BMJ Case Reports

Unusual association of diseases/symptoms

Rectal adenocarcinoma in an adult with imperforate anus at birth

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Summary

A 43-year-old man with imperforate anus surgically corrected at birth presented with bleeding per rectum and constipation. On investigations, there was an adenocarcinoma at 2 cm from anal verge involving mid and lower rectum. He underwent abdominoperineal resection. During surgery, he was found to have a rectourethral fistula. The relationship between imperforate anus, rectourethral fistula and carcinoma of rectum is disclosed.

BACKGROUND

Colorectal cancer is one of the common gastrointestinal tract cancers in the elderly. Its occurrence in patients less than 50 years of age should make one look out for factors which predispose to its development. It is associated with various genetic, environmental and dietary factors. Chronic exposure of rectal mucosa to urine due to congenital rectourethral fistula could also predispose to development of rectal cancer as seen in patients with ureterosigmoidostomy. We present a case of rectal adenocarcinoma associated with a congenital rectourethral fistula which in turn was a part of imperforate anus. There are very few reports of this association in the literature.

CASE PRESENTATION

A 43-year-old man presented with complaints of painless bleeding per rectum with constipation since past 2 months. He gave history of being treated surgically at birth for imperforate anus. A perineal operation was done, details of which were not available. He had a history of recurrent urinary tract infections and urgency of bowel movements since childhood.

INVESTIGATIONS

Colonoscopy revealed a polypoidal growth at 2 cm from anal verge with rest of the colon being normal. Biopsy revealed adenocarcinoma. Contrast-enhanced CT of abdomen and pelvis revealed circumferential lesion in lower and mid rectum without lymphadenopathy, normal liver and no ascites. There was no abnormality of the urinary tract.

DIFFERENTIAL DIAGNOSIS

- Rectal cancer
- Rectal polyp

TREATMENT

He underwent abdomino perineal resection with end colostomy. At surgery, there was circumferential thickening of mid and lower rectum with no perirectal fat involvement. No peritoneal, omental or liver involvement. While performing anterior dissection in the perineal phase, it was noticed that rectum was placed anteriorly and there was difficulty in finding plane between anterior wall of rectum and urethra. While dissecting this plane, he was noticed to have a rectourethral fistula. There was however, no involvement of the urethra by the tumour. The fistula was excised. Urethra was sutured in layers over the urinary catheter and suprapubic catheterisation was done to divert urine.

In view of intraoperative findings, on retrospective questioning patient gave history of pneumaturia since childhood.

OUTCOME AND FOLLOW-UP

The pathological stage of tumour was pT3N0M0, stage IIA with tumour-free longitudinal and radial margins.

The patient is currently undergoing chemoradiotherapy.

DISCUSSION

In this patient, the development of a malignant tumour in rectum at the age of 43 years without having any familial predisposition to bowel cancer points the possible role of rectourethral fistula as the predisposing factor. Rectal cancer is usually associated with rectourethral fistula when there is malignant invasion of urethra. Its occurrence with congenital rectourethral fistula is very rare, although a few cases of colorectal cancer associated with anorectal malformations have been reported.[1–3] Congenital rectourethral fistula leads to numerous complications, but whether it increases the risk of rectal cancer is not known. We know that ureterosigmoidostomy done for urinary diversion increases the risk of colorectal cancer by 100-fold.[4] Several theories have been proposed to explain this carcinogenesis in colon but the most favoured one is the increased formation of N-nitroso compounds in colon by bacterial flora from urinary nitrates.[4] The latency between ureterosigmoidostomy and development of

colorectal cancer is 6–50 years with mean time of 21 years.[5] But cases of colon cancer have been reported even after exposure of colonic mucosa to urine for as little as 8 months probably because the initial exposure of the colonic mucosa to the urine is enough to start the chain of events leading to development of tumours.[6]

We postulate that by similar mechanisms that lead to increased risk of colorectal cancer after ureterosigmoidostomy, there is an increased risk of colorectal cancer in cases of rectourethral fistula as seen in our case.

In our patient, the rectourethral fistula was discovered while attempting separation of anterior wall of rectum from posterior wall of urethra during abdominoperineal resection. Urethral injuries can occur because of abnormal anatomy in patients with anorectal malformations.[2] Preoperative genitourinary evaluation could be helpful in identifying urethral involvement to prevent urinary tract injury during surgery and plan the surgery better.[2]

Learning points

- Patients of anorectal anomalies might be at an increased risk of developing rectal cancer.
- Patients of rectal cancer with history of anorectal anomalies should undergo detailed genitourinary evaluation.

Competing interests None.

Patient consent Obtained.

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