CYTOMEGALOVIRUS PROCTITIS: REPORT OF CLINICAL, ENDOSCOPIC AND PATHOLOGIC FEATURES IN A PATIENT WITHOUT EVIDENCE OF IMMUNODEFICIENCY

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Cytomegalovirus (CMV) infections are recognized as important complications in immunocompromized patients. However only few reports mentioned the occurrence of CMV gastrointestinal infections in previously otherwise healthy persons.

We describe a case of CMV proctitis who is lack of immunocompromized condition.

A 65-year-old heterosexual man had repeat history of anal bleeding. Colonoscopic examination revealed a markedly erythematous fragile and about 4 cm in size of polypoid mass, with punch-out ulcers at the rectum just beyond the anal verage. Endoscopic biopsies revealed chronic inflammatory cells infiltration, angiomatous proliferation at submucosa, with CMV inclusion in some endothelial cells. Anti-CMV antibody IgG titer were 1:64(+) initially, raised to 1:128(+) one month later then down to 1:64(+) after another one month.

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Key words: ***cytomegalovirus, proctitis***

Cytomegalovirus (CMV) infections in adults are not uncommon, but usually are subclinical. With the recent increased number of immunocompromised patients, CMV is recognized as an important cause of disease in these cases[3] but the gastrointestinal involvement is still infrequent and usually seen in the context of overwhelmingly generalized CMV infections. There are some reports of isolated gastrointestinal tract manifestation of CMV infections in patients with ulcerative colitis renal transplantation recipients and patients with acquired immune deficiency syndrome (AIDS). Few reports mentioned the occurrence of CMV gastrointestinal infections in previously otherwise healthy persons[8]. We describe a case of CMV proctitis who is lack of immunocompromised condition.

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CASE REPORT

A 65-year-old heterosexual man

with repeat history of anal bleeding was admitted because of right hemiparesis following a motorcycle accident. On admission, he developed quadriplegia the next day, serial X-ray and imaging diagnostic examinations showed fracture of the 7th cervical spine and cervical spinal stenosis due to ossified posterior longitudinal ligament (OPLL). Laminectomy was performed in the third day of admission. Unfortunately, three days after operation, he suffered from anal bleeding. Colonoscopic examination was performed three days after revealed a markedly erythematous, fragile and about 4 cm in size of polypoid mass, with punched-out ulcers and exudative coating at the posterior wall of rectum just beyond the anal verge. In addition, about a 1.5 cm in size of bridging pseudopolyp was demonstrated in the vicinity of the mass (Fig. 1), multiple endoscopic biopsies showed chronic inflammatory cells infiltration, and angiomatous proliferation involving submucosa, with CMV inclusion shown in some endothelial cells (Fig. 2). Serologically, anti-CMV antibody IgG titer was 1:64 (+) initially, it raised to 1:128 (+) in one month duration then went down to 1:64 (+) in another month's interval. Although anti-CMV antibody IgM was negative, peripheral white blood cell count showed persistent monocytosis lasting for over two months. Clinically, this patient showed no evidence of immuno-deficiency, no lymphocytopenia, normal immunoglobulin analysis and no evidence of opportunistic infection except the presenting CMV infection. Therapeutic trial with Salazopyrin (Sulfasalazin) 500 mg TID was started. Follow-up colonoscopic examination was done a month later and showed remarkable improvement of afore-mentioned lesion (Fig. 3).

DISCUSSION

CMV infection had been regarded as an infectious disease mainly affecting children. In the review paper of Wong *et al*[2] in 1962 only 41 cases of adult CMV were found in the literature. Adult CMV infection increased markedly in recent days especially in those immunocompromised patients, in most of these cases gastrointestinal involvement usually is part of a systemic infection. Few cases of CMV infection localized in gastro-intestinal tract in otherwise healthy adults without showing evidence of immuno-incompetency have been reported lately; endoscopic biopsies of two patients with gastric ulcer only were found to have typical intranuclear CMV inclusion in the glandular cells by Andrade *et al*[7] one CMV primary infection in a patient with idiopathic proctitis was reported by Cunningham *et al*[9] and Villar *et al*[8] had reported a young patient who was a traffic accident victim had CMV infection presenting with a picture of acute erosive esophagitis following multiple blood transfusions. Our patient did not have a definite history of chronic proctitis before this episode, but he had history of several times of anal bleeding, although he himself allegedly thought or speculated it was hemorrhoid bleeding, there was no medical confirmation so far. From the endoscopic picture the lesion looked like a chronic one; there was a bridging pseudopolyp. Laboratory findings also favor it was not an acute CMV infection, although its diagnostic value was dubious. This patient had picture of peripheral monocytosis, and it persisted for more than two months. All of these data might just reflect it was a

Fig. 1(A)

Fig. 1(B)

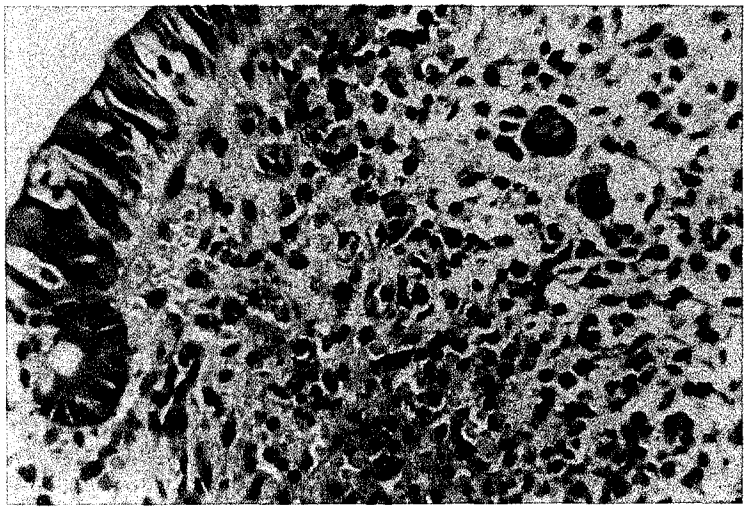

Fig. 2

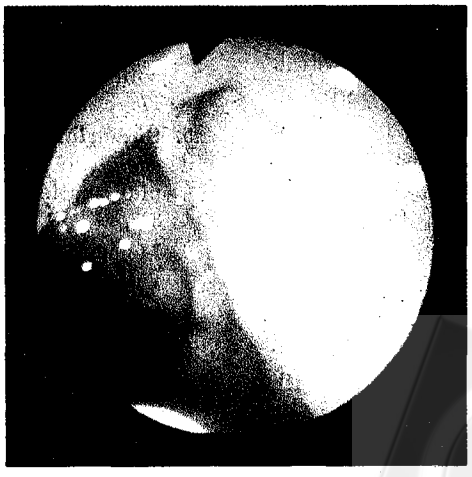

Fig. 3

reactivation of latent CMV infection.

The histopathologic examination of endoscopic biopsy of our patient revealed CMV inclusions in endothelial cells of blood vessels, this is different from reports of many authors[10]. In these reports the CMV inclusions were found in epithelial cells of gastrointestinal mucosa. In reports of Foucar *et al*[11] and Meiselman *et al*[6] CMV vasculitis was noted to be a predominant picture in their cases. All of them were immuno-compromised and fatal. There were other 12 cases in literatures reviewed by Foucar *et al*[11] with significant lower GI bleeding and having evidence of CMV vasculitis in the colon mucosa, most of these cases had underlying chronic ulcerative colitis, although half of them were fatal, the other half cases had better outcome. Our case was similar to the latter ones.

Goodman *et al*[12] reviewed seven cases of CMV inclusions in the colon, most of them had the inclusions in granulation tissue of either ulcers or perforation without associated vascular endothelial inclusions. He recognized these findings were consistent with a superinfection of damaged colonic tissue, and subsequent exacerbation of the underlying disease process. The case reported by Cunningham *et al*[8] had epithelial inclusions only, although he showed the evidence of acute CMV infection including IgM antibody and seroconversion, the possibility of superinfection could not be ruled out. Our case is more favorably inclined to be a reactivation latent infection, presenting with the picture of CMV vasculities, in which it is possible that a CMV infection may play a critical role in its pathogenesis.

Acyclovir had been employed with temporary improvement in one of two reported cases of Meiselman *et al*[6], Salazopyrin was prescribed in the reported case of Cunningham *et al* with dramatic improvement. Our patient did not have evidence of immunodeficiency, so we tried Salazopyrin treatment and resulted in a propitious upshot. The auspicious consequence of Salozopyrin therapy in this situation is difficult to elucidate or interpret for evaluation since it may involve the underlying idiopathic proctitis which also responds to the medication as well. Thus it makes a final diagnosis either of pure CMV proctitis or concomitantly superimposed idiopatic proctitis in this particular case a dilemma.

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巨細胞濾過性病毒直腸炎

——一 例 報 告——

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巨細胞濾過性病毒感染是免疫不全病人重要的併發症之一，原本身體健康的病人感染此病毒的情形並不多見，僅有少數的報告中曾提及此種病例。

本文即非免疫不全病人罹患巨細胞濾過性病毒直腸炎的病例報告。病人65歲，男性，非同性戀者，有經常性肛門出血現象，大腸鏡發現肛門上方有一紅腫脆弱的腫瘤，約4公分大且有潰瘍現象。切片檢查報告指出，黏膜下層有炎性細胞浸潤、血管增生及少數肉皮細胞含有巨細胞濾過性病毒。最初 IgG 型抗巨細胞濾過性病毒抗體約1:64倍，一個月後昇至1:128倍，二個月後又降至1:64倍。(中消醫誌 1988; **5**: 119-123)

關鍵語：巨細胞濾過性病毒，直腸炎