The Turkish Journal of Pediatrics
Volume 17 / Number 3-4 / July - October 1975 / pp 125 - 128

Leiomyosarcoma in the Rectum in the Newborn Period*

(A Case Report)

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Leiomyosarcoma is a malignant tumor composed of elements of smooth muscle. In adults it occurs in the uterus, gastrointestinal tract, kidney, prostate, bladder, etc. Very few cases have been reported in the newborn period.[1]

An embryonic leiomyosarcoma of the rectum, with rectal prolapse which caused difficulty in defecation came to our attention. We believe that our case of rectal localization of this tumor in the newborn period is the first case in the medical literature.

Case Report

A 12 day old infant was admitted to the Newborn Surgical Unit at Hacettepe Children's Hospital on Nov. 5, 1971. Her chief complaints were ano-rectal mass and difficulty in defecation. The pregnancy and delivery were normal. The mother and father were both 28 years old and healthy. The patients' birth weight was 3 kg. She was normal until 5 days before admission. Her mother noticed for the first time in the baby, a mass of about 5 cm. in diameter protruding through the anus, especially during defecation.

On physical examination she was found to be a well developed-active baby; weight was 3 kg., height was 50 cm., and pulse 140/minute. Examination of other systems revealed no abnormality except for

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a 5 x 5 x 4 cm mass protruding through the anus. The mass was solid, dark bluish in color, and there were some ulcerations and bleeding at a few points. Hb; 12 g/100 ml, W.B.C. 23.600/mm^3, urine examination and serum electrolytes were normal. Chest and abdominal X-rays were also normal.

After routine pre-operative preparation the patient was taken to the operating room and under general anesthesia the mass was reduced back into the rectum. Following this a left paramedian incision was used for laparotomy. A tumor of 5 x 6 x 8 cm. was found in the rectum and recto-sigmoid region. No signs of intra-abdominal metastasis, perforation or peritonitis were noticed. Because of the very low localisation of the tumor and general condition of the patient, only a partial excision of the tumor could be performed. Distal resection had to be carried out inside the tumor about 2 cm. below the pelvic peritoneum. The rectum was closed in 2 layers with 4 "0" silk, interrupted sutures. The pelvic peritoneum was approximated above them. Sigmoid end colostomy was performed. Post-operative course was uncomplicated. The sigmoid colostomy started to function at 48 hours and the next day the patient was started on oral feedings.

Pathologic examination of the tumor was reported as low grade malignant leiomyosarcoma (Figure 1,2). Because of the low grade malignancy of this tumor, radical surgery was postponed until the patient would become suitable for it. The patient was carefully followed and rectal examinations along with rectoscopy were performed several times. There was no tumoral growth in the rectum. No chemotherapy or radiotherapy was used. No local or systemic recurrence was found at that time.

On 5.6.1972 re-laparotomy was performed. There were no gross signs of tumor in the rectum. The tumor had disappeared spontaneously. Liver, mesentery, spleen, stomach and intestine were found to be normal. The black silk sutures that were previously used to close the rectum were easily found again. Both ureters were found and retracted. The rectum was dissected as in a Swenson pull-through procedure until one and a half centimeters from the anocutaneal line and perineal anastomosis was performed between the rectum and sigmoid colon; post-operative course was uncomplicated.

Microscopic examination of the resected rectum revealed no tumor cells at all.

The patient was examined at intervals and her defecation was found to be normal. There were no signs of metastasis.

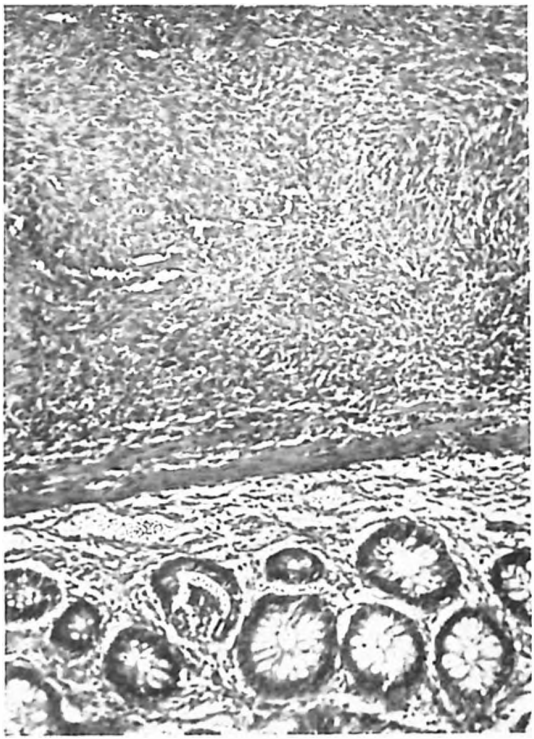

Figure 1
Photomicrograph of lesion.

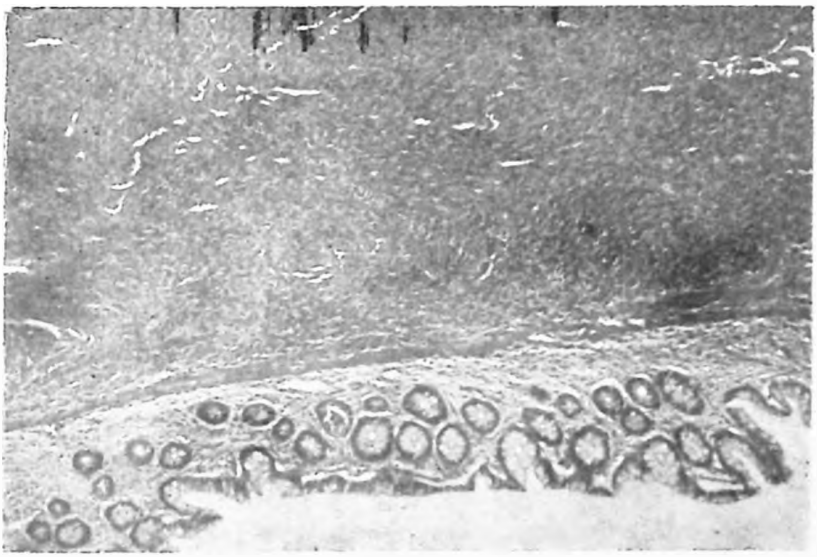

Figure 2
Photomicrograph of lesion.

Discussion

Leiomyosarcomas in the newborn period are extremely rare. Such tumors at the site of Meckel's diverticulum in the terminal ileum were reported by El Shafie et al in 1971.[1] But in our case, localization of the tumor was in the rectum and rectosigmoid.

Clinical manifestations of leiomyosarcoma in the newborn period may be an acute infectious process of the mass, or the leading point of an intussusception, or symptoms of intestinal obstruction. None of the classic signs of malignancy could be detected. In our patient the tumor presented itself as a symptom of rectal prolapse and difficulty in defecation rather than malignancy symptoms and, because of this, the tumor was able to be diagnosed in the early neonatal period.

During the first operation we could perform only a partial resection, but in re-laparotomy about 6 months later, we observed spontaneous regression of the same tumor. Microscopic examination of the resected rectum revealed no tumor cells at all. This raises the question of the possibility of spontaneous regression of leiomyosarcoma. Spontaneous regression is well known for some tumors, especially for neuroblastomas. But we could not find any case of regression of leiomyosarcoma in the literature.

The Swenson operation is one of the well established operations in the definitive treatment of Hirschsprung's disease. Although our patient did not have Hircshprung's disease, in order to obtain intestinal continuity, we performed the Swenson procedure. We believe that it is useful to perform this operation in some of the cases which are similar to ours.

Summary

Malignant tumors of the rectum are uncommon in the newborn period. This report presents leiomyosarcoma of the rectum in a newborn which we believe to be the first case in the medical literature.

Six months after the partial resection of the tumor, resected material which was left in the first intervention showed no tumor cells.

REFERENCE

1. El Shafie L, Spitz, İkeda M: Malignant Tumors of the Small Bowel in Neonates Presenting with Perforation. J Pediatr. Surg. 6: 62, 1971.