



CASE REPORT

Juvenile Polyp with Osseous Metaplasia Presenting with Fecal Incontinence in Young Patient : Case Report of Unusual Presentation

Fatima Alrowaiei¹, Mohammed Awadh^{2*}, Thamer Alabbasi³, Abdulla Darwish⁴

¹Resident in Pathology, Bahrain Defense Force Royal Medical Services, Bahrain

²Consultant Histopathologist, Bahrain Defense Force Royal Medical Services, Bahrain

³Consultant Surgeon, Bahrain Defense Force Royal Medical Services, Bahrain

⁴Consultant Histopathologist, Bahrain Defense Force Royal Medical Services, Bahrain

*Corresponding author:

Mohammed Awadh, Consultant Histopathologist, Bahrain Defense Force Royal Medical Services, Bahrain.

E-mail: Mib141@hotmail.com

Received date: September 17, 2024; **Accepted date:** January 20, 2025; **Published date:** March 31, 2025

Abstract

Osseous metaplasia is rare in colorectal lesions and exceedingly rare in colonic polyps. We present a case report of rectal juvenile polyp with stromal osseous metaplasia in a 21-year-old male who presented with fecal incontinence episodes throughout the day and night over the past 2 years. The patient underwent a colonoscopy and polypectomy, which showed a juvenile polyp with osseous metaplasia. The patient's primary concern was to fully recover postoperatively. Juvenile polyp with osseous metaplasia may be added to the differential diagnosis of patients with fecal incontinence. This work aims to discuss pathogenesis and review the literature on this rare condition.

Keywords: Colorectal polyp, Juvenile polyp, Osseous metaplasia, Colorectal tumors, Intestinal imaging

Introduction

Juvenile polyps are benign, non-cancerous growths in the gastrointestinal tract; typically, they can develop sporadically or in juvenile polyposis syndrome.^{1,2} Osseous metaplasia is a rare phenomenon described in many neoplastic conditions and rarely in non-neoplastic lesions.³ It is undeniably a remarkable histological feature that has not yet shown clinical significance, and the underlying pathogenesis remains unclear.⁴⁻⁷

Here, we report a case of juvenile rectal polyp with osseous metaplasia incidentally discovered in a 20-year-old male who presented with a unique chief complaint.

Case report

A 20-year-old male Arab patient with no significant medical history presented to the general surgery clinic, reporting fecal incontinence episodes throughout the day and night over the past 2 years, which became worse in the last month of his presentation. He denied experiencing specific neurological symptoms or undergoing previous perianal surgeries. He had no history of smoking, alcohol, or drug use, and there were no records of prior medical visits or tests. Physical examination was remarkable for weak voluntary contraction of the external anal sphincter. Otherwise, the remaining examination, such as stool analysis for infectious pathogens, was unremarkable.

Initially, the patient was advised to have Magnetic resonance imaging (MRI) of the pelvis to eliminate the possibility of any neurological disorders, such as demyelination or spinal cord lesions. However, the MRI revealed a small nodule from the lateral rectal wall, likely residual fecal material or polyp. Apart from that, the rest of the report showed no significant findings. Therefore, the patient underwent a complete colonoscopy, revealing a large polyp at the lower mid-rectum. [Figure 1]. The polyp was excised using a hot snare, and multiple random colonic biopsies were obtained and sent for histopathological analysis. Grossly, the main specimen consisted of a single tan soft polypoidal tissue with scattered grey-white areas measuring 1.3 x 1.2 x 0.4 cm and multiple fragments of soft tan tissue measuring 1.5 x 0.3 cm in aggregate. The polyp was bisected.



Figure 1: Large polyp at the lower mid-rectum on endoscopic examination.

On microscopic examination, the rectal polyp showed mucosal ulceration, granulation tissue, heavily mixed inflammation, and dilated glands with splaying of muscularis mucosae, features consistent with juvenile polyp (Figure 2 A-D). Additionally, the polyp showed areas of calcification, bony metaplasia, and mucin production. No dysplasia or malignancy was seen. Overall, appearances are in keeping with juvenile polyps with osseous metaplasia. The remaining separate colonic biopsies were within normal limits.

Discussion

To our current understanding, osseous metaplasia is uncommon in colonic polyps, and there have been few documented cases of osseous metaplasia

in juvenile rectal polyps. The rarity of this phenomenon limits the possibility of conducting larger studies to determine the actual cause of this phenomenon.

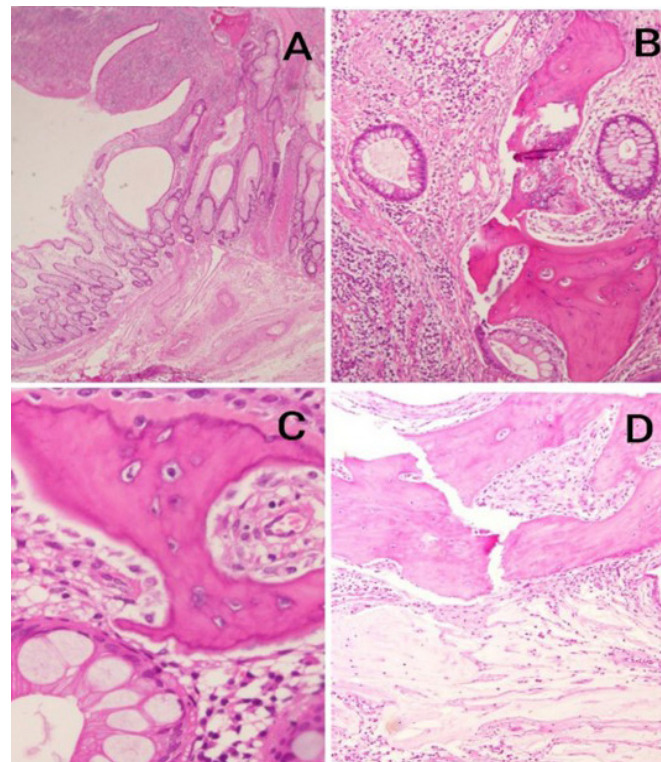


Figure 2: (A) Low power view shows dilated glands, surface ulceration with inflamed stroma. (B) High power view shows heavy inflamed stroma with central osseous metaplasia and basophilic calcification. (C) Osseous metaplasia adjacent to a gland. (D) Osseous metaplasia with extravasated mucin in the stroma.

Osseous metaplasia involves the abnormal development of bone tissue in areas where it typically doesn't occur. It can be found in cancerous and non-cancerous conditions and is characterized by the creation of mature bone within soft tissues.^{1,3, 6,8} In 1981, Sperling reported the occurrence of bone formation in a benign rectal polyp. Since then, this phenomenon has been observed in several gastrointestinal polyps featuring osseous metaplasia.^{9,10}

There have been approximately 22 documented cases of osseous metaplasia in juvenile rectal polyps, excluding this case.^{1,4-6,10-16} The age at the time of diagnosis ranged from 3-17 years; our case was diagnosed at the age of 20 years; nonetheless, it is still considered a young age group. A literature

review done in 2021 revealed that seven out of 9 cases of juvenile polyps with osseous metaplasia were males.^{12,17}

Furthermore, the polyps ranged in size from 10 to 30 mm, with an average size of 13.68 mm (13 mm in our case) and were consistently found in the rectum, as occurs in our patient. Most cases, including ours, showed no evidence of juvenile polyposis syndrome. However, one case in 2020 was reported in the context of juvenile polyposis syndrome.^{1,18,22}

In 2021, a literature review was conducted to compare and summarize previously reported cases of colorectal polyps with osseous metaplasia. Out of 28 cases, 20 were in non-dysplastic colorectal polyps, which include 11 inflammatory polyps and nine juvenile polyps. These figures suggest that osseous metaplasia is more common in non-dysplastic polyps.²² However, with new cases still being reported, it remains unclear whether osseous metaplasia is more prevalent in inflammatory or juvenile polyps.

The rarity of this case lies in the unusual complaint of fecal incontinence and histological findings. Most reported cases presented with rectorrhagia, one with constipation, and three with mass protrusion.¹ Additionally, in 2024, Le JH et al. reported a case of JP with periumbilical pain and unintentional weight loss of 4.5 kg.⁶

Several mechanisms have been suggested, yet the pathogenesis of osseous metaplasia remains unknown. Osteogenic stimulation of fibroblast transformation into osteoblasts was believed to be caused by inflammation.^{4,6,10} Numerous theories have been proposed on this subject. Recent research has indicated that the expression of bone morphogenetic proteins (BMPs) in fibroblasts might contribute to the pathogenesis of osseous metaplasia. Most BMPs are part of the TGF- β superfamily and are essential for new bone formation, except for BMP-1, which is a metalloprotease and indicates cartilaginous differentiation.^{1,6,19}

In 2001, Imai et al. showed the presence of BMP-2, BMP-4, BMP-5 and BMP-6 in colonic adenocarcinomas with osseous metaplasia using immunohistochemistry (IHC). In 2003, Kypson et al.,

found that BMP-2 was significantly overexpressed in tumor cells from rectal adenocarcinoma cases featuring osseous metaplasia compared to those without bone formation.^{5,10,19,20} Furthermore, *in vitro* studies have demonstrated that mouse fibroblasts exposed to four transcription factors (Oct3/4, Sox2, c-Myc, and Klf4) can generate pluripotent stem cells capable of differentiating into all three germ cell layers. Similar results have been observed in adult human fibroblasts. This raises the possibility that mature fibroblasts in intestinal lesions might be influenced by these or similar transcription factors, resulting in the generation of stem cells that can become osteoblasts. However, more research is needed to support this theory.^{5,10,19,20}

Histologically, benign and malignant lesions with osseous metaplasia often exhibit mucin production and extravasation.^{1,4} Alper et al. reported mucin production as a standard feature in two cases of colon adenocarcinomas.²¹ Additionally, three previously reported cases documented mucin extravasation histologically in juvenile polyps.^{1,6,14} Our case also demonstrated mucin production; however, limited available data makes it unclear whether mucin should be considered a definitive histological feature of these polyps. This warrants further discussion. In conclusion, the connection between our patient's primary complaint and osseous metaplasia in juvenile polyps remains unclear. The precise mechanism behind osseous metaplasia is still widely debated.

Conflicts of Interest

Nil

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