International Journal of Health Sciences and Research
www.ijhsr.org ISSN: 2249-9571

*Case Report*

# Heterotopic Ossification in Gastrointestinal Neoplasms: A Rare Occurrence

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## ABSTRACT

Osseous metaplasia is described in a variety of organs in both neoplastic and non-neoplastic conditions. In gastrointestinal tract, it is a very rare phenomenon. Only few cases have been reported in literature in colonic polyps especially in juvenile polyps and colonic malignancies. We report three cases of heterotopic ossification in juvenile polyp and one in colonic adenocarcinoma with review of literature. Many mechanisms of its pathogenesis are hypothesized but exact cause remains still unclear

***Key Words:*** Juvenile polyp, osseous metaplasia, rectal polyp

## INTRODUCTION

Osseous metaplasia (OM) in gastrointestinal tract is a rare phenomenon. It has been described in neoplastic lesions involving kidneys, liver, breast, skin and in non-neoplastic lesion like mucocoele of appendix. [1] In gastrointestinal tract; it is reported in colonic polyps and adenocarcinoma. Review of literature shows only 9 documented cases of osseous metaplasia in Juvenile polyp. Reported cases of heterotopic ossification in malignant tumors of large intestine in English literature are less than twenty. [2] Here we report four cases of heterotopic ossification in GI tract, three in juvenile polyp and one in adenocarcinoma from our Centre.

**CASE 1**

An eight year old boy presented to our outpatient department (OPD) with intermittent bleeding per rectum of two months duration. Colonoscopy showed a single polyp of 1 X0.5 cm with surface ulceration in the rectum (Figure1a). Polypectomy was done and sent for histopathological examination

Gross examination showed a polyp measuring 1x 0.5 cm. Microscopy showed a colonic polyp with focal ulceration. Glands were cystically dilated with secretions. Lamina propria was odematous and showed chronic inflammatory infiltrates. A focus of osseous metaplasia composed of bony trabeculae was seen. Hence a diagnosis of a juvenile polyp with osseous metaplasia was made (Figure 1b).

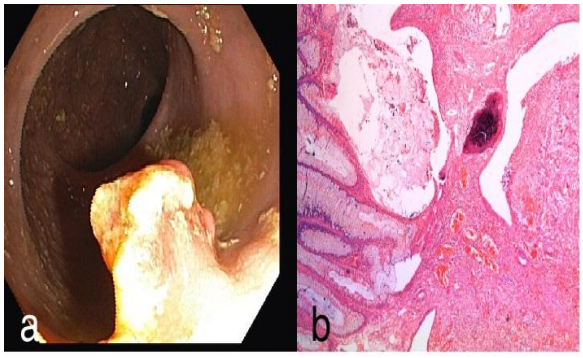


**Fig 1: Case 1 a) Endoscopic picture of polyp measuring 1cm with surface ulceration**
**B) Microscopy of the juvenile polyp with osseous metaplasia (H&E, 100 X)**

**CASE 2**

A two year old girl presented to the OPD with complaints of bleeding per rectum of two months duration. Colonoscopy showed a pedunculated polyp measuring 1.5x0.5cm with surface ulceration (Figure 2a). This was snared and cauterized and sent for histopathological examination.

Grossly it was a polyp measuring 1.5x0.5cm with a stalk of 0.5 cm. Microscopic examination showed ulceration and edematous lamina propria infiltrated by mononuclear cells. Glands were dilated and showed cystic changes with secretions .Osseous metaplasia composed of mature bony trabeculae was seen and the stalk was unremarkable. The case was reported as a juvenile rectal polyp with foci of osseous metaplasia (Figure 2b).

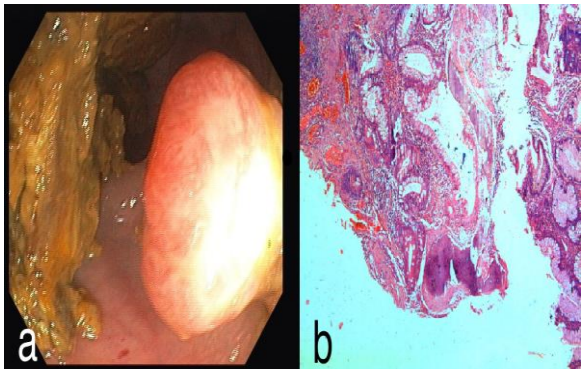


**Fig 2: Case 2 a) Endoscopic picture of polyp measuring 2cm**
**b) Microscopy of polyp with osseous metaplasia. Inflammatory infiltrates and hemorrhagic areas seen (H&E,100 X)**

**CASE 3:**

A four year old boy presented with bleeding per rectum of one month duration. Colonoscopy showed a small polyp less than 1 cm with a lobulated surface (Figure 3a). This was removed and cauterized and sent for histopathological examination

Gross examination showed a rectal polyp measuring 1 cm. Microscopy showed edematous lamina propria with focal ulceration. Lamina propria showed mononuclear cells and acute inflammatory cell infiltration. Glands were dilated having secretions. A focus of osseous metaplasia composed of normal looking osseous trabeculae was present. Features were in favor of a juvenile rectal poly with foci of osseous metaplasia (Figure 3b).

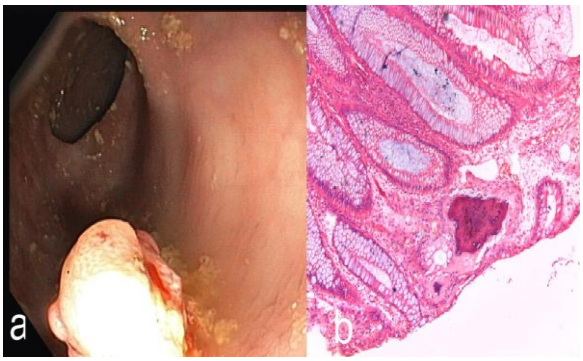


**Fig 3: Case 3 a) Endoscopic picture of the polyp measuring 1cm**
**b) Microscopy of the juvenile polyp with osseous metaplasia (H&E, 100X)**

**CASE 4**

A male aged sixty three was admitted with bleeding per rectum of fourteen days and loose stools of seven days duration. Examination revealed a soft abdomen with audible bowel sounds. On colonoscopy, an ulcero proliferative lesion in mid sigmoid region was found. CT abdomen showed a circumferential neoplasm of 6-7 cm with moderate luminal narrowing without obstruction (Figure 4a). Biopsy was done and report was given as adenocarcinoma with osteoid metaplasia (Figure 4b). Later, patient underwent exploratory laparotomy with radical sigmoidectomy and colorectal anastomosis. Sample was sent for histopathological examination.

Rectosigmoid segment of 15cm in length was received in our department. Ulceroproliferative lesion in the midsigmoid measured 6cm in length (Figure 5a). Cut section showed greyish white area involving the whole thickness reaching the serosa. Microscopy revealed a moderately differentiated adenocarcinoma. There was a focus of osteoid metaplasia composed of mature bony trabeculae (Figure 5b). Lymphatic invasion was present involving 7/12 lymph nodes salvaged.

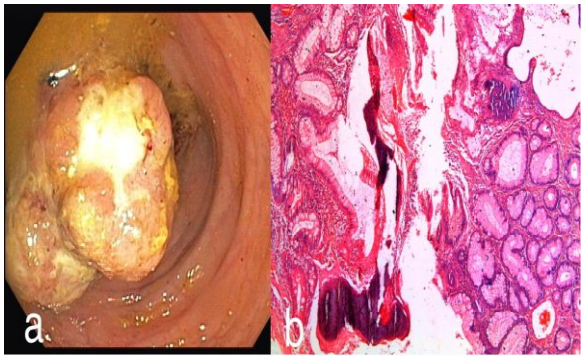


**Fig 4: Case 4 a) Endoscopy showing an ulceroproliferative lesion in the midsigmoid region**
**b) Biopsy showing adenocarcinoma with osseous metaplasia (H&E, 100X)**

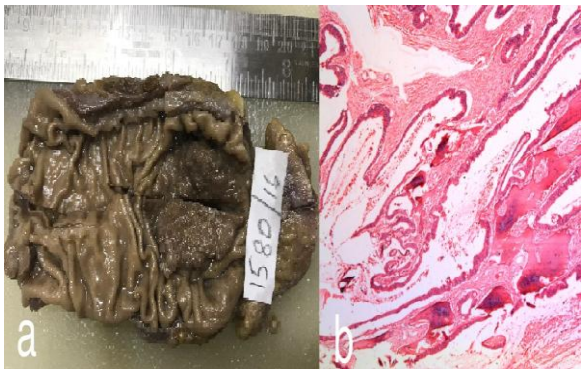


**Fig 5: Case 4: a) Sigmidectomy specimen measuring 10 cm in length with an ulceroproliferative growth of 5cm**
**b) Microscopy of adenocarcinoma showing osseous metaplasia (H&E, 100X)**

## DISCUSSION

OM has been described in a range of pathological entities including trauma, benign and malignant tumors. It has been described in renal cell carcinoma, hepatocellular carcinoma, lung adenocarcinoma, bronchi of older patients, carcinoma of endometrium and skin cancers. In stomach, metaplastic bone formations have been reported in hyperplastic polyps carcinoids and adenocarcinomas. It has been found in primary sites as well as in the secondary sites in malignancies. [1] The incidence of osseous metaplasia in GI tract is only 0.4% as described by Dukes etal. [1] Juvenile rectal polyps are rare in occurrence and those with metaplastic changes are still rarer. [3] We encountered three cases in our Centre. There was a male preponderance and all were above two years of age. Osseous metaplasia in juvenile polyp was first described by Sperling in 1981. [3] Similar cases were reported by Bhattacharya et al, Garg et al and Rashida et al. [4-6] Osteoid metaplasia in juvenile polyp is very rare and it has no clinical and prognostic significance. Osseous metaplasia has been described in inflammatory polyp, serrated adenoma and tubular adenoma. [7-10]

Gruber in 1913 was first to describe heterotopic ossification in a gastric adenocarcinoma. [9] It was in 1923, Hasegawa described 2 cases of rectal carcinoma with bone formation. [1] Since then only few cases (about 20) have been described in colorectal carcinoma, though being one of the most common malignancies. Osseous metaplasia is most frequently associated with Mucinous tumors. Haques et al reported four cases of metaplastic bone formation in GI tract, one in Barrett's oesophagus and three cases were mucin producing tumors of appendix, transverse colon and rectum. [6] Mucin is thought to prompt a stromal reaction and ossification. Our case did not have mucin production. Mogoanta etal and Sharmila Devi etal have described osseous metaplasia in rectal adenocarcinoma. [1,9] Osseous metaplasia has not shown to alter the prognosis of the patient.

Various mechanisms have been suggested regarding pathogenesis of osseous metaplasia. Marks and Atkinson in 1964 postulated that osseous metaplasia may be due to the metaplastic transformation of fibroblasts to osteoblasts. [5] Imai et al described the role of bone morphogenetic protein, BMP-2, 4, 5, 6 in the pathogenesis of osseous metaplasia. [2] Rhone and Horowitz in 1976 postulated that the metaplasia of pluripotent mesenchymal cells into osteoblasts is under the influence of factors generated by epithelial cells. One possible mechanism includes the production of bone morphogenetic proteins BMPP-5, BMP-6 by epithelial cells and BMP-2 and BMP-4 by adjacent fibroblasts. [1] Mucin extravasation, chronic and active ulceration, necrotic tissue and stromal fibroblast proliferation have been associated with OM. [7] BMP released from rapidly dividing

epithelial cells of tumors have been suggested in adenocarcinoma. In non-neoplastic polyps, osteogenetic stimulation is thought to be from inflammatory process. In vitro studies in mouse and human fibroblast cultures show that four transcription factors oct ¾, Sox-2,C-myc, klf4 are related to the generation of pluripotent stem cells from stromal fibroblasts. The in vivo mechanism is largely unknown. [4] The exact pathogenesis remains obscure and requires further study.

## CONCLUSION

Only single reports of osseous metaplasia in juvenile polyp have been published so far. We have described three cases in juvenile polyp and one in adenocarcinoma. Heterotopic ossification has been an incidental finding on microscopy often. Histopathologists should be aware of this entity to avoid an over diagnosis of carcinosarcoma which carries a worse prognosis. The bone is always histologically benign. The phenomenon of heterotopic ossification does not have clinical significance. However, diagnosis of carcinoma should be borne in mind when calcification of GI tract is detected radiographically.

**No Conflict of Interests**

**Authors' Contributions:**

Dr Shamila Mohamed Ali has contributed, analysed, interpreted the literature data and prepared the manuscript.

Dr Reddy Polu Somasekhara has contributed to the conception and design of the study, manuscript review and has given final approval of the version. He is the guarantor of the work.

Dr Kannam Ramaswamy has contributed the clinical details and expertise in these cases

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How to cite this article: Ali SM, Somashekara RP, Ramaswamy K. Heterotopic ossification in gastrointestinal neoplasms: a rare occurrence. Int J Health Sci Res. 2018; 8(8):340-343.

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