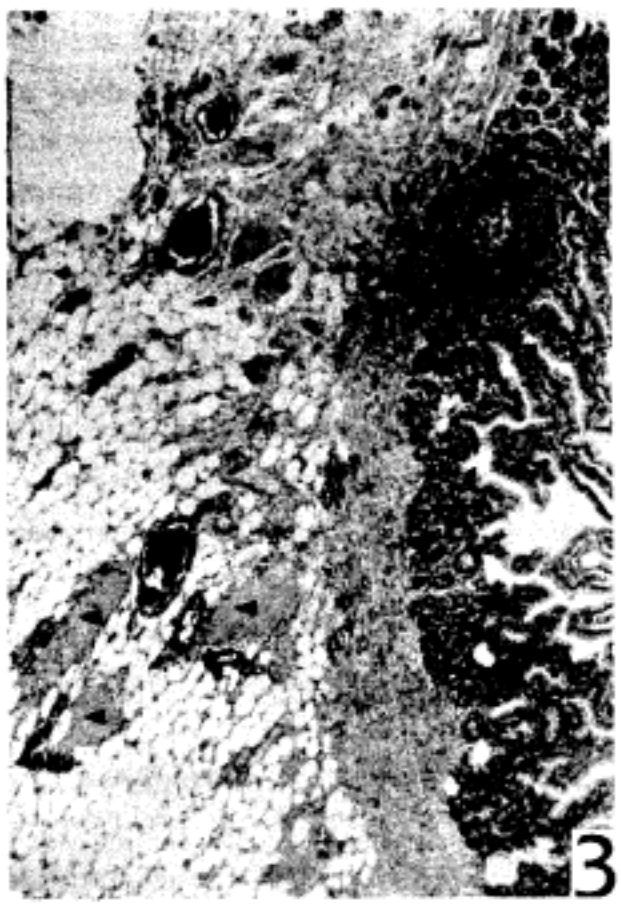

Fig. 3. Well developed lymphoid follicles with flattened inner muscular layers and scattered outer muscular bundles in surrounding fatty tissue (arrow heads). (H&E, × 40)

Fig. 4. Mucosa of perforating site revealed congestion, hemorrhage with fibrinous exudates and infiltration of inflammatory cells. (H&E, × 100)

Microscopically intestinal mucosa was well developed with lymphoid follicles but muscular wall showed underdeveloped, irregular and thinned (Fig. 2). Internal muscular layer was thinned and outer muscular layer wer incompletely developed, showing scattered muscular bundles in surrounding fatty tissue (Fig. 3). Mucosa of perforating site exhibited necrosis and hemorrhage with flattened mucosa and infiltration of inflammatory cells. Serosa was covered with fibrinous exudates. There was no evidence of other aberrant element of digestive tracts (Fig. 4). With above features this case was interpreted as congenital jejunal diverticulum complicated by perforation.

COMMENTS

Fraser[1] indicates the frequency of diverticula in the gastrointestinal tract occurs in order of colon, duodenum, esophagus, stomach and jejunum. So jejunal diverticulosis is a very rare malformation and was reported 0.015-0.32% in the literature in bowel resection cases or 0.33% in autopsy cases when by the sufflating technic into jejunum. If acquired cases are excluded from the above reported cases congenital jejunumal diverticulosis is very uncommon.

Jejunal diverticulosis is found most commonly immediately distal to 96 cm from Treitz ligament, and the size varies from a few milimeters to large outpouching (8-9 cm in diameter) but average are 1-4 cm. in diameter[7-14].

The great majority of jejunal diverticula give rise to no symptoms and those found incidentally in the course of roentgenographic examination, abdominal exploration or at necropsy. However, the triads of obscure abdominal pain, anemia and dilated loops of jejunum suggest for jejunal diverticulosis. Uncomplicated and asymptomatic diverticulosis of jejunum does not require surgical treatment. The resection with end-to-end anastomasis is the procedure of choice in the surgical treatment of jejunal diverticulosis[15-17]. Baskin and Mayo[12] reported that in their series of 87 patients with jejunal diverticulosis, nine (10.4%) developed complications that required surgical treatment. The complications included: acute or chronic obstruction, acute or chronic hemorrhage, foreign body or tumor within a diverticulum, internal fistula, perforation, intussusception, volvulus, pneumoperitoneum and macrocytic anemia. Commonest complication was stated as intestinal obstruction.

There are two types of diverticula in the small bowel: congenital and acquired. Congenital type is usually solitary, and situated at the antemesenteric border with all the coates of the intestinal wall. On the other hand acquired diverticula occur on the mesenteric border and lack the muscular coats consisting of mucosa and submucosa[6,8,11]. We considered this case as congenital type because it was solitary, and located at the antemesenteric border with muscular layers.

Etiology of congenital diverticulua is still obscure but two theories are dominated. The one is enteric duplication theory because duplication arises in the dorsal (antemesenteric) portion of the gut and gas well defined muscular wall with well developed mucosa[18]. In addition, ectopic gastric mucosa may be found and lumen may or may not communicated with gut at one or more sites. Those findings strongly indicate that congenital diverticulum is arising from enteric duplication. The other theory is inherent defect in the smooth muscle or connective tissue of the intestine although clear evidence of such defects in congenital type is lacking.[8] This case revealed flattened inner muscles and incomplete external muscles with abnormal scattered muscular bundles in surrounding fatty tissue also suggested that inherent defects in the muscular layers also played some roles in diverticulosis. Therefore, it was considered that two mechanisms played important role in the pathogenesis of diverticular disease.

SUMMARY

A Rare case of congenital jejunal diverticulum complicated by perforation in a 4 year old Korean male was reported with discussion of its clinical manifestation and pathogenesis.

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=국문초록=

선천성 공장게실 1예보고

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최윤백

공장의 게실(憩室)은 1807년 Cooper에 의해 처음 기술되었으며 0.01~0.32%의 빈도를 나타내고 있다고 보고되어 있어서 극히 드문 질환중의 하나이다.

저자들은 4세의 남아에서 발생한 공장의 선천성게실 1예를 경험하고 문헌고찰과 함께 보고하는 바이다.

본 증례는 4세의 남아로서 심한 복통과 양측 견갑부 동통을 주소로 내원하였으며 당시 복부 X-ray단순촬영상 양측 횡경막하부에 유리공기음영이 나타나 장천공을 의심하여 수술을 시행하였다. 수술소견상 Treitz 인대 하방 70cm부위 공장의 장간막반대부에 직경 1.5cm정도의 천공이 발견되었고 그 주위에는 섬유소성 삼출액이 관찰되었다. 다른부위는 정상이었으며 또 다른 게실은 발견되지 않았다. 수술은 게실을 포함한 공장의 부분절제술 및 층수돌기 절제술을 시행하였다.

육안적소견상 직경 4.5cm의 게실이 장간막 반대부에서 발견되었으며 정상 공장내강과의 연결부위는 직경이 3cm이었다. 천공부위는 게실의 정점에 위치하고 있었으며 섬유소성삼출액이 장표면을 덮고 있었고 활면상 장벽의 출혈이 관찰되었다. 현미경소견상 게실은 잘 발육된 소장점막으로 피복되어있었으며 림프절도 관찰되었고, 장벽은 근육층이 얇아져 있었으며 내측근은 얇게 구성되어있었고 외측근은 불완전하게 발육되었으며 근육속으로 지방조직이 산재하고 있었다. 천공부위 점막에는 울혈, 출혈, 섬유소성삼출액이 관찰되었을 뿐만아니라 섬유소성삼출액이 장막밖에서도 관찰되었으며 이러한 소견들을 종합하여 저자들은 공장에 선천성으로 발생된 게실이라 진단하였고 게실의 장천공으로 인한 합병증이 생겨 수술하게 되었다고 생각되었다. 또 이 증례를 통하여 선천성게실의 병인은 근육층의 이상발육과 장중복이 다 관여되리라 추측되었다.