

## CASE REPORT

# Coincidence or not? A rare case of rectal leiomyosarcoma amidst incidental findings of anal squamous cell carcinoma: case presentation and literature review

Gosta Iwasiuk and Jason Nealy\*

Department of General Surgery, Community Memorial Hospital, Ventura, CA, USA

\*Correspondence address. E-mail: jnealy@cmhshealth.org

## Abstract

Leiomyosarcoma accounts for only a small percentage of colorectal malignancies and remains a difficult disease to diagnose and treat. Here, we describe a case of leiomyosarcoma in a middle-age female first identified on colonoscopy. The cancer had invaded the posterior vaginal wall necessitating an abdominoperineal resection with partial vaginectomy and pelvic floor reconstruction with neovagina creation. Final pathologic analysis revealed invasion into the posterior vaginal wall without direct involvement of the vaginal squamous mucosa. Interestingly, a second carcinoma was found in the anal epithelium. Histology demonstrated this to be an anal squamous cell carcinoma positive for P16 immunohistochemistry, indicating HPV infection. Little is known as to whether leiomyosarcoma is also related to other carcinomas of the GI tract. This case of rectal leiomyosarcoma with temporal and spatial relationship to anal canal squamous cell carcinoma begs the question if HPV can trigger other neoplasms.

## INTRODUCTION

Leiomyosarcoma is a smooth muscle, mesenchymal neoplasm representative of 10% of all smooth muscle tumors. The GI tract is an uncommon location for leiomyosarcoma. When found in the alimentary tract, the order of prevalence is as follows: stomach, followed by the small intestine, then colon, and finally the rectum. Leiomyosarcoma of the rectum accounts for 0.1% of all colorectal malignancies [1–3]. Though uncommon, ~300 reported cases of rectal leiomyosarcoma are cited in the literature [1]. Due to the infrequent occurrence of leiomyosarcoma of the rectum, little is known about the epidemiology, behavior, oncological treatment and prognosis. No literature to date has documented a case of leiomyosarcoma of the rectum occurring concomitantly with squamous cell carcinoma of the anus.

Leiomyosarcoma most commonly occurs in the fifth or sixth decades of life and varies upon the location of the sarcoma. The most common presenting symptoms of rectal leiomyosarcoma are hematochezia and perianal pain [4]. When located in the rectum it is often detected by palpation on digital rectal exam, as 80% of rectal leiomyosarcomas are located in the distal third of the rectum and generally grow into the lumen [5].

## CASE REPORT

A 57-year-old Caucasian female presented to the outpatient general surgery clinic after referral from her primary care provider for a rectal mass visualized on colonoscopy. She admits to three episodes of hematochezia in the past year; her last

Received: June 19, 2017. Accepted: September 12, 2017

Published by Oxford University Press and JSCR Publishing Ltd. All rights reserved. © The Author 2017.

This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/4.0/>), which permits non-commercial re-use, distribution, and reproduction in any medium, provided the original work is properly cited. For commercial re-use, please contact [journals.permissions@oup.com](mailto:journals.permissions@oup.com)

episode being one week ago. She denies constitutional symptoms including fever, weight loss, fatigue and night sweats. Additionally, she denies abdominal pain, nausea, vomiting or melena. Her social history includes a 30-pack year smoking history as well as alcohol dependence. Pertinent family history includes her father, deceased from a colon malignancy at the age of 63. Past medical history includes gastroenteritis, low back pain and osteopenia.

Digital rectal exam demonstrates a fungating, anteriorly fixed, firm mass in the 12 o'clock position. Tissue was friable and gross blood was observed on a gloved finger. The mass was palpated 5 cm from the anal verge and was nearly obstructing. External hemorrhoids were also observed.

An exam under anesthesia allowed better visualization of the mass and incisional biopsy. As the tumor was better appreciated, concern arose for possible invasion into the posterior wall of the vagina. The mass measured  $3.4 \times 2.5 \times 1.0 \text{ cm}^3$  and was grossly infiltrating into the colonic mucosa and is seen invading the anoderm. Immunohistochemical analysis was performed demonstrating positive markers for actin, desmin and Ki-67. Negative for S-100, CD34, CD117 and pankeratin.

PET/CT demonstrated a 4.5 cm low rectal tumor with no evidence of metastasis or lymph node involvement. MRI of the pelvis demonstrated a 5 cm mass with possible extension beyond the anterior rectal wall. Loss of the fat plane between the mass and the posterior wall of the vagina was appreciated. The mass extended to the level of the internal anal sphincters and mass effect was evident upon the right internal anal sphincter. Again, no lymph node involvement was identified.

In total, an open abdominoperineal resection with ostomy creation, TAH-BSO, partial vaginectomy, perineum reconstruction and neovagina creation with left gracilis flap was performed. A liver biopsy was also obtained given her history of alcohol dependence and possible metastatic spread.

Final pathologic analysis revealed a  $5 \times 4.5 \times 4.5 \text{ cm}^3$  mass invading the posterior vaginal wall without direct involvement of the vaginal squamous mucosa. Interestingly, a second carcinoma was found in the anal epithelium. Histology demonstrated an anal squamous cell carcinoma positive for P16 immunohistochemistry, indicating HPV infection. Liver biopsy demonstrated minimal histological changes.

## DISCUSSION

The incidental finding of a second lesion, anal squamous cell carcinoma makes this case report unique among an extensive literature review of leiomyosarcoma. One case report demonstrates rectal leiomyosarcoma occurring 17 years following radiation therapy for prostate cancer [2]. Another, describes leiomyosarcoma in a patient who had previously been irradiated for stage 2 squamous cell cancer of the anus [3]. Overall, it has been noted that 5–12% of radiation-induced sarcomas are leiomyosarcomas [3]. However, no case reports were identified of rectal leiomyosarcoma and anal squamous cell carcinoma occurring concomitantly in a patient with no prior history of radiation. This brings up the question of whether these were two mutually exclusive events, or if HPV infection has a role not only in proliferation of squamous cell carcinoma but in leiomyosarcoma as well.

The complexity of rectal leiomyosarcoma lies in its rarity as well as the invasive nature of this cancer. The choice between local excision versus abdominoperineal resection lies in evaluation of tumor size and location; either  $<2.5 \text{ cm}$  allowing local excision and  $\geq 2.5 \text{ cm}$  suggesting APR [1, 5, 6]. Yet, recurrence rates suggest abdominoperineal resection to be superior to local excision [1, 5]. As stated in a retrospective case study of 12 rectal leiomyosarcomas from 1976 to 1995, local recurrence rates were much higher in patients receiving wide local excision than those receiving abdominoperineal resection (100 vs. 20%) [4]. An additional case study, found the recurrence rate to be 67.5% for local excisions and 19.5% for abdominoperineal resection [6]. Adjuvant radiation or chemotherapy was found to be poor. Furthermore, prognosis for those with documented leiomyosarcoma is poor due to its propensity to develop in other areas of the GI tract following resection. In the above study, prognosis for all 12 participants was found to be 83% at 1 year and 46% at 5 years [6].

With only 300 cases in literature, additional studies are necessary to define the epidemiology and course of leiomyosarcoma of the rectum. Although it has been shown that leiomyosarcoma of the rectum can occur following pelvic irradiation, little is known as to whether leiomyosarcoma is also related to other carcinomas of the GI tract, such as GIST tumors for example. Our case presentation of rectal leiomyosarcoma with temporal and spatial relationship to anal canal squamous cell carcinoma begs the question if HPV can trigger other neoplasms or is this just one of those coincidences? Aesop warned us that one swallow does not make a summer, but it does raise the possibility.

## CONFLICT OF INTEREST STATEMENT

None declared.

## REFERENCES

1. Chattopadhyay G, Ray D, Chakravarty S, Mandal S. Leiomyosarcoma of rectum and anal canal—a diagnostic challenge: a review. *World J Colorectal Sur* 2008;1:Art. 14 PubMed. Web. 29 Jan. 2016.
2. Freedberg DE, Bao F, Stein JA. Rectal leiomyosarcoma after pelvic irradiation. *Clin Gastroenterol Hepatol* 2013;11:A28 Clinical Key. Web. 29 Jan. 2016.
3. Futuri S, Donohoe K, Spaccavento C, Yudelman I. Rectal leiomyosarcoma: a rare and long-term complication of radiation therapy. *BMJ Case Rep* 2014;ClinicalKey. Web. 29 Jan. 2016.
4. Wang HS, Chen WS, Lin TC, Lin JK, Hsu H. Leiomyosarcoma of the rectum: a series of twelve cases. *Chin Med J* 1996;57:280–3. ClinicalKey. Web. 29 Jan. 2016.
5. Eriguchi N, Aoyagi S, Hara M, Tanaka E, Hashimoto M. A case of giant leiomyosarcoma of the rectum. *Kurume Med J* 1998;45:137–41. PubMed. Web. 29 Jan. 2016.
6. George MD, Demetri D, Jeffrey Morgan MD, Raut CP. *Local Treatment for Gastrointestinal Stromal Tumors, Leiomyomas, and Leiomyosarcomas of the Gastrointestinal Tract*. UpToDate, 23 Apr. 2015. Web. 12 February 2016.