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CASE REPORT

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Juvenile rectal polyp with osseous metaplasia: A rare case report

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ABSTRACT

Heterotopic bone formation is seldom detected in lesions of gastrointestinal pathology. We report a rare case of osseous metaplasia in a juvenile rectal polyp in a four year old boy with a brief review of literature. As per the literature survey, this appears to be ninth case of osseous metaplasia in a juvenile rectal polyp.

ARTICLE HISTORY

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KEYWORDS

Osseous metaplasia; juvenile polyp; rectal polyp

Introduction

Osseous metaplasia occurring in colonic polyps, is extremely unusual [1]. There are only few case reports of neoplastic and non neoplastic gastrointestinal polyps with osseous metaplasia [2,3]. An electronic literature search undertaken revealed only eight cases of juvenile colonic polyps with osseous metaplasia and fourteen cases of other morphological types of colorectal polyps with bone formation [4,5]. Though occurrence of bone in the polyp is an incidental finding and does not have any clinical or prognostic significance, the mechanism of bone formation is still a topic of significant debate [2].

Case Report

A four year old boy was brought to the paediatric surgery clinic with history of intermittent bleeding on passing stools since one month. Mother had noticed an anal projection during defeacation since one week. Proctoscopic examination revealed a solitary pedunculated polyp in the rectum measuring 1×1cm. Polypectomy was done under general anesthesia and sent for histopathology.

Gross examination revealed a 1×1cm red pedunculated polyp with greywhite and grey brown areas on cut section. Microscopy showed a polyp focally lined by columnar epithelium. Surface of polyp was ulcerated and covered by granulation tissue. Polyp consisted of cystically dilated mucosal glands of varying sizes lined by columnar mucinous epithelium with inspissated mucin. Stroma was edematous with foci of mature bony trabeculae and was infiltrated by mixed inflammatory infiltrate [Fig 1 and 2].

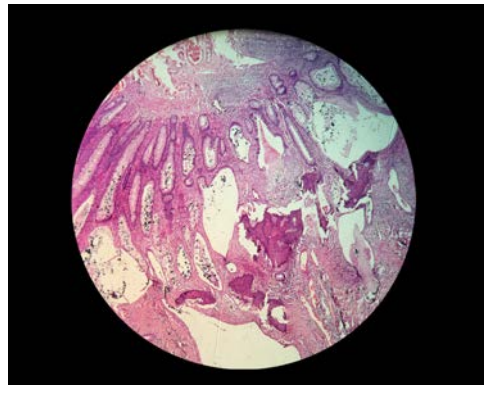

Figure 1. Polyp consisting of cystically dilated glands with osseous metaplasia in the stroma (H and E ×40).

Discussion

Osseous metaplasia is very rare in colon and only few case reports have been published from India [4]. Only 22 cases of osseous metaplasia have been reported in colonic polyps in literature reviewed and among these juvenile polyps

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account for eight cases. Others include 6 inflammatory polyps, 6 tubular/tubulovillous adenomas and 2 serrated adenomas. Out of 22 reported cases, the mean age of occurrence of polyps was 33.6 years, Male to female ratio being 2.3:1. Size of colonic polyps ranged from 5-50mm [Table 1].

Amongst the 8 cases with juvenile polyps, majority of the patients were males (75%), with a mean age of 7.9 years (range 3-15 years). The polyp size varied from 5mm to 20mm, with a mean size of 13.3mm and majority being located in the rectum (87.5%) [Table 1].

Table 1. Summary of reported cases of benign colorectal polyps with osseous metaplasia [4].

Serial No.	Year	Author	Age in years	Gender	Site	Size of polyp in mm	Type of polyp
1	1964	Mark	10	M	Rectum	NK*	Juvenile polyp
2	1981	Sperling [6]	25	M	Rectum	10	Inflammatory
3	1992	Drut	5	M	Rectosigmoid	10	Juvenile polyp
4	1992	Drut	4	M	Rectum	5	Juvenile polyp
5	1992	Castelli [7]	22	F	Rectum	10	Inflammatory
6	1994	Groisman	67	M	Rectum	18	Tubulovillous adenoma
7	1994	Groisman [8]	3	F	Rectum	20	Juvenile polyp
8	1996	Cavazza	NK*	NK*	NK*	NK*	Tubulovillous adenoma
9	1999	McPherson	73	M	Cecum	20	Tubulovillous adenoma
10	2000	Rothstein	NK*	NK*	Sigmoid	25	Tubular adenoma
11	2005	Al-daraji	85	F	Sigmoid	15	Tubular adenoma
12	2008	White	63	F	Transverse	NK*	Tubular adenoma
13	2009	Oono [9]	39	M	Rectum	12	Inflammatory polyp
14	2009	Ahmed [10]	15	M	Rectum	18	Juvenile retention polyp
15	2010	Wilsher	50	M	Rectosigmoid	25	traditional serrated adenoma
16	2012	Odum [11]	74	M	Rectum	10	Inflammatory polyp
17	2012	Montalvo	62	M	Rectum	50	traditional serrated adenoma
18	2012	Bhat [2]	5	F	Rectum	15	Juvenile retention polyp
19	2013	Bhattacharya [1]	14	M	Rectum	10	Juvenile polyp
20	2013	Garg [3]	6	M	Rectum	15	Juvenile polyp
21	2013	Bhawna [5]	42	F	Rectum	14	Inflammatory polyp
22	2014	Zemheri [4]	9	M	Rectum	8	Inflammatory polyp
23	2016	Present case	4	M	Rectum	10	Juvenile polyp

*NK-Not Known

Juvenile polyps are hamartomatous lesions which may show the phenomenon of osseous metaplasia [3]. Mark (1964) first described bone formation within juvenile rectal polyp [4]. Sperling (1981) described the phenomenon of bone formation within an inflammatory rectal polyp [6].

The largest case review (52 cases) of osseous metaplasia in the gastrointestinal tract (excluding liver and pancreas) was by Ansari et al in 1992. In this review, the mean subject age was 55 years, and the diagnosis in 47 out of the 52 cases were adenocarcinomas. The majority of cases were in the colon, the most common site being the rectum (21/52 cases). Histologically the malignant lesions with osseous metaplasia were commonly seen with the presence of mucin extravasation [5,12].

Marks and Atkinson (1984) suggested that osseous metaplasia may be due to transformation of fibroblasts into osteoblasts [9]. Many theories have been published regarding this mechanism. Recent studies have suggested the expression of bone morphogenic proteins (BMPs) in regard to osseous metaplasia [13]. BMP 2 and BMP4 were noted in stromal fibroblasts [1]. In inflammatory polyps, persistent inflammation resulted in osteogenic stimulation suggesting that inflammation plays an important role in benign polypoidal lesions [5,9].

Mouse and human fibroblast in vitro cultures have found to generate pluripotent stem cells under the influence of four specific transcription factors (Oct3/4, Sox2, c-Myc, and Klf4). Later on, these stem cells have exhibited the capability to differentiate into different cell types [14,15]. This mechanism explains that, the mature fibroblast located within the stromal component of these intestinal lesions may be under the influence of these similar transcription factors, leading to differentiation of stem cells to osteoblasts [1].

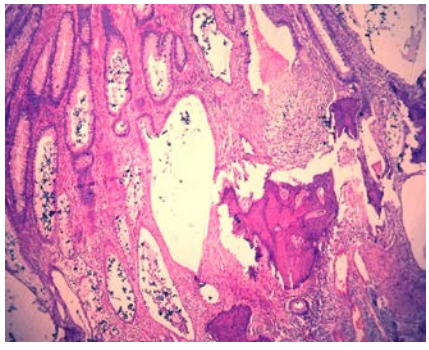

Figure 2. Stroma of the polyp showing osseous metaplasia (H and E ×400).

Conclusion

Osseous metaplasia occurring in colonic polyps is extremely rare. As per the literature survey, this appears to be ninth case of osseous metaplasia in a juvenile rectal polyp.

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