



Carcinoma rectum in adolescent male-An unusual presentation

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Abstract

A 13 year old male presented with complaint of pain in abdomen, bloating and altered bowel habits since 3 to 4 months. He was cachexic, abdomen was distended and bowel sound were present but decreased. Detailed examination and investigatory workup was done. CT scan revealed mass in rectum near anal verge involving pelvic musculature and posterior wall of urinary bladder. Sigmoid colostomy with incision biopsy which was suggestive of signet ring cell adenocarcinoma. Chemotherapy and radiotherapy was given and patient was periodically followed up. But the tumor was aggressive in nature and patient was put on palliative chemotherapy. The patient could not tolerate chemotherapy and lost his life to cancer within 2 years of diagnosis.

Keywords: rectal cancer, adolescent, signet ring cell

Introduction

According to Globocan 2020, colorectal is third most common cancer (10.0 %) after breast and lung cancer and it is the second most common cause of cancer related mortality (9.4%) [1]. Incidence of colorectal cancer is much lower in India as compared to western countries, and it is the seventh leading cancer in India. Mean age of rectal cancer in India is approximately 40-45 yr [2]. Carcinoma rectum is rare cancer in children and adolescents belonging to age range of less than 20 years. The cancer is the second most common cause of cancer related death. Early diagnosis and management have resulted in good overall survival and reduced mortality [3]. Routine colonoscopy is usually not recommended in children and adolescents and hence the cases usually present in advance stage [4-6].

Case Details

A 13 year old male presented with complaint of pain in abdomen, bloating and altered bowel habits since 3 to 4 months. Initially he visited general practitioner and received symptomatic treatment but his symptoms persisted. The child presented at our institute with similar complaints after 2 months. His family history was non-contributory.

On examination, he was cachexic, abdomen was distended and bowel sound were present but decreased. Per rectum examination was done which revealed anal stenosis and mass could be felt with great difficulty. Ultrasonography and straight X-ray of abdomen in erect posture was also done which was suggestive of lower GI tract obstruction without other significant abnormality. Further, CT scan was also done which revealed 13.2x6.4x5.8 cm mass in rectum and anal region 6 mm away from anal verge. The lesion involved bilateral bulbococcygeus, bilateral levator ani and left gluteus minimus muscles. Involvement of Posterior urinary bladder wall was also involved.

Oncosurgeon's opinion was taken and Patient was subjected to sigmoid colostomy with incision biopsy and left sided colostomy in situ. Biopsy was suggestive of signet ring cell adenocarcinoma of rectum. Patients was given neoadjuvant FOLFOX regimen. This was followed by radiotherapy with 50 Gray/25# along with concurrent 5 FU for 3 months.

After 3 months, follow up CT scan was done which was suggestive of left sided colostomy in situ with disease encasing both distal ureters causing Hydroureteronephrosis. Oncosurgeon opinion was again taken and it was observed that the disease was unresectable. Thus DJ stenting was done and palliative chemotherapy with FOLFOX was started for 4 months. Patient was then followed up regularly and after 3 months MRI Pelvis was done which revealed 2.6x 3.5x 7 cm growth in lower and upper rectum with mesorectal fact extension with mesorectal fascia and bilateral levator ani involvement. Again palliative chemotherapy with FOLFOX 6# was given for next 3 months.

Further, patient underwent mutation analysis which was negative for NRAS (2, 3, 4), BRAF, KRAS mutation. Patient was non affording for Irinotecan, and thus was put on CAPOX regimen. After 3 months, patient could not tolerate chemotherapy and was given BSC. The patient lost his life to cancer within 2 years of diagnosis.

Discussion

Colorectal carcinoma is one of the common GI malignancy among adults and elderly and is the second most common cancer in India [7-9]. The cancer is rare in pediatric age group and usual age of diagnosis is second decade of life. It is observed in higher proportions of males as compared to females with male: female ratio of 2:1 [10-11]. The incidence of rectal cancer has been estimated to be 1:1,000,000 [8,9].

We have described a case scenario of pediatric signet ring cell

adenocarcinoma of rectum in a 13 year old child with poor prognosis. Nair *et al* documented that adenocarcinoma of colorectal region in pediatric age group is often observed in setting of hereditary cancer syndromes accounting to 5-10% of all cancers [12]. However in present study, family history of colorectal or any other cancers was absent in maternal as well as paternal side.

The child presented with vague symptoms such as abdominal pain and bloating. Thus, the initial diagnosis was missed and later when presented at our centre, complete workup was done. Child was cachexic, and per rectum examination revealed stenosis and mass. Literature suggest that the mode of presentation as well as prognosis vary significantly between adults and child. Child present with no specific symptoms such as abdominal pain, change in bowel habits, vomiting, bleeding per rectum, and weight loss [10, 11]. Investigations such as ultrasonography, erect X-ray abdomen and CECT abdomen were helpful in identifying the mass in anorectal area. Though, in adults, most common site of colon cancer is left side within 25 cm of the anus including the recto- sigmoid junction [10, 11]. But in children, the site of involvement vary and may affect any part of colon and rectosigmoid junction [13]. In this scenario, a mass was observed in rectum and anal region 6 mm away from anal verge. Mirchandani *et al* [8] and Sultan *et al* [14] documented that children with colorectal cancer usually present with advanced stage and aggressive nature of cancer at the time of diagnosis. In our case, soft tissue involvement was observed i.e. the mass was associated with involvement of pelvic musculature such as bulbococcygeus, levator ani, gluteus minimus muscles along with posterior wall of urinary bladder. The nature of cancer is different in children as compared to adults. Adults usually presents with moderately differentiated adenocarcinoma whereas mucinous adenocarcinoma is of predominant type in children [10, 14]. The severe form of cancer in children could be due to mucin which is thought to absorb water, swell and is responsible for invasive nature of cancer. Due to pooling of mucin, the tumor grow rapidly and interferes with the immune recognition of carcinoma cells caused by mucopolysaccharide coating [10, 13, 14].

Upon biopsy, the signet ring cell adenocarcinoma was observed. This signet ring subtype is known to be aggressive and grows rapidly involving regional lymph node as well as is associated with diffuse peritoneal seeding. Thus this form of cancer has worst prognosis. Pandey *et al* [10], Nair *et al* [12] and Tiwari *et al* [15]. also reported mucinous adenocarcinoma and signet ring subtype of adenocarcinoma in children and its association with worst prognosis.

Initially sigmoid colonoscopy with biopsy along with adjuvant chemotherapy and radiotherapy was done. Though, resection of mass is the preferred management option in such cases [10] but as the patient presented in advanced stage, resection was not possible. However, the role of chemotherapy in pediatric colorectal cancer remains controversial [12], we prescribed FOLFOX regimen similar to adults in pediatric dose. This regimen has shown clear benefit even in advanced stage in adults [8, 16]. Apart from this, new agents such as irinotecan, oxaliplatin and leucovorin can also be used as an adjunct to 5 fluorouracil. [10] Also, molecular biology and DNA sequencing techniques are being utilized to determine the biological markers of colorectal carcinoma that may be associated separately with colorectal

cancer in adults and pediatric age group [8, 17].

In our country, one of the important determinant of choice of chemotherapeutic agent is socioeconomic status of the family according to Nair *et al* [12]. In our case, patient belonged to low socioeconomic status and could not afford bevacizumab, and thus was put on palliative chemotherapy. Gradually, he could not tolerate chemotherapy and put on BSC. Overall, the survival of child was less than 2 years. Similarly, Pandey *et al* [10] also concluded that colorectal carcinoma in children is usually a fatal disease and 5-year survival rate among them range between 7 to 12%. The factors associated with poor prognosis in these cases are delay in diagnosis, advance stage at the time of diagnosis, signet ring cell adenocarcinoma and low socioeconomic status. [10]

Conclusion

Colorectal carcinoma is rare in children and adolescents and require high index of suspicion. Any child who present with abdominal pain, bloating and altered bowel habits along with cachexia with or without rectal bleeding should be examined and investigated carefully to rule out malignancy. Rectal carcinoma is associated with worst prognosis in child and survival rate was less than 2 year in this case.

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