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CASE REPORT

Three synchronous gastrointestinal tumours

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Abstract Mucinous cystadenomas of the appendix are rare, with significant malignant potential [Lo and Sarr in Hepatogastroenterology 50(50):432–437, 2003]. Carcinoid tumours are similarly uncommon, rarely occurring in Meckel's diverticula but are still the commonest tumour of Meckel's diverticulae [Nies et al. in Dis Colon Rectum 35(6):589–596, 1992; Modlin and Lye in Cancer 97(4):934–959, 2003; Sutter et al. in Schweiz Med Wochenschr Suppl 89:20S–24S, 1997; Weber and McFadden DW in J Clin Gastroenterol 11(6):682–686, 1989]. A 77-year-old woman presented to our clinic with a 6-week history of non-specific lower abdominal pain. A pelvic ultrasound showed an 8 × 3 × 2.5 cm mass in the right iliac fossa. Colonoscopy and CT confirmed this mass and also revealed a left colonic tumor. At laparotomy, three tumours were identified; in the appendix, a Meckel's diverticulum, and the descending colon. A subtotal colectomy and diverticulectomy were performed. Histology confirmed a T3N0 Dukes B colonic adenocarcinoma, a carcinoid of Meckel's diverticulum and a mucinous adenoma of the appendix. The patient is tumor free to date, 5 years after presentation. The presence of three synchronous tumours of different histological origin in the gastrointestinal tract has not previously been described in the literature.

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Introduction

Synchronous tumours of the colon are considered reasonably common, being identified in up to 20% of colonic resection segments. Carcinoid tumours are rare, and the presence of a carcinoid tumour in Meckel's diverticulum is so rare, that to date, less than 120 cases have been described in the literature [1, 2]. Carcinoid tumours are neuroendocrine tumours originating from the neural crest and can be diagnosed at any age [3]. Depending on the size of the carcinoid, metastasis of these tumours is common. There is also evidence that carcinoid tumours are associated with other gastrointestinal tumours, including adenocarcinomas and ovarian tumours [4–6]. Mucinous adenomas of the appendix are similarly rare being thought to arise from the mucinous metaplasia of peritoneal inclusion cysts, with mucinous adenomas being identified in only 0.6 % of all appendiceal resections [6, 7]. We report a patient who had three different, independent gastrointestinal tumours, each of differing histological origin, which to the best of our knowledge has not previously been reported in the literature.

Case report

A 77-year-old woman was admitted to our hospital following a 6-week history of non-specific lower abdominal pain, which was severe but intermittent. She had a recent change in bowel habit with episodes of diarrhoea, without

weight loss, a change in appetite, or urinary symptoms. She had a history of pulmonary fibrosis but was asymptomatic in this regard. There was also a family history of gastric cancer.

On examination, the abdomen was soft and non-tender, with a palpable mass in the right iliac fossa (RIF), which was freely mobile. Digital rectal examination was normal. Blood tests showed a raised CEA of 5.37 ng/ml. Plain film of the abdomen showed a radio-opaque shadow in the right hemi-pelvis. Pelvic ultrasound scan was performed which identified an 8 × 3.3 × 2.5 cm oblong cystic structure superficially in RIF showing some calcification at its cephalic end. Its appearance was non-specific, and was thought to be a mesenteric cyst. Colonoscopy showed an unsuspected annular mucosal lesion of the descending colon, which was biopsied. A barium enema was performed to evaluate the proximal colon and showed a 4.5 cm long "apple-core" lesion in the mid-descending colon (Fig. 1a). CT of the abdomen and pelvis showed an 8 × 3.5 cm calcified, thin-walled and well-demarcated mass, and the descending colon tumour (Fig. 1b, c). Histology confirmed an invasive adenocarcinoma, and surgery was undertaken.

Laparotomy was performed and at this time a large appendiceal mass, along with a 2.5 cm tumour in a Meckel's diverticulum and a 5 cm long carcinoma of the descending colon were identified. A subtotal colectomy with Meckel's diverticulectomy was performed. Histology confirmed a T3N0 Dukes B adenocarcinoma of the colon, a neuroendocrine carcinoma in a Meckel's diverticulum, and a mucinous adenoma of uncertain malignant potential of the appendix. The patient was followed up for 5 years clinically, by CT, tumour markers, and colonoscopy, with no evidence of recurrence.

Pathological findings

The resected surgical specimens comprised a 45 cm length of large intestine with the 8 × 3.3 × 2.5 cm appendiceal mass, and a wedge resection of small bowel with a Meckel's diverticulum measuring 2.5 cm situated on its anti-mesenteric border. Histological examination of the two specimens, showed a Meckel's diverticulum containing a carcinoid tumour (Fig. 2a). This tumour was composed of a rounded mass of closely packed tumour cells showing peripheral palisading and rosette formation. There was moderate nuclear pleomorphism and mitoses identified. The tumour had invaded the bowel wall beyond the muscularis propria and was present at the serosal surface.

The colonic specimen showed an invasive moderately differentiated adenocarcinoma of 5 cm in length, located 9 cm from the distal resection margin of the specimen, which invaded through the muscularis propria (Fig. 2b). The resection margins were free of tumour and there was no peritoneal involvement. Lymphovascular space invasion was absent and 15 regional lymph nodes were negative for tumour.

The appendiceal lesion was a mucinous tumour with mild epithelial atypia and pools of mucin dissecting

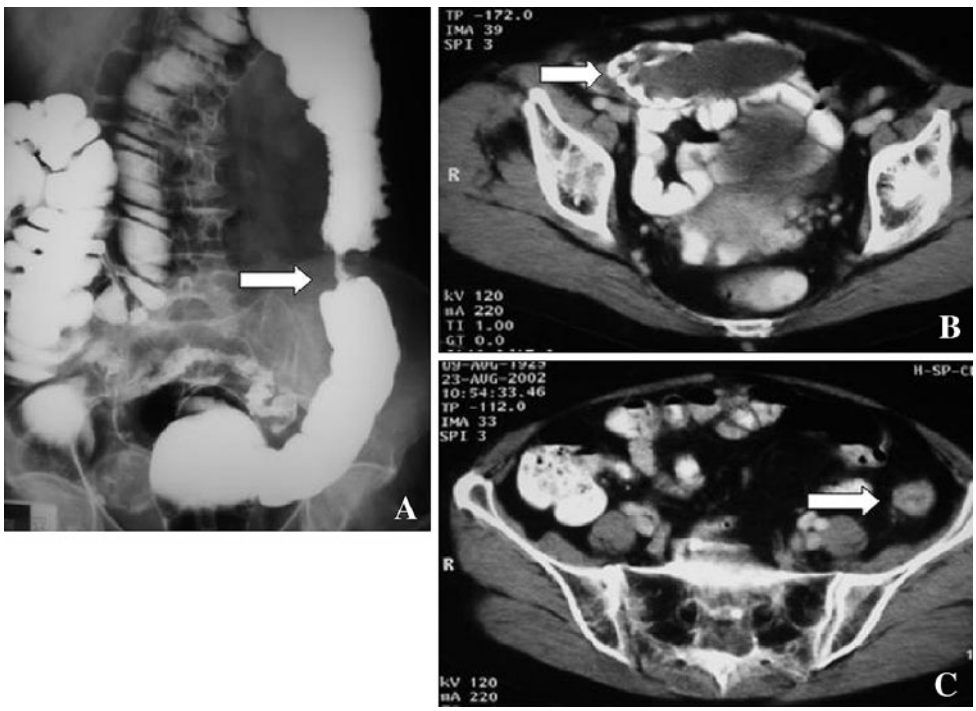

Fig. 1 **a** Barium enema showing 4.5 cm "Apple-core" lesion of descending colon (*arrow*). **b** CT scan of abdomen showing large calcified mass in the pelvis (*arrow*). **c** CT scan showing descending colon tumour (*arrow*)

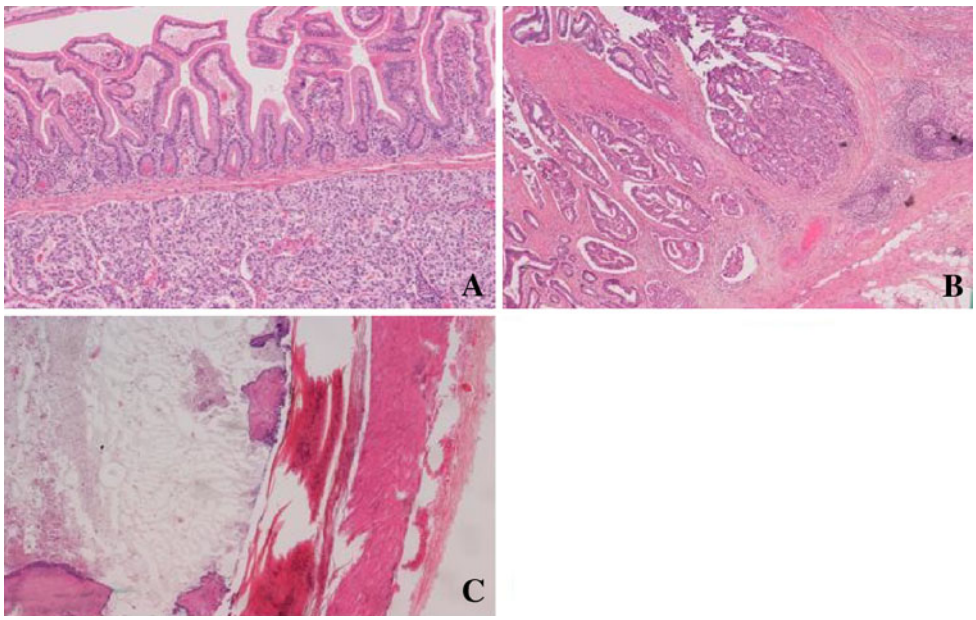

Fig. 2 **a** Carcinoid tumour of Meckel's diverticulum (H&E, ×400) showing closely packed cells with peripheral palisading and rosette formation, **b** Adenocarcinoma of colon (H&E, ×100) showing invasion through the muscularis propria and **c** Mucinous cystadenoma of appendix (H&E, ×40) showing mucin within the lumen of the appendix

through the wall and was classified as a mucinous tumour of the appendix of uncertain malignant potential (Fig. 2c).

Discussion

In this unusual case of three tumours, only two of the tumours had been identified prior to surgery. The tumour in the descending colon had been identified radiologically and verified with a histological diagnosis at colonoscopy. CT had identified the calcified mass in the RIF, which increased the likelihood of malignancy; however, tumour type or histological origin was not known. At surgery, the decision was made to perform a subtotal colectomy as the risk of a mucinous cystadenoma, and the consequent development of pseudomyxoma peritonii, along with the presence of a confirmed adenocarcinoma, was felt to be too great to perform more localised resections. The mucinous adenoma was successfully removed with no evidence of spillage. The colonic carcinoma had excision margins 9 cm above and below the lesion. As optimal management of this tumour would suggest a right hemi-colectomy [8], a subtotal colectomy was deemed the most appropriate management for the presence of these two synchronous tumours.

Concurrently, a third tumour, the carcinoid tumour of Meckel's diverticulum, was identified and resected. Similar to the cystadenoma, histological diagnosis was unknown, but carcinoid tumours are the most common malignant tumours of Meckel's diverticulum with two thirds of patients remaining asymptomatic at initial diagnosis [2]. Tumour size influences the likelihood of metastases as does the gender, but with early metastases even in small tumours, aggressive surgical management is justified [1, 2, 7, 9].

In this patient, surgical management with close follow-up was felt to be appropriate as at presentation there was no evidence of carcinoid syndrome, any evidence of metastases on CT, or lymph node invasion identified on histology.

This case presented a very unusual collection of tumours, each of which presented its own challenges in appropriate management, and the synchronous occurrence of these three histologically separate tumours has to the best of the authors' knowledge not previously been reported.

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References

1. Nies C, Zielke A, Hasse C, Ruschoff J, Rothmund M (1992) Carcinoid tumors of Meckel's diverticula. Report of two cases and review of the literature. Dis Colon Rectum 35(6):589–596. doi:10.1007/BF02050541
2. Sutter PM, Canepa MG, Kuhrmeier F, Marx A, Martinoli S (1997) Carcinoid tumor in Meckel's diverticulum: case presentation and review of the literature. Schweiz Med Wochenschr Suppl 89:20S–24S
3. Weber JD, McFadden DW (1989) Carcinoid tumors in Meckel's diverticula. J Clin Gastroenterol 11(6):682–686. doi:10.1097/00004836-198912000-00018
4. Grossmann I, Akkersdijk GJ (2003) Carcinoid tumor in a Meckel's diverticulum: hypothesis on mutual embryological origin. Int Surg 88(1):41–46
5. Habal N, Sims C, Bilchik AJ (2000) Gastrointestinal carcinoid tumors and second primary malignancies. J Surg Oncol

75(4):310–316. doi:10.1002/1096-9098(200012)75:4<306::AID-JSO14>3.0.CO;2-3
6. Kothari T, Mangla JC (1981) Malignant tumors associated with carcinoid tumors of the gastrointestinal tract. J Clin Gastroenterol 3(Suppl 1):43–46. doi:10.1097/00004836-198100031-00009
7. Lo NS, Sarr MG (2003) Mucinous cystadenocarcinoma of the appendix. The controversy persists: a review. Hepatogastroenterology 50(50):432–437
8. Barry M, Collins CG, McCawley N, McGuinness J, Leahy AL (2007) Synchronous appendiceal tumours. Surgeon 5(2):111–113
9. Shebani KO, Souba WW, Finkelstein DM, Stark PC, Elgadi KM, Tanabe KK, Ott MJ (1999) Prognosis and survival in patients with gastrointestinal tract carcinoid tumors. Ann Surg 229(6):815–821. doi:10.1097/00000658-199906000-00008 discussion 822–813
10. Modlin IM, Lye KD, Kidd M (2003) A 5-decade analysis of 13,715 carcinoid tumors. Cancer 97(4):934–959. doi:10.1002/cncr.11105