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Rectal Adenocarcinoma with Osseous Metaplasia

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A rare case of osseous metaplasia in a moderately differentiated rectal adenocarcinoma in a 79-year-old man is reported. A possible pathogenesis of this phenomenon is discussed.

Introduction

Osseous metaplasia is a very rare finding in gastrointestinal malignances. Dukes, who first described osseous metaplasia in rectal adenocarcinomas in 1939, estimated its incidence in these tumors as less than 0.4%, but to date only about 20 cases have been reported in the world literature [1, 2, 4, 11].

We describe a case of osseous metaplasia in a rectal adenocarcinoma revealed in routine pathology material in a 79-year-old man.

A Case Description

A 79-year-old man was admitted to the General Surgery Ward with a one-week history of abdominal pain, constipation, distension and vomiting. The results of physical examination and abdominal ultrasound suggested bowel obstruction. Partial rectosigmoidectomy with colostomy was performed due to a rectal tumor. The patient remained well 16 months after the operation.

The surgical specimen consisted of a 15cm-long segment of the large intestine with a $4.0 \times 5.0 \times 0.8$ cm superficially ulcerated, whitish tumor nearly completely encircling the lumen of the rectum and situated 3cm from the distal resection margin.

Histologically, the tumor was a moderately differentiated adenocarcinoma extending through the muscularis propria into the adjacent perirectal adipose tissue with lymphatic invasion, abundant fibrovascular stroma and sparse peritumoral lymphocytic infiltration. The resection margins were free of tumor tissue. Two of ten perirectal lymph nodes showed metastases (pT3N1). Many irregular spicules of the woven bone at varying stages of development were haphazardly distributed within the tumor (Fig. 1). They were composed of the osteoid with lacunar spaces containing osteocytes and surrounded by the rim of osteoblasts (Fig. 2). Some spicules were



Fig. 1. Partly calcified bony spicule infiltrated by rectal adenocarcinoma. HE. Magn. $50\times$.



Fig. 2. A spicule embedded in hypercellular stroma with transition of undifferentiated mesenchymal cells into osteoblasts resembling intramembranous ossification. HE. Magn. 200×.

located within the area of hypercellular stroma, and a distinct transition from undifferentiated mesenchymal cells to osteoblasts was observed. Cancer infiltration of osteoid depositions was noted, but the majority of spicules were located at a large distance from the carcinomatous glands. Some spicules were calcified. Hemopoietic tissue was not revealed. There was no ossification in the metastases to perirectal lymph nodes.

Discussion

Osseous metaplasia is a rare phenomenon in the gastrointestinal tract. It was reported in gastric, appendiceal and colorectal adenocarcinomas, either in primary tumors or in metastases, in rectal adenomas, as well as in gastric carcinoids [1–4, 8, 9, 11]. Heterotopic bone was also found in some non-neoplastic conditions, including juvenile, hyperplastic and Peutz-Jeghers polyps, Barrett's esophagus and solitary rectal ulcer syndrome [1, 4–7].

The pathogenesis of this condition is unclear. The formation of bone spicules is probably the consequence of mesenchymal precursor cell metaplasia; these cells transform into osteoblasts capable of osteoid production [11]. The stimuli responsible for this abnormal pathway of differentiation are unknown. It seems that some growth factors, e.g. TGF 1 and 2 or other paracrine factors locally secreted by epithelial (neoplastic or non-neoplastic) or inflammatory cells, may play an important role in this phenomenon [10]. Alkaline phosphatase revealed by Randall et al. [8] in osteoblasts and the surrounding mesenchymal and cancer cells is probably involved in mineralization of newly formed spicules, but its role in stimulation of ectopic bone formation is unknown. It was stressed that osseous metaplasia-like dystrophic calcification frequently occurred in the close vicinity of tumor necrosis, extracellular mucus or squamous metaplasia [11]. A repeated local trauma or special properties of rectal mucosa were also considered as predisposing factors [6].

It seems that osseous metaplasia is a lesion without any clinical significance. However, it could be misinterpreted as

bone invasion of adenocarcinoma or an extremely rare neoplasm in the colorectum – a carcinosarcoma, especially in small biopsy specimens.

References

- 1. *Ansari MQ, Sachs IL, Max E, Alpert LC:* Heterotopic bone formation in rectal carcinoma: case report and literature review. Dig Dis Sci 1992, 37, 1624–1629.
- 2. Dukes CE: Ossification in rectal cancer. Proc R Soc Med 1939, 32, 1489–1494.
- Groisman GM, Benkov KJ, Adsay V, Dische MR: Osseous metaplasia in benign colorectal polyps. Arch Pathol Lab Med 1994, 118, 64–65.
- Haque S, Eisen RN, West AB: Heterotopic bone formation in the gastrointestinal tract. Arch Pathol Lab Med 1996, 120, 666–670.
- Kaushik VY, Moriarty KJ, Lipscomb GR, Bisset DL, Wells S: Osseous metaplasia associated with solitary rectal ulcer syndrome. CME J Gastroenterol Hepatol Nutr 2001, 4, 37–39.
- Nakajama H, Iwane S, Mikami T, Nara H, Yamagata K, Morita T, Yagihashi S: Osseous metaplasia in benign rectal polyps. J Clin Gastroenterol 1997, 25, 558–559.
- Narita T, Ohnuma H, Yokoyama S: Peutz-Jeghers syndrome with osseous metaplasia of the intestinal polyps. Pathol Int 1995, 45, 388–392.
- Randall JC, Morris DC, Tomita T, Clarke Anderson H: Heterotopic ossification: a case report and immunohistochemical observation. Hum Pathol 1989, 20, 86–88.
- Sampsei JW, Callaway F: Gastric carcinoid with ossification. Am J Surg 1972, 124, 108–111.
- Teot LA, O'Keefe RJ, Rosier RN, O'Connell JX, Fox EJ, Hicks DG: Extraosseous primary and recurrent giant cell tumours: transforming growth factor- 1 and - 2 expression may explain metaplastic bone formation. Hum Pathol 1996, 27, 625-632.
- Van Patter HT, Whittick JW: Heterotopic ossification in intestinal neoplasms. Am J Pathol 1955, 31, 73–91.

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