

Case Report

Tuft of Hair Per Ano - An Unusual Presentation of Pediatric Benign Ovarian Teratoma

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Abstract

It is unusual to find a tuft of hair protruding per ano in a pediatric patient. A ten year old female child presented with complaints of hair coming out per rectum for 2 years. She also had associated complaints of mild abdominal discomfort and constipation. Clinically, the child was lean and emaciated with no complaints of bleeding per rectum. The preoperative Contrast Enhanced Computed Tomography (CECT) showed an ill-defined heterogenous right adnexal mass, completely adherent to adjacent recto-sigmoid colon with loss of fat planes.

Rectosigmoidoscopy revealed indenting extraluminal mass in posterolateral rectum with hair infiltration intraluminally. On laparotomy mass was excised into along with infiltrating bowel wall with diversion stoma. The histopathology report was suggestive of mature benign solid teratoma. The child is doing well in last follow up of six months and planned for stoma reversal.

Keywords: Mature ovarian teratoma; Hair per anus; Malignant degeneration; Pressure necrosis

Introduction

Ovarian tumors are rare in children, accounting for 1% of childhood malignant tumours, with a different biological behavior, being mostly benign as compared to their adult counterpart [1]. Complications of ovarian teratomas have been well described in adults like torsion (16%), spontaneous rupture (1% to 4%), malignant transformation (1% to 2%) and involvement of adjacent organs (<1%) [2]. Urinary bladder and rectum are the most common adjacent pelvic structures involved on spontaneous rupture or direct invasion of ovarian teratomas. Unlike adult literature, complications like involvement of adjacent colon with protrusion of hair tuft per rectum is seldom reported in pediatric population.

Hereby, we report a case of solid mature ovarian teratoma in a 10 years old female child, who presented with tuft of hair protruding per anus. In addition, she had features of long standing constipation. To our best knowledge, such a rare presentation is not reported so far in the pediatric literature.

Case Presentation

We came across a 10-year old female child with complaints of hair coming out per rectum through the anal canal for last 2 years. The hair tuft was getting large with passage of time. The child often had refusal to feeds due to abdominal bloating and constipation. The features had aggravated in the last 3-4 weeks. Clinically the child appeared lean and emaciated.

Abdominal examination was suggestive of a palpable, non-tender, firm globular lump of size 8 cm × 10 cm × 8 cm with limited mobility. Per rectal examination depicted a tuft of hair strands with impacted normal colored stool with no blood staining. The hair strands continued proximally and were not easily detached. Proximal limit could not be palpated.

Rest of the examination was unremarkable. No such lump, organomegaly or lymphadenopathies were noted elsewhere. Roentgenogram of abdomen showed fecal loaded gut. Elective sigmoidoscopy was done that revealed extraluminal mass pushing the rectum in postero-lateral wall and occluding the lumen with intrusion at the superior end where it gave rise to tuft of hair intraluminally. Ultrasonography report was consistent with pelvic mass with heterogenous consistency CECT scan abdomen and pelvis were suggestive of a large well capsulated, heterogenous right adnexal mass (size 10 cm × 10 cm × 9 cm) protruding into the adjacent sigmoid colon and adherent to surrounding structures. Tumour was completely adherent with the posterior wall of rectum with loss of fat planes. Intralesional specks of calcifications were noted along with mesenteric lymphadenopathy and inter bowel fluid (Figure 1). AFP and B-HCG levels were in normal range preoperatively with 2.16 and 1.10 respectively.

On exploratory laparotomy, a large heterogenous well defined right ovarian mass was identified. The mass measured about 10 cm × 10 cm × 10 cm in all dimensions, invading the adjacent rectosigmoid. It was infiltrating the distal sigmoid colon for an approximate length of 8 cm posteriorly and invaded it at two different places. A tuft of hair was noted on dissection of the tumor which was lying in the rectosigmoid colon and the same was protruding per anus. The left ovary and the adnexal structures were displaced due to mass effect and they beared no significant findings. The mesocolon was free and the adjoining bowel loops and viscera were separated with careful dissection. Mass was carefully dissected without rupture and preserving the tissue planes. Salpingo-oophorectomy was done along with excision of infiltrated bowel (Figure 2). Diversion colostomy was done at the level of proximal sigmoid colon. The excised specimen

Citation: Chhabra A, Nazki S, Behera S, Nagpal N, Vaneet KS. Tuft of Hair Per Ano - An Unusual Presentation of Pediatric Benign Ovarian Teratoma. *Int J Pediatr Surg.* 2021;2(2):1019.

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Publisher Name: Medtext Publications LLC

Manuscript compiled: Aug 10th, 2021

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weighed 600 gm and filled with thick mucoid fluid and tuft of hair interspersed with fat and cartilaginous matter (Figure 3). However, well-formed skeletal segments were absent.

The post-operative course was uneventful and discharged on full feeds after five days. Histopathologic evaluation confirmed well differentiated mature benign teratoma of rectum (Figures 4 and 5). Patient is doing well and awaiting reversal of bowel continuity.

Discussion

Ovarian mature teratomas are the most common histologic

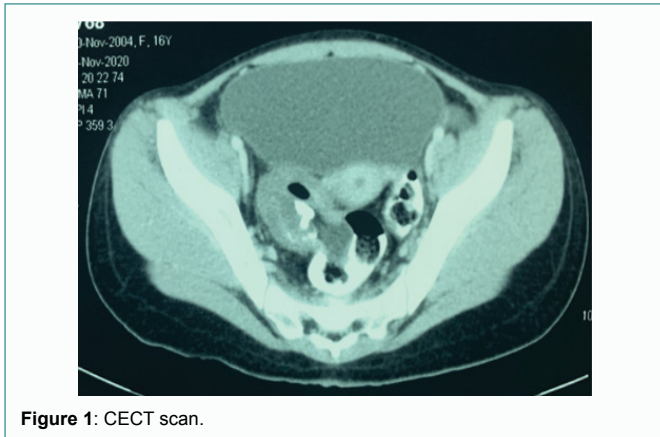


Figure 1: CECT scan.



Figure 2: Adjoining bowel loops and viscera were separated with careful dissection.

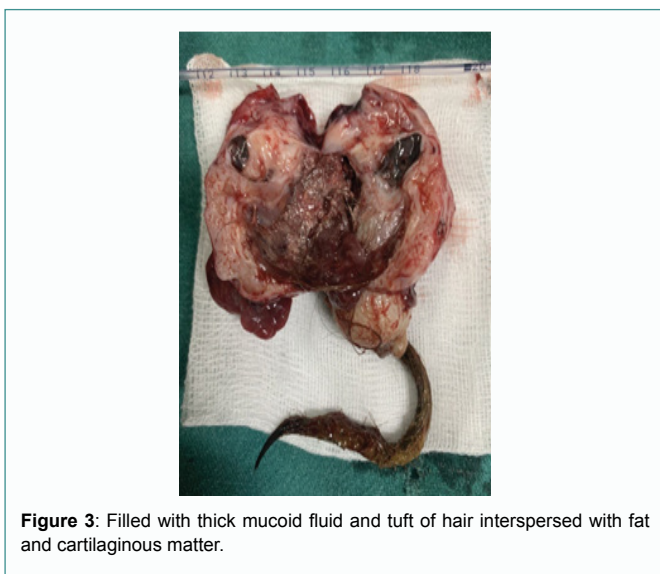


Figure 3: Filled with thick mucoid fluid and tuft of hair interspersed with fat and cartilaginous matter.



Figure 4: Lobules of mature cartilage, mucus secreting glands and fibromuscular tissue (H and E \times 40).

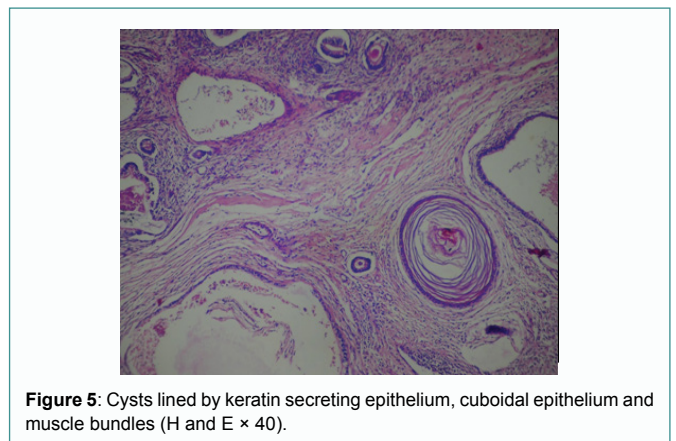


Figure 5: Cysts lined by keratin secreting epithelium, cuboidal epithelium and muscle bundles (H and E \times 40).

subtype of pediatric ovarian germ cell tumors, mostly benign in nature [3]. Although malignant transformation has been described as one of the causes for fistula formation into adjacent organs, but it is not necessary always and can be a result of local inflammatory changes. For instance, Kizaki et al. [4], reported a case in a 45 years old female with a left ovarian teratoma which was adhered to the adjacent rectum due to extensive inflammation. She underwent left salpingo-oophorectomy and upper anterior resection of rectum in view of possible malignancy, however, histopathology depicted only inflammation without any malignant changes.

Shiels et al. [5], considered certain events like rupture of cyst, dense adhesions with adjacent structures and pressure necrosis which may cause entero-ovarian fistula formations. Similarly, Kim et al. [6], reported a case of benign ovarian dermoid cyst with rectal fistula formation in a 17 years old female, who presented as passage of sebaceous material per rectum. She underwent laparoscopic excision of cyst along with primary repair of fistula.

Although malignant degeneration is an important cause reported in cases of cysto-enteric fistulas, but it is not essentially evident in all cases as reported by Cebesoy et al. [7]. They considered cyst rupture and leakage of fluid could be the primary causative factors. Mature cystic teratoma can present with range of symptom, from being asymptomatic in nature to various complications such as torsion, rupture, and malignant change. One of the rare complication in the form of super infection in a young girl without preexisting risk factors has been reported [8].

Conclusion

Fistula formations into adjacent organs are extremely rare in cases

of mature ovarian teratomas, even in adults, and often associated with malignant degeneration. Meanwhile, it should be kept in mind that fistulisation in benign teratomas is possible under circumstances like cyst rupture, torsion, leakage of cystic fluid and pressure necrosis. In our case, tumor involved the rectum with intraluminal tuft of hair, which is seldom reported in benign teratomas. Thus, the primary surgeon should be aware of this uncommon presentation and treat accordingly.

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