Synchronous Squamous Cell Carcinoma of the Anorectum and Proctosigmoid—a Case Report

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Abstract

Primary squamous cell carcinoma of the rectum is extremely rare. Squamous cell carcinoma of the anorectum is a known entity evaluation and treatment protocols have been well defined. The occurrence of squamous cell carcinoma of rectosigmoid in concurrence with squamous cell carcinoma of the anal canal is extremely rare and the first case to be reported in the literature. A 48-year-old male patient presented with difficulty in passing stools. The clinical examination, colonoscopy, and the histopathology revealed synchronous proctosigmoid and the anorectal neoplasm. A case like this will pose a diagnostic challenges; squamous cell carcinoma of the rectum is not defined in the medical literature; synchronous squamous cell carcinoma will pose both diagnostic dilemmas and treatment challenges like whether to conserve the sphincter or not. Because squamous cell carcinoma of the anorectum can be treated by chemoradiotherapy, even this case is initially thought of treating by neoadjuvant chemoradiotherapy followed by low anterior resection followed by adjuvant treatment. Since there was no medical literature to support this treatment, ultimately multidisciplinary tumor board decision was made for abdominoperineal resection. Patient was treated with abdominoperineal resection and adjuvant chemoradiation. The diagnostic dilemmas and the management issues have been discussed.

Keywords CA rectosigmoid · CA anorectum · Synchronous malignancies · Squamous cell carcinoma · Abdominoperineal resection

Introduction

The most common type of cancer in the rectum is adenocarcinoma. The primary squamous cell carcinoma of the rectum is extremely rare in its occurrence. It is probably 0.10 to 0.25 per 1000 cases of carcinoma rectum [1]. Very meager literature is available till date to know the etiopathogenesis, epidemiology, and management of such cases. Every case reported will form a building block for studying the nature of such a disease.

Occurrence of squamous cell carcinoma of the rectum in concurrence with squamous cell carcinoma of the anal canal is a unique occurrence which has not been reported till date in the medical literature.

Our report marks the first report of such an occurrence. The following article will discuss the diagnostic dilemmas and management bias we encountered during the management of this case.

Case Report

A 48-year-old Indian male presented to us with complaints of painful defecation and bleeding per rectum since 6 months. There was no history of abdominal pain or symptoms suggestive of intestinal obstruction. The patient is a known case of type 2 diabetes mellitus since 2 years and on regular treatment. There was no history of tobacco use or use of alcohol. There was no history of any similar complaints in the family. There was no history of homosexuality or anal intercourse.

Abdominal examination was normal. On per rectal examination, a circumferential ulceroproliferative growth is noted, involving the anorectum, starting from the anal verge up to 2–3 cm. The luminal narrowing is up to 80%. The rectal mucosa beyond 2–3 cm felt normal. There are no enlarged lymph

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nodes in the inguinal region or neck. The systemic examination was normal.

A punch biopsy from the growth in the anal canal was taken; histopathology revealed it to be poorly differentiated squamous cell carcinoma (Fig. 1). Colonoscopy revealed a neoplastic lesion in the rectosigmoid junction (Fig. 2). Colonoscopic findings were an ulcerated friable growth in the anal canal, followed by normal rectal mucosa. A rectosigmoid polypoidal eccentric friable growth 14–19 cm from the anal verge was noted (Fig. 3). Biopsy from the proximal lesion showed moderately differentiated squamous cell carcinoma (Fig. 4).

A contrast enhanced CT scan of the abdomen and pelvis showed double lesions, one at the rectosigmoid junction and another at the anal canal. In view of double lesions, final diagnosis of synchronous primary squamous cell carcinoma of the rectosigmoid cT3N0M0 [Stage IIA] and squamous cell carcinoma of the anal canal cT2N0M0 (Stage II) as per AJCC 2010 was made.

In the baseline blood investigations, CT thorax was normal.

Since the patient had a synchronous rectosigmoid and anorectal malignancy, he was treated with abdominoperineal resection and permanent colostomy. The gross specimen and the post-operative histopathology confirmed the synchronous primary squamous cell carcinoma of the rectosigmoid and anal canal. Rectosigmoid disease was T3 with a single node showing metastasis. Distance between the anorectal disease is 7.5 cm. Intervening colonic mucosa was unremarkable. The

patient is now being planned for adjuvant treatment by chemoradiation.

Discussion

Rectal carcinomas are one of the 10 leading sites of cancer in Bangalore [2]. However, adenocarcinoma is the most common histology constituting 90% of the rectal carcinoma [3]. Occurrence of squamous cell carcinoma is extremely rare, and till date, only case reports and few small series of the patients have been reported in the literature [1]. The largest of the case series is in Russian, reporting 107 patients; however, the details of the patients are not known and probably the anorectal cases have been over reported as rectal cases [1, 4].

Excluding the Russian study, till date, around 83 cases have been reported as primary rectal squamous cell carcinoma in the literature [3, 5]. Ours will be the 84th case. However, our case is not only primary squamous cell carcinoma of rectum, but also a synchronous malignancy.

Synchronous malignancies are two or more histologically distinct simultaneously detected malignancies within a period of 6 months. Synchronous malignancies of the rectum and the anal canal have been reported in the past [6, 7]. In all the reports, the histology of the rectum is adenocarcinoma. Our patient had squamous cell carcinoma of both the rectum and the anal canal.

Fig. 1 H&E stained sections. **a** Anal canal tumor: $\times 10$ showing nests of squamous cells with desmoplastic reaction. **b** Anal canal tumor: $\times 40$ showing squamous cells of marked pleomorphism, mitotic activity, and inflammatory infiltrate (suggestive of poor differentiation). **c** Rectosigmoid tumor: $\times 10$ squamous cells with papillaroid configuration with keratin pearls. **d** Rectosigmoid tumor: squamous cells of moderate pleomorphism and mitotic activity. (Both **c** and **d** are suggestive of moderate differentiation)

Fig. 2 Contrast enhanced CT scan abdomen and pelvis. **a** Coronal section. **b** Saggital section. The arrow marks represent the rectosigmoid growth and the anorectal growth with intertwining normal rectal mucosa

The exact etiopathogenesis of the occurrence of squamous cell carcinoma in the rectum is not understood. Only scientific speculations are made.

1. The occurrence could be as a consequence of inflammatory bowel disease, or any chronic infection or secondary to radiation [1]. In our patient, there is no such history.
2. It could be due to pluripotent stem cells undergoing squamous metaplasia and hence carcinoma [8]. However, as the author states, usually, the carcinomas arising from pluripotent stem cells are poorly differentiated. In contrast, our patient had a moderately differentiated squamous cell carcinoma. But, this theory cannot be totally ruled out.
3. Human papilloma virus has a well-established association with various squamous cell carcinomas in human beings. It is attributed to the causation of squamous cell carcinomas of the head and neck, penis, anal canal, and skin in males [9]. Possible infection of the rectal mucosa and the anal canal could be one of the reasons for such a synchronous occurrence in our patient. However, our patient denies of any anal intercourse. We could not get a HPV DNA assay done on our patient.

Fig. 3 **a** External appearance of the anal tumor. **b**, **c**, **d**—colonoscopy images where **b**—friable anorectal growth; **c**—normal rectal mucosa; **d**—proctosigmoid growth

Fig. 4 Abdominoperineal resection specimen: **a**—proctosigmoid growth breaching the serosa; **b**—normal mesorectum; **c**—anorectal growth

The management of such a patient poses a real practical challenge. Initially, the patient was considered for sphincter-saving surgery, but in view of poorly differentiated histology of anal canal tumor and possibility of extra mucosal spread of tumor from the anal canal, the lesion cannot be determined without surgical resection and histological diagnosis. Hence, a multispecialty tumor board decision was made and the patient was considered for the abdominoperineal resection. Since the rectal disease is of T3 status and a single node showed metastatic spread, the patient has been advised chemoradiation as an adjuvant treatment.

Conclusion

Synchronous squamous cell carcinoma of the rectosigmoid and the anorectum is extremely rare and the first case to be reported in the medical literature. This case posed a lot of

diagnostic dilemmas and difficulties, during the management of this case. However, the patient was treated with abdominoperineal resection and adjuvant chemoradiation. In view of limited literature of synchronous squamous cell carcinoma of the rectosigmoid and anal canal, more cases need to be studied to understand the natural history of the disease and formulate guidelines for the management of these cases. I would be grateful if you could publish the article without funding as I do not have any sources to fund the article. The cost of the article is also borne by me.

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