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Case Report

Synchronous Primary Anorectal Melanoma and Sigmoid Adenocarcinoma: A Case Report

Aaron Lim[1], Benjamin Grant[1], John Avramovic[1,2], Yik-Hong Ho[1,2], Corrine Wallace[3]

[1]*Department of Surgery, Mater Hospital Pimlico, Townsville, Australia*

[2]*School of Medicine & Dentistry, James Cook University, Douglas, Townsville, Australia*

[3]*Department of Anatomical Pathology, Sullivan Nicolaides Pathology, Mater Hospital Pimlico, Townsville, Australia*

Synchronous primary anorectal melanoma and colorectal adenocarcinoma is extremely rare, with only 5 cases reported in the literature. Here, a case is reported and the currently available literature is summarized. A 72-year-old white male presented with changes in his bowel habits and unintentional weight loss. Colonoscopy revealed a polypoid mass in the distal rectum extending to the anal verge anteriorly and a circumferential polypoid mass in the distal sigmoid colon. Biopsies were taken, which revealed poorly differentiated melanoma of the anorectal mass and moderately differentiated adenocarcinoma of the sigmoid mass with nodal involvement. Computed tomography of the abdomen showed liver metastasis. An extended abdominoperineal resection was undertaken for palliation, relief of symptoms, and definitive histology to guide further management. Consequently, a resection of the hepatic metastasis was attempted; however, macroscopic deposits were discovered on 7 of 8 liver segments perioperatively. He was subsequently referred to medical oncology for palliative chemotherapy. Synchronous primary anorectal melanoma and colorectal adenocarcinoma is rare, this being the sixth report found in the literature. In summary of the available cases, all synchronous cancers were located in the rectosigmoid and had very similar presentations. Most presented relatively late and were generally treated with abdominoperineal resection, which appears to be the best treatment option. Overall, prognosis appears to be dismal. General and colorectal surgeons should always be aware of the possibilities of simultaneous primary cancers because this can affect treatment modalities and prognosis for the patient.

Corresponding author: Aaron E Lim, BPhrm, MBBS(Hons), Department of Surgery, Mater Hospital Pimlico, 21–37 Fulham Road, Pimlico, QLD 4812, Australia.
Tel.: +61421351893; Fax: +61730148788; E-mail: aaron.lim@uqconnect.edu.au

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Anorectal melanoma is rare, accounting for less than 1% of all melanomas and less than 3% of all anorectal cancers.[1–3] Multiple primary colorectal cancers are also uncommon, the incidence of which is estimated to be about 2.5% to 5% of all colorectal cancers.[4–6] As such, synchronous primary anorectal melanoma and primary colorectal adenocarcinoma is exceedingly rare, with only 5 cases reported in the literature.[2] Here, a case of synchronous primary anorectal melanoma and adenocarcinoma of the distal sigmoid colon, and a review of the literature are presented.

Case Report

A 72-year-old white male presented in early January 2014 with changes in his bowel habits and unintentional weight loss. Specifically, he complained of blood and mucus in his stools, fecal incontinence, changes in the frequency of his motions, tenesmus, anorexia, and 10-kg weight loss during the previous 4 months. His past medical history included hypertension and gout. He denied any family history of colorectal cancers. He also had a 40–pack year history of smoking and drank approximately 10 standard drinks a day.

Colonoscopy revealed a polypoid mass in the distal rectum extending to the anal verge anteriorly and a circumferential polypoid mass in the distal sigmoid colon. Biopsies were taken, which revealed poorly differentiated melanoma of the anorectal mass and moderately differentiated adenocarcinoma of the sigmoid mass. Computed tomography (CT) of the abdomen showed a hypodense lesion in segment VI of the liver which was highly suspicious of metastasis (Fig. 1).

He subsequently underwent an extended abdominoperineal resection (APR) for palliation, relief of symptoms, and definitive histology to guide further management. Perioperatively, the anorectal mass was found to be approximately 7×6 cm, with some invasion into the adjacent structures, likely invading anteriorly into the prostate. The sigmoid mass was approximately 5×4 cm and did not involve or invade the adjacent structures.

Histopathology from the extended APR confirmed synchronous low-grade adenocarcinoma (Fig. 2) of the distal sigmoid and anorectal malignant melanoma (Fig. 3), with lymph node involvement by both malignancies. Immunohistochemistry performed on the anorectal tumor showed positive staining with S100 and pan-melanoma marker, whereas cytokeratin AE1/AE3 stained the surrounding normal intestinal mucosa but did not stain the tumor cells.

Postoperatively, the patient's recovery was uneventful and he was consequently referred for potential resection of the likely right hepatic metastasis. However, macroscopic deposits were discovered on 7 of 8 liver segments perioperatively, which did not appear to be visible on repeat CT of the abdomen after APR. Biopsies were taken and histopathology confirmed both metastatic melanoma and adenocarcinoma. He was subsequently referred to medical oncology for palliative chemotherapy.

Discussion

Anorectal melanomas are rare and exhibit biologic behaviors different from those of cutaneous melanomas.[1–3] They are more aggressive and have poorer long-term survival rates.[2] Differential diagnoses for anorectal melanoma include colorectal carcinoma, prolapsing rectal polyp, or thrombosed external haemorrhoids.[2] Preoperative diagnosis of anorectal melanoma may be difficult, and they can be misdiagnosed as a different type of neoplasm, as with the case of synchronous collision malignant melanoma and adenocarcinoma of the rectum reported by Dias *et al*.[7]

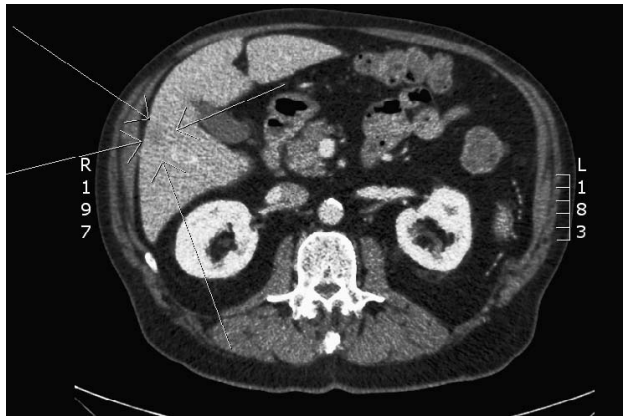

Fig. 1 Contrast-enhanced computed tomography showing a hypodense lesion in segment VI of the liver (arrows).

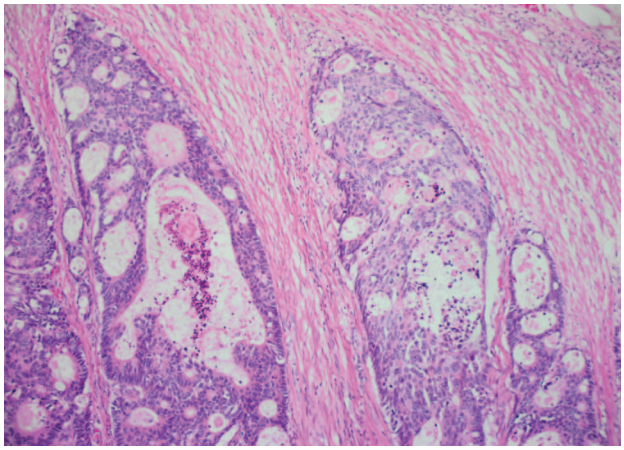

Fig. 2 H&E (×100) stained distal sigmoid lesion: moderate to well-differentiated adenocarcinoma.

Synchronous colorectal cancers are uncommon, occurring in 3% to 5% of patients, with an even lower incidence (about 2.5%) when patients with Lynch syndrome are excluded. [6] They are defined as 2 or more distinct primary tumors diagnosed within 6 months of an initial colorectal cancer, separated by normal bowel and not due to direct extension or metastasis.[4,5,8] Conversely, collision tumors are composed of 2 histologically distinct neoplasms coinciding at the same location. They come from 2 histogenetic events that occurred in proximity, and therefore these tumors have 2 different cell lineages that grow until these lesions get intimately juxtaposed.[7] As such, collision tumors were also included in the literature search because they were originally 2 separate primary tumors.

Primary anorectal melanoma has been very rarely described with concomitant or synchronous primary colorectal adenocarcinoma.[2] In searches of the PubMed, Google Scholar, and Medline databases using the search terms "colorectal melanoma," "adenocarcinoma," "synchronous neoplasms," and "collision neoplasms," alone and in various combinations, we found 5 reported cases of synchronous or collision primary anorectal melanoma and colorectal adenocarcinoma, with the full text from one report unattainable and only available in Europe.

Patients reported were all older than 50 years, with a slightly higher female predominance.[2,3,7,9] Most synchronous cancers presented with similar symptoms of rectal bleeding and changes in bowel habits.[2,3,7] All synchronous colorectal cancers were located either in the rectum or the sigmoid.[2,3,7,9] This may be accounted for by the fact that most colorectal adenocarcinomas are found in the rectum or

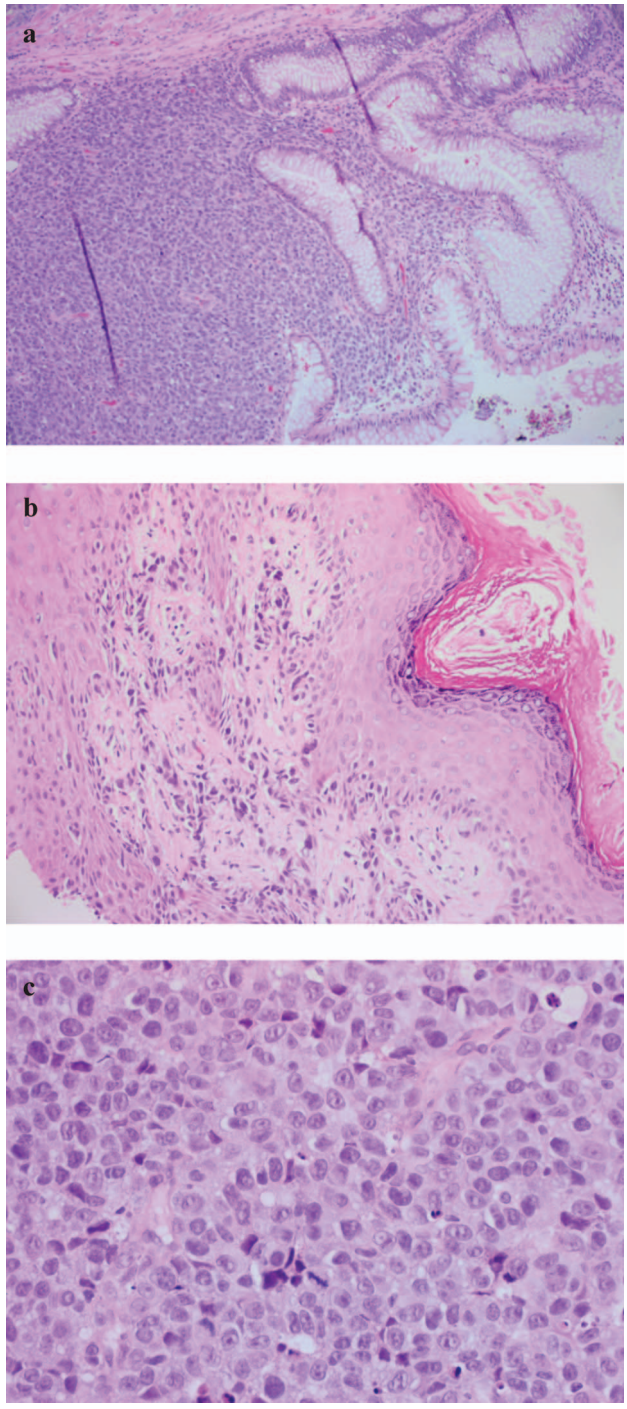

Fig. 3 H&E-stained anorectal lesion. (a) Sheets of poorly differentiated cells (×100). (b) Atypical lentiginous proliferation of melanocytes (×200). (c) Higher-powered view of melanoma (×400).

sigmoid. Most patients, including the one reported here, presented relatively late with distant metastasis, perhaps because of the aggressive nature of anorectal melanoma compared with cutaneous

melanoma.[2,7] All but one case was managed by APR.[7]

There is no consensus at this moment regarding which surgical approach is preferred for the treatment of synchronous primary anorectal melanoma and colorectal adenocarcinoma, partly because of its rarity. However, there are a few opinions concerning the surgical treatment of anorectal malignant melanoma. First, wide local excision (WLE) is often considered a procedure of choice because of the high morbidity and mortality rates and lack of survival advantage of APR.[1,10,11] Second, APR can improve locoregional control rates, which can improve prognosis. Because WLE has less morbidity than APR and avoids the necessity of colostomy, most authors consider it as the first choice of treatment for anorectal malignant melanoma when it is technically feasible.[1,10,11]

Nevertheless, although studies have shown that WLE provides a better quality of life, neither WLE nor APR provides a survival benefit.[1,10,11] One study showed that WLE of anorectal melanoma can result in increased local recurrence compared with APR; however, these results have not been duplicated.[12] Furthermore, it has been shown that in patients with stage IV disease, such as our case patient, APR can reduce bleeding or colonic obstruction, resulting in improved quality of life.[11] Because of the fact that many patients present with late disease involving the sigmoid colon, an APR appears to be an appropriate choice in treating synchronous primary anorectal melanoma and colorectal adenocarcinoma.

The mean survival time for anorectal malignant melanoma is 20 months with treatment, and disease-free survival of 6.7% to 12% at 5 years.[10,11] The mean 5-year survival rate of both metastatic anorectal melanoma and colorectal cancer is poor, being 0% and 5.7%, respectively.[10,11] It would have been useful if data on the prognosis of the patients in the case studies were reported along with their subsequent medical management. However, it can be extrapolated from the data on metastatic anorectal melanoma alone that prognosis is dismal. In addition, patient factors contributing to an increased risk of synchronous primary anorectal melanoma and colorectal adenocarcinoma were difficult to ascertain because of the limited amount of literature available and variability in the reporting of patient data.

In conclusion, synchronous primary anorectal melanoma and colorectal adenocarcinoma is rare, this being the sixth report found in the literature. Overall, all synchronous cancers were located in the rectosigmoid and had very similar presentations. Most presented relatively late and were generally treated with APR, which appears to be the best treatment option. General and colorectal surgeons should always be aware of the possibilities of simultaneous primary cancers because this can affect treatment modalities and prognosis for the patient.

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