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Case Report / Vaka Sunumu

Pathology / Patoloji

Inflammatory rectal polyp with osseous metaplasia

Kemik metaplazisinin eşlik ettiği inflamatuvar rektal polip

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SUMMARY

To present a rare case of osseous metaplasia in an inflammatory rectal polyp. A 9-year-old boy admitted to the clinic with a 6 month history of intermittent rectal bleeding that occurred approximately 15 times . An 8 mm polyp in diameter was totally excised from rectal wall. Histologically, elongated, dilated, mucin filled colonic glands were scattered within stroma which was composed of both acute and chronic inflammatory and granulation tissue. In the stroma there was a small focus of new bone formation, which was covered with intact colonic mucosa. We presented an extremely rare case report of a metaplastic bone formation in a rectal inflammatory polyp.

Key words: *Rectum, osseous metaplasia, inflammatory polyp*

ÖZET

Ender bir olgu olan "inflamatuvar rektal polipte osseöz metaplazi" sunumu. Dokuz yaşında erkek hasta 6 aydır ara ara olan yaklaşık 15 kez yineleyen rektal kanama yakınmasıyla başvurdu. Rektal duvardan 8 mm çapta polip eksize edildi. Histolojik olarak, akut ve kronik inflamasyon ve granülasyon dokusunda oluşan stromada dağınık uzamış, genişlemiş, müsinle dolu kolonik glandlar görüldü. Stromada küçük bir odakta normal kolonik mukoza ile çevrili yeni kemik oluşumu dikkati çekti. Oldukça ender görülen rektal inflamatuvar polipte metaplastik kemik oluşumu olgusu literatür bilgileri ile sunuldu.

Anahtar kelimeler: *Rektum, osseöz metaplazi, inflamatuvar polip*

INTRODUCTION

Osseous metaplasia is a heterotopic bone formation[1]. While it is rarely seen in various types of polyps of the stomach, colon, and rectum of the gastrointestinal tract, its occurrence in colonic polyps is extremely rare[2-4]. Mark, in 1964, firstly descripted bone formation within a rectal polyp[4]. The mechanism of this metaplastic change has not been completely understood[1-3]. Clinically and prognostically, its significance is not known[1].

Herein, we report a case that is osseous metaplasia in an inflammatory rectal polyp in a 9-year-old boy with an accompanying literature review.

CASE REPORT

A 9-year-old boy admitted to the clinic with a 6-month history of intermittent rectal bleeding which was independent from defecation and occurred approximately 15 times during this period and. The bleedings were light colored drips without coagulum. Hemorrhoid or anal fissure formation was not detected during digital anal examination, and only a suspect lesion could be palpated. Biochemical test results were within normal limits. A flexible colonoscopic examination was performed and the only pathologic finding was a polyp 8 mm in diameter which was totally excised from the rectal wall (Figure 1). During one year of follow up period, the patient was free of any symptoms.

On gross examination, tan-brown colored polypoid mass, which was 0.8 cm in diameter, was seen. It was bisected on its long axis and submitted for histological examination. Microscopically, surface of the polyploid mass was covered by fibrinopurulent

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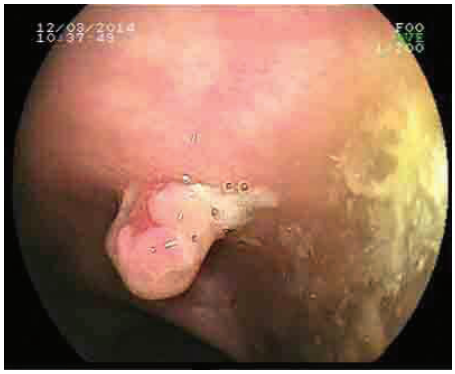

Figure 1. Rectal polyp on endoscopic examination.

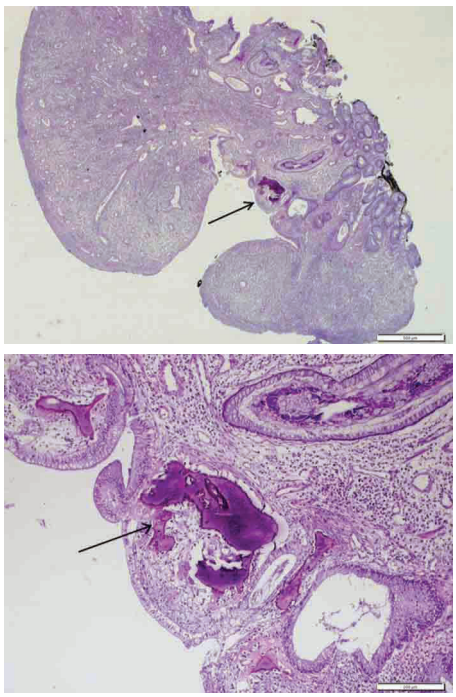

Figure 2A. Inflammatory polyp with osseous metaplasia (arrow)(H&Ex20), B. Bone trabecula in polyp stroma (arrow) (H&Ex100).

debris. Elongated, dilated, mucin filled colonic glands were scattered within stroma which was composed of both acute and chronic inflammatory and granulation tissue (Figure 2A). In the stroma, there was a small focus of new bone formation that was covered with intact colonic mucosa (Figure 2B). Bone marrow cells were not visible.

DISCUSSION

Osseous metaplasia in colonic polyps is extremely rare. In English-written literature, only twenty-one cases have been reported up to date together with our case. We reviewed and summarized the subject of osseous metaplasia in colon polyps (Table 1). The patients comprised thirteen men and five women, besides two whose genders were not described. The mean age of the patients was 69 yeras (range: 3-85 years). Mean diameter of polyps was 16.3 mm (range: 8-50 mm). Histologically, six lesions were neoplastic (three tubular adenomas and three tubulovillous adenomas), while the remaining fifteen lesions were non-neoplastic entities (six inflammatory polyps, seven juvenile polyps and one traditional serrated adenoma). Site of involvement was rrectum-rectosigmoid (n=16), sigmoid colon (n=2), cecum (n=1), and transverse colon (n=1).

Osseous metaplasia can be seen in both benign and malignant conditions. The pathogenesis of osseous metaplasia remains unknown. Many theories have been published concerning its pathogenetic mechanism.

Osteogenic stimulation was considered as the result of inflammatory process[2]. Necrosis, inflammation, pre-existing calcification, increased vascularity and extracellular mucin deposition are reported to be associated with heterotopic bone formation in tumors[7]. Both benign and malignant lesions with osseous metaplasia are commonly associated with mucin production and extravasations. Benign lesions with osseous metaplasia are often seen with active chronic inflammation and or ulceration[7,8,13]. In 1939, Dukes reported a case series of osseous metaplasia in rectal carcinoma. In these series, long duration of symptoms, low grade tumors with low metastatic potential and presence of necrosis were shown as contributing factors[4]. He also suggested osseous formation after dystrophic calcification of necrotic tissue[4,17]. In 1964, Marks and Atkinson suggested that osseous metaplasia might result from transformation of fibroblasts into other types of mesodermal tissue, especially osteoblasts[4,13]. In 1989, Randall et al.[18] showed expression of alkaline phosphate - which is a marker of bone synthesis- in osteoblast- like cells of metaplastic bone and to a lesser degree in glan-

Table 1. Summary of reported cases of benign colorectal polyps with osseous metaplasia.

Year	Author	Age	Gender	Site	Size (mm)	Types of Polyp
1964	Mark (4)	10	M	Rectum	NI	Juvenile
1981	Sperling (5)	25	M	Rectum	10	Inflammatory
1992	Drut (4)	5	M	Rectosigmoid	10	Juvenile
1992	Drut (4)	4	M	Rectum	5	Juvenile
1992	Castelli (6)	22	F	Rectum	10	Inflammatory
1994	Groisman (7)	67	M	Rectum	18	Tubulovillous adenoma
1994	Groisman (7)	3	F	Rectum	20	Juvenile
1996	Cavazza (8)	NI	NI	NI	NI	Tubulovillous adenoma
1999	McPherson (9)	73	M	Cecum	20	Tubulovillous adenoma
2000	Rothstein (10)	NI	NI	Sigmoid colon	25	Tubular adenoma
2005	Al-daraji (11)	85	F	Sigmoid colon	15	Tubular adenoma
2008	White (12)	63	F	Transverse colon	NI	Tubular adenoma
2009	Oono (13)	39	M	Rectum	12	Inflammatory polyp
2009	Ahmed (14)	17	M	Rectum	18	Juvenile retension polyp
2010	Wilsher (15)	50	M	Rectosigmoid	25	traditional serrated adenoma
2012	Odum (4)	74	M	Rectum	10	Inflammatory polyp
2012	Montalvo (16)	62	M	Rectum	50	traditional serrated adenoma
2012	Bhat (2)	5	F	Rectum	15	Juvenile retension polyp
2013	Bhattacharya (3)	14	M	Rectum	10	Inflammatory polyp
2013	Garg (1)	6	M	Rectum	13	Juvenile polyp
2014	Current case	9	M	Rectum	8	Inflammatory polyp

dular cells of metastatic colonic adenocarcinoma and proliferating mesenchymal cells. Recent studies have suggested the role of expression of bone morphogenetic proteins (BMPs) in the pathogenesis of osseous metaplasia[19]. BMPs are members of the TGFβ superfamily and play a major role in the formation of new bone, except BMP-1 which is a metalloproteinase and a marker for cartilaginous differentiation. In immunohistochemical analysis, BMP-2, BMP-4, BMP-5, and BMP-6 were demonstrated in colonic adenorcacinomas with osseous metaplasia by Imai et al.[20] In 2001. BMP-2, BMP-4, BMP-5, and BMP-6 were found to be present in the cytoplasm of tumor cells and within the osteoblast-like cells of newly formed bone, and BMP-2 and BMP-4 were shown in stromal fibroblasts. In 2007, Liu et al.[19] immunohistochemically demonstrated expression of BMP-1, BMP-4 and BMP-6 in both stroma and epithelium in various cases with osseous metaplasia.

Pathogenesis of osseous metaplasia is still under investigation. Its exact in vivo pathogenetic mechanism is still unknown but in vitro studies which were done with mouse and human fibroblast cultures showed that four transcription factors (Oct ¾, Sox-2, c-myc, Klf4) are related with the production of pluripotent stem cells. Subsequently, these stem cells are shown to differentiate into different cell types of all three germ layers[21,22].

An osseous metaplasia is an incidental finding and it is clinically and prognostically insignificant. Its exact pathogenesis is unknown but transformation of the fibroblasts into osteoblasts is suggested. Here, we presented an extremely rare case of a metaplastic bone formation in an inflammatory rectal polyp.

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