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Endobronchial metastasis from primary anorectal melanoma

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Patient: Male, 64
Final Diagnosis: Metastatic anorectal melanoma with endotracheal metastasis
Symptoms: Fatigue • weight loss • hematochezia • cough
Medication: None
Clinical Procedure: Biopsy of anal mass • rigid bronchoscopy
Specialty: Internal medicine • oncology • pulmonology

Objective: Rare disease

Background: Anorectal melanoma is a rare cancer with a poor prognosis. The mean survival after diagnosis is 15–25 months. At the time of diagnosis, 61% of patients have local regional lymph node metastases, and 21% have distant metastases. The lungs are a common site for metastasis for all tumors including melanoma. However endobronchial metastasis is a rare phenomenon. Endotracheal metastases are even rarer, occurring in only 5% of patients with extrapulmonary endobronchial metastases. It is most commonly seen in breast, colorectal, and kidney cancers. It is extremely rare for cutaneous melanoma. The mean survival after diagnosis is only 15 months and treatment options are limited.

Case Report: We report the case of a 64 year-old gentleman with newly diagnosed metastatic anorectal melanoma. A 3 cm by 3 cm bluish-black, oval-shaped, exophytic mass protruding from his anus was found on physical exam. Endobronchial and endotracheal metastasis to the trachea were discovered on computed tomography and he was subsequently taken to the operating room for argon plasma coagulation laser recanalization of his trachea via rigid bronchoscopy, and resection of his anal mass.

Conclusions: We have presented the first known case of anorectal melanoma with endobronchial metastasis. Palliative APC laser recanalization was used to prevent asphyxiation from the endotracheal mass. Endobronchial metastasis is uncommon and can be easily mistaken for primary bronchogenic carcinoma. It should always be considered when evaluating patients with new lung masses.

Key words: anorectal • melanoma • endobronchial • metastasis • treatment • bronchoscopy

Full-text PDF: http://www.amjcaserep.com/download/index/idArt/889291

Background

Anorectal melanoma is a rare cancer with a poor prognosis. It accounts for less than 4% of all cancers originating from the anorectal region, and less than 1% of all melanomas [1]. It was first reported by Moore in 1857 [2]. Isolated case-reports and case-series have been reported since then. It mainly affects the elderly, typically between the sixth to eighth decade of life [3]. The tumor originates in the anorectal transition zone, above the dentate line, where melanocytes typically reside. In this area there are abundant lymphatics and blood vessels that allow the tumor to grow and metastasize quickly. At the time of diagnosis, 61% of patients have local regional lymph node metastases, and 21% have distant metastases [4]. It comes as no surprise that the mean survival after diagnosis is 15–25 months [5]. Furthermore, the presenting symptoms are usually non-specific and include bleeding, tenesmus, pruritus, pain, and change in bowel habits. This leads to a delay in diagnosis [6]. New advances have implicated the KIT receptor tyrosine kinases in playing an integral role in the development and function of melanocytes and subsequent development of melanoma [7].

The lungs are one of the most common sites for metastatic disease from all solid tumors, including anorectal melanoma. While pulmonary metastasis is common, endobronchial metastasis is a rare phenomenon. The frequency of endobronchial metastasis has been disputed in the literature. King and Castleman reported that 18% of patients had tumor infiltrating bronchi.8 Later, Braman and Whitcomb found the incidence to be 2%, in their own autopsy series. 8 This discrepancy is a result of the different definitions used for endobronchial metastasis. King included tumors with bronchial invasion, while Braman only included metastasis that developed in the bronchial epithelium, usually via the mucosal lymphatics. 8,9 Furthermore, while extrapulmonary endobronchial metastases are very infrequent, endotracheal metastases are even more rare, occurring in only 5% of patients with extrapulmonary endobronchial metastases [10].

The most common cancers that can cause endobronchial metastases are breast, colorectal, and kidney. Other extrathoracic malignancies such as melanoma, cervical, uterine, and bladder have also been reported [1]. Endobronchial metastasis is usually a late finding in the course of a patient's disease. The most common symptoms include cough, dyspnea, and hemoptysis [10]. The mean survival after diagnosis is only 15 months and treatment options are limited [10].

We present a case of a 64 year-old gentleman who was diagnosed with metastatic anorectal melanoma with numerous endobronchial metastases, including a nearly obstructing endotracheal mass. To our knowledge, this is the first reported case in the English literature of primary anorectal melanoma causing endotracheal metastasis.

Case Report

A 64 year-old Caucasian male with a past medical history significant for tobacco abuse, alcoholism, pancreatitis, and hemorrhoids was brought into the emergency department by his landlord because of concerns over progressive deterioration in his health. The patient stated that over the past six months he had become more fatigued and was having trouble carrying out activities of daily living. His condition had declined to the point where he could no longer walk to the bathroom because of generalized weakness. During this same time period, he had lost about 20 kg. He reported a non-productive cough, but denied dyspnea, hemoptysis, or chest pain. His only other complaint was that he had noticed a mass growing out of his anus, which intermittently bled and caused pain. He was still having regular bowel movements despite the mass. He was unsure if it was hemorrhoids and had been using Tucks medical pads for symptom control. On physical exam, he was a cachectic-appearing male with an approximately 3×3 cm bluish-black, oval-shaped, exophytic mass protruding from the anus. In addition, numerous bluish, firm, immobile, non-tender subcutaneous nodules were present throughout his chest, abdomen, and pelvis. Laboratory testing revealed a normocytic anemia and a carcinogenic embryonic antigen (CEA) was normal. A chest radiograph revealed scattered bilateral nodular opacities.

Over the course of the first day of hospitalization, he had increasing hypoxemia, tachycardia and relative hypotension. A repeat chest radiograph was unchanged and CT angiography (CTA) of the chest was performed to evaluate for pulmonary embolism. The CTA revealed widespread lymphadenopathy and tracheal, pleural, pulmonary, cutaneous and subcutaneous nodules most compatible with malignancy. In addition, there was debris in the bilateral mainstem bronchi extending inferiorly with occlusion of the bronchial tree to the level of the subsegmental lower lobe bronchi. Diffuse tree-in-bud opacities and nodular lung opacities were thought to represent endobronchial tumor (Figure 1). The following day, a CT of the abdomen and pelvis was performed to evaluate disease burden. There was extensive lymphadenopathy throughout the entire abdomen and pelvis with innumerable nodules in the peritoneum, retroperitoneum, paraspinous and gluteal muscles, and subcutaneous soft tissues concerning for metastases (Figure 2). There was also a large soft tissue, exophytic mass involving and extending outward from the anus, measuring 3.7×3.5 cm (Figure 3).

Due to concern for possible occlusion by the endotracheal mass, a flexible bronchoscopy was performed which revealed

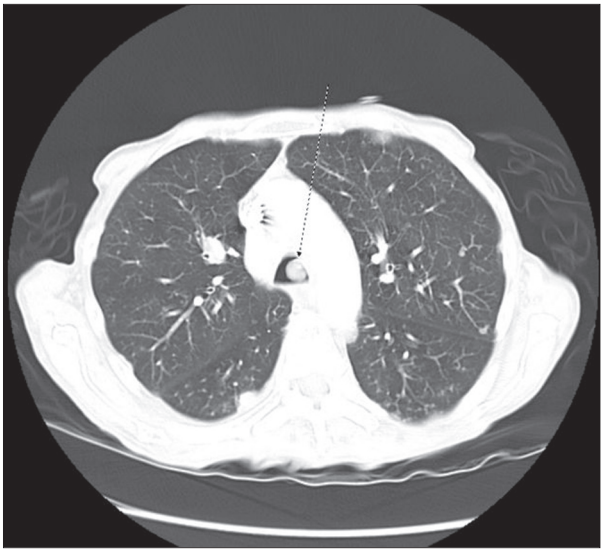

Figure 1. Chest CTA demonstrating endotracheal metastasis.

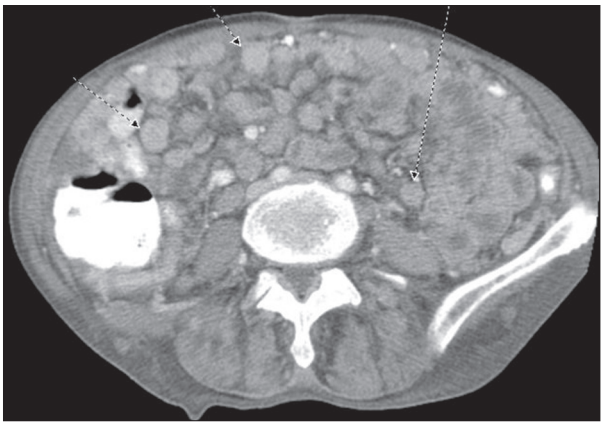

Figure 2. CT abdomen/pelvis demonstrating severe tumor burden.

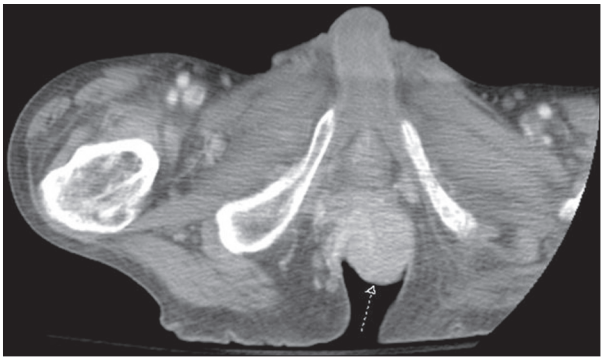

Figure 3. CT pelvis demonstrating primary anorectal melanoma.

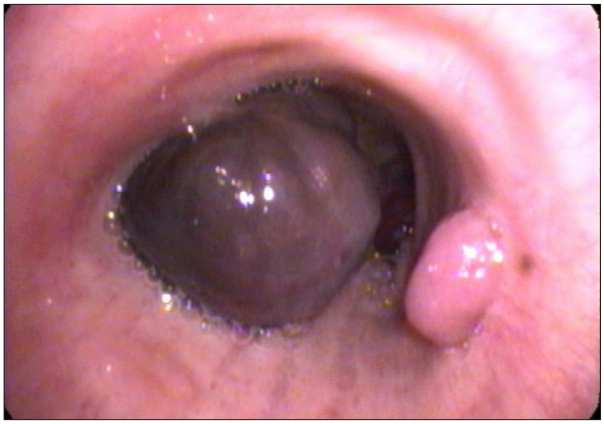

Figure 4. Endoscopic view of large endotracheal metastasis obtained during flexible bronchoscopy.

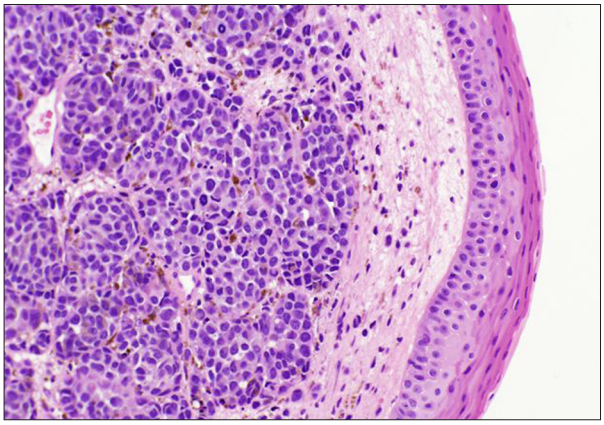

Figure 5. Metastatic tracheal biopsy hematoxylin and eosin stain at 20 times magnification.

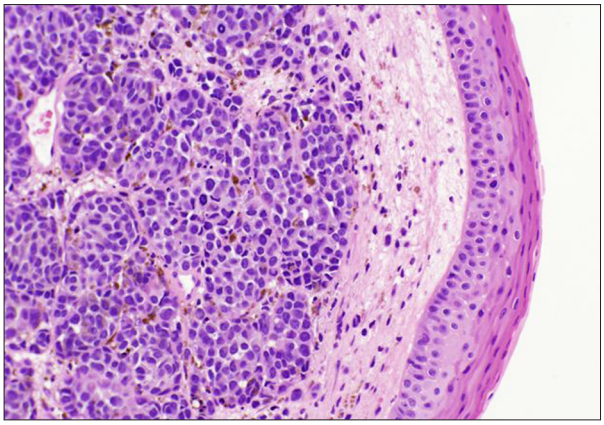

Figure 6. Anal Biopsy hematoxylin and eosin stain at 20 times magnification.

a large, hyperpigmented, pedunculated mass in the mid-lower trachea obstructing about 75% of the airway (Figure 4). A smaller hyperpigmented, polypoid lesion was also noted and biopsied. The right mainstem bronchus was almost completely occluded with a large, hyperpigmented pedunculated mass. The left mainstem bronchus was normal in appearance and diameter. The endotracheal mass was nearly obstructive and it was thus felt that he should undergo rigid bronchoscopy with resection of the mass to prevent complete occlusion of the airway.

General surgery was also consulted to evaluate his anal mass. Given the concern for malignancy they felt that a biopsy was warranted. On hospital day number five, he was taken to the operating room for rigid bronchoscopy with removal of the primary endotracheal mass using the barrel of the rigid bronchoscope, with argon plasma coagulation for hemostasis, followed by resection of his perianal mass. Both samples were sent for pathology which revealed metastatic melanoma. The endotracheal mass biopsy demonstrated bland squamous mucosa without evidence of junctional melanocytic atypia. There is a submucosal infiltrate of atypical epithelioid cells most consistent with a metastatic focus (Figure 5). In contrast, the perianal biopsy revealed squamous mucosa with junctional nests of atypical epithelioid melanocytes in addition to a submucosal infiltrate of atypical epithelioid nests. Both the cytological atypia and histological findings supported the anal mucosa as the primary site of tumor origin (Figure 6). Given his poor prognosis, the patient elected for palliative care, and was discharged from the hospital with home hospice. He died at home nine days following discharge.

Discussion

Despite advances in the understanding of the molecular basis for anorectal melanoma, prognosis is dismal. The only definitive therapy is surgical resection. There is debate as to which surgical approach is optimal, namely abdominoperitoneal resection versus local resection. In a recent 20 year series by Yeh et al., 46 patients with anorectal melanoma were studied. Twenty-seven underwent local excision and 19 underwent abdominoperitoneal resection. Recurrence rates were identical in each group (75%) with 5 year median survivals of 32% for abdominoperitoneal resection and 35% for local excision [11]. Given the high morbidity associated with abdominoperitoneal resection the current surgical trend is for local excision. Both chemotherapy and radiation therapy are generally not effective. While demonstrating promise, immunotherapy thus far, has not been widely used [7].

The lungs are a common site of metastatic disease from solid tumors, however, endobronchial metastases are rare. Endobronchial metastases from melanoma are even more infrequent. In a review from 1962–2002, Sorensen, found a total of 204 patients with endobronchial metastases originating from 20 different primary extrapulmonary tumors [10]. In this review, 8 of the cases were caused by metastatic melanoma. In our search of the entire English literature we have found only 14 cases of metastatic melanoma causing endobronchial metastases [8,9,12–20]. The incidence of intratracheal metastasis is even more uncommon.

Kobayashi describes a patient that developed isolated pulmonary metastasis without recurrence at the primary site nine years after receiving treatment for her primary tumor, sinonasal melanoma [21]. Of interest, this is the only case of pulmonary metastasis that originated from a mucosal melanoma, but it did not cause an intrinsic mass in the bronchi [21]. Our patient had a vastly different presentation with very aggressive and diffusely metastatic disease. To our knowledge, this is the first documented case of primary anorectal melanoma causing both endobronchial and endotracheal metastases.

Endobronchial metastases most commonly present with cough, shortness of breath, and hemoptysis [10]. Our patient initially presented with only cough. This was surprising, especially given the nearly obstructing endobronchial tumor discovered on flexible bronchoscopy. His acute decompensation with hypoxemia, tachycardia and relative hypotension on his first day of hospitalization was likely related to intermittent complete bronchial obstruction from this mass.

Treatment for endobronchial metastasis has changed over the years as novel endobronchial therapies have emerged. In general, assessing the efficacy of various therapies is difficult because most subjects with endobronchial metastasis have diffuse metastatic disease and treatment is usually palliative. Older therapies included combined chemotherapy and radiation therapy with or without surgery. Newer modalities include the use of laser evaporation [15,18], endobronchial brachytherapy, stenting, and photodynamic therapy [22]. For our patient, mechanical debridement and argon plasma coagulation recanalization was utilized.

Stranzel et al. treated 11 patients with iridium-192 as palliative endobronchial brachytherapy over seven years. Complete endoscopic response was observed in three patients and in five others partial opening of the initially obstructed airway was achieved. Eight of the patients had relief of their symptoms [23]. Despite modern advances in therapies, surgical resection should still be considered for appropriately selected patients since long progression-free survivals have been reported after surgery, albeit not for melanoma [20].

Conclusions

Anorectal melanoma is a rare but often fatal disease. We have presented the first known case in the English literature of anorectal melanoma with endobronchial metastasis. By the time our patient received medical care his disease was widely metastatic and unsuitable for conventional therapy. Therapeutic bronchoscopy was used to prevent asphyxiation from the endotracheal mass. Endobronchial metastasis is uncommon and can be easily mistaken for primary bronchogenic carcinoma of the lung. When evaluating patients

with endobronchial masses, it should always be considered that these may represent metastatic disease from an extrathoracic primary tumor.

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Conflict of interest

None declared.

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