CASE REPORT

Rectal leiomyosarcoma: a rare and long-term complication of radiation therapy

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SUMMARY

Leiomyosarcoma of the rectum can develop as a late complication in patients with a history of pelvic irradiation. We report the case of a patient who developed rectal leiomyosarcoma 13 years after receiving radiation for treatment of a stage 2 squamous cell cancer of the anus. This was detected on physical examination. Based on a discussion with the patient, we decided to manage conservatively. Overall, leiomyosarcomas constitute 5–12% of radiation-induced sarcomas. Rectal leiomyosarcoma is rare, accounting for 0.1–0.5% of all malignant tumours of the rectum. As radiation therapy plays a major role in the management of anal cancer, it is important that clinicians are aware of the possible development of radiation-induced sarcomas that may occur decades after initial management.

years prior, he was diagnosed with a T2 N0 stage 2 squamous cell anal cancer after symptoms of rectal bleeding. At that time, the patient underwent treatment for anal cancer with mitomycin at 10 mg/m² intravenously on days 1 and 2 and fluorouracil 1000 mg/m²/day continuous intravenous infusion on days 1–4 and 29–32. The patient also underwent 5 weeks of concurrent radiation therapy. The radiation dose was 3780 cgy+1620 cgy cone down, totalling 5400 cgy. The treatment resulted in a complete clinical response.

A colonoscopy in 2008 demonstrated no recurrence of disease. An oesophagogastroduodenoscopy was also performed at the time for symptoms of gastroesophageal reflux and showed mild gastritis.

The patient was evaluated by a colorectal surgeon and was noted on digital rectal examination to have a mobile mass in the posterior midline rectum.

BACKGROUND

We report a case of leiomyosarcoma of the rectum in an elderly man who underwent chemoradiation therapy for anal cancer. While there are case reports in the literature of sarcomas developing in the gastrointestinal tract following pelvic radiation therapy for gynecological and prostate malignancies, to the best of our knowledge there are no cases reported of leiomyosarcoma developing within the rectum following radiation treatment for anal cancer.

CASE PRESENTATION

A 90-year-old man presented in 2013 with bright red bleeding per rectum and rectal pain. Thirteen

INVESTIGATIONS

Colonoscopy confirmed an irregular friable 5 cm polyp that was inadvertently fractured during digital rectal examination. Biopsy of the polyp revealed proliferation of uniform tightly packed sheets of spindle cells, numerous mitotic figures, and occasional pleomorphic nuclei. Immunohistochemical stains were positive for desmin and negative for CD117 (figure 1). These findings were consistent with leiomyosarcoma. MRI of the pelvis demonstrated short segment wall thickening and abnormal signal intensity near the anal–rectal junction (figure 2).

Figure 1 Immunohistochemical stains showing positive staining of the tumour cells for desmin and smooth muscle actin, and negative staining for CD34, S100 and CD117. The immunohistochemical stains confirm the smooth muscle origin of the tumour.

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Figure 2 MRI of the pelvis demonstrates short segment wall thickening and abnormal signal intensity near anorectal junction. The abnormal signal extends into the perirectal fat. This represents the reported site of neoplasm.

TREATMENT

The patient was offered surgical management via abdominoperineal resection, but he declined.

OUTCOME AND FOLLOW-UP

The plan is to repeat physical examination every 3 months, undergo CT of the abdomen and pelvis every 6 months and proctoscopy within 1 year.

DISCUSSION

Therapeutic radiation for a variety of primary cancers has been linked to subsequent malignancies. While radiation-associated sarcomas are uncommon, constituting 3–6% of all sarcomas, it is a well recognized event.^{1–3}

Identified risk factors for developing radiation-induced sarcoma include young age at initial treatment, genetic factors and treatment-related factors including high radiation dose and concomitant chemotherapy. Radiation-induced sarcomas are often high-grade tumours that are difficult to completely resect. Furthermore, the prognosis of radiation-induced sarcoma is poor in comparison to spontaneously developing sarcoma.³

In a review of 130 patients by Gladly *et al*,³ the median time interval between radiotherapy and development of a radiation-associated sarcoma is 10 years, although this varies with histological subtype.⁴ The most common histological types of radiation-associated sarcomas are pleomorphic malignant fibrous histiocytoma (PMFH) (26%), angiosarcoma (21%), fibrosarcoma (12%), leiomyosarcoma (12%) and malignant peripheral nerve sheath tumour (9%).

Overall, leiomyosarcomas constitute 5–12% of radiation-induced sarcomas. Rectal leiomyosarcoma is rare accounting for 0.1–0.5% of all malignant tumours of the rectum.^{5,6} There are only four other case reports of rectal leiomyosarcoma developing in the context of pelvic irradiation. Three of these cases were patients with gynecological malignancies. Despite the high prevalence of prostate cancer, there is only one case of rectal leiomyosarcoma developing after external beam radiation therapy for prostate cancer.^{7–10} We report a case of rectal leiomyosarcoma detected 13 years after chemoradiation for anal cancer in an elderly man.

In 2013, it is estimated that there will be 7060 new cases of anal cancer and an estimated 880 people will die of this disease.¹¹ Rates of new anal cancer cases are rising on average 2.2% each year over the past 10 years. As radiation therapy plays a major role in the management of anal cancer, it is important that clinicians are aware of the possible development of radiation-induced sarcomas that may occur decades after initial management.

Learning points

- ▶ Rectal leiomyosarcoma may be a late effect of anal irradiation.
- ▶ Close surveillance of patients with anal cell carcinoma who have undergone radiation is recommended.
- ▶ Discuss with patients the risk of secondary malignancies with radiation.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Cahan WG, Woodward HQ, Higinbotham NL, *et al*. Sarcoma arising in irradiated bone report of eleven cases. *Cancer* 1948;1:3–29.
- 2 Brady MS, Gaynor JJ, Brennan MJ. Radiation-associated sarcoma of bone and soft tissue. *Arch Surg* 1992;127:1379–85.
- 3 Gladdy RA, Qin LX, Moraco N, *et al*. Do radiation-associated soft tissue sarcomas have the same prognosis as sporadic soft tissue sarcomas? *J Clin Oncol* 2010;28:2064–9.
- 4 Singer S, Maki RG, O'Sullivan B. Soft tissue sarcoma. In: DeVita VT, Lawrence TS, Rosenberg SA, eds. *Cancer principles and practice of oncology*. 9th edn. Philadelphia, PA: Lippincott Williams & Wilkins, 2011:1533–4.
- 5 Figueiredo JA, Andrade AFZB, Carneiro BGMC, *et al*. Rectal leiomyosarcoma, three-year follow-up. *J Coloproctol* 2012;32:73–5.
- 6 Cuffy M, Abir F, Longo WE. Management of less common tumors of the colon, rectum and anus. *Clin Colorectal Cancer* 2006;5:327–37.
- 7 Drumea K, Sabo E, Zuckerman E, *et al*. Leiomyosarcoma of the colon in the aftermath of pelvic irradiation for endometrial carcinoma. *Am J Gastroenterol* 1993;88:1302.
- 8 Basu I, Lemonas P. Leiomyosarcoma of the rectum following pelvic irradiation: a difficult histological diagnosis. *Ann R Coll Surg Engl* 2012;94:e44–5.
- 9 Caporale A, Angelico F, Cosenza MU, *et al*. A late complication of pelvic radiotherapy: leiomyosarcoma of the rectum report of a case and review of the literature. *Hepatogastroenterology* 2003;50:1933–6.
- 10 Freedberg DE, Bao F, Stein JA. Rectal leiomyosarcoma after pelvic irradiation. *Clin Gastroenterol Hepatol* 2013;11:A28.
- 11 *SEER cancer statistics factsheets: anal cancer*. Bethesda, MD: National Cancer Institute. <http://seer.cancer.gov/statfacts/html/anus.html>

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