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CASE REPORT

Pediatric case of colonic perivascular epithelioid cell tumor complicated with intussusception and anal incarceration: A case report

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Abstract

BACKGROUND

Perivascular epithelioid cell tumor (PEComa) represents a group of rare mesenchymal tumors. PEComa can occur in many organs but is rare in the colorectum, especially in children. Furthermore, PEComa is a rare cause of intussusception, the telescoping of a segment of the gastrointestinal tract into an adjacent one. We describe a rare case of pediatric PEComa complicated with intussusception and anal incarceration, and conduct a review of the current literature.

CASE SUMMARY

A 12-year-old girl presented with abdominal pain and abdominal ultrasound suggested intussusception. Endoscopic direct-vision intussusception treatment and colonoscopy was performed. A spherical tumor was discovered in the transverse colon and removed by surgery. Postoperative pathologic analyses revealed that the tumor volume was 5.0 cm × 4.5 cm × 3.0 cm and the tumor tissue was located in the submucosa of the colon, arranged in an alveolar pattern. The cell morphology was regular, no neoplastic necrosis was observed, and nuclear fission was rare. The immunohistochemical staining results were as follows: Human melanoma black 45 (HMB 45) (+), cluster of differentiation 31 (CD31) (+), cytokeratin (-), melanoma-associated antigen recognized by T cells (-), smooth muscle actin (-), molleya (-), desmin (-), S-100 (-), CD117 (-), and Ki67 (positive rate in hot spot < 5%). Combined with the results of pathology and immunohisto-chemistry, we diagnosed the tumor as PEComa. Postoperative recovery was good at the 4 mo follow-up.

CONCLUSION

The diagnosis of PEComa mainly depends on pathology and immunohisto-

chemistry. Radical resection is the preferred treatment method.

Key Words: Perivascular epithelioid cell tumor; Colonic; Intussusception; Anal incarceration; Endoscopic direct-vision intussusception treatment; Case report

Core Tip: Perivascular epithelioid cell tumor (PEComa) of the colon is rarely encountered in the clinic, especially in pediatric patients. We describe a rare case of PEComa complicated with intussusception and anal incarceration in a 12-year-old female. We performed endoscopic direct-vision intussusception treatment and surgical removal. The diagnosis of PEComa mainly depends on pathology and immunohistochemistry. Radical resection is the preferred treatment method.

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INTRODUCTION

Colonic perivascular epithelioid cell tumor (PEComa) is rare in clinical practice, especially in children. Intussusception caused by PEComa is even rarer. This report describes a pediatric case of colonic PEComa with intussusception and anal incarceration treated with endoscopic intussusception reduction. This is the first report of such a case. Furthermore, we review the studies on colorectal PEComa indexed in the PubMed database and accessed with the keywords "Colonic PEComa" and "Rectal PEComa". A total of 30 cases were retrieved, and we provide a detailed analysis and summarization of these cases here.

CASE PRESENTATION

Chief complaints

A 12-year-old girl presented with abdominal pain as the first manifestation.

History of present illness

The patient had developed paroxysmal angina pectoris around the umbilicus and lower abdomen 17 d prior, accompanied by nausea and vomiting. Intussusception was diagnosed in a local hospital by ultrasound, and was reduced by air enema. Contrast-enhanced computed tomography (CT) scan showed abnormal enhancement on the left side of the transverse colon with intussusception, which was considered as polyps (Figure 1). Supplementary colonoscopy showed a spherical protuberance of 5 cm in diameter in the transverse colon (Figure 2). The patient was transferred to our hospital for further diagnosis and treatment.

History of past illness

The patient had no previous medical history.

Personal and family history

There was no relevant personal or family history of colon tumor.

Physical examination

The patient's vital signs were stable, the abdomen was flat and soft, the left lower abdomen was tender, and there was mild rebound pain.

Laboratory examinations

Results of routine blood, liver function, and coagulation and tumor marker tests were within the normal ranges.

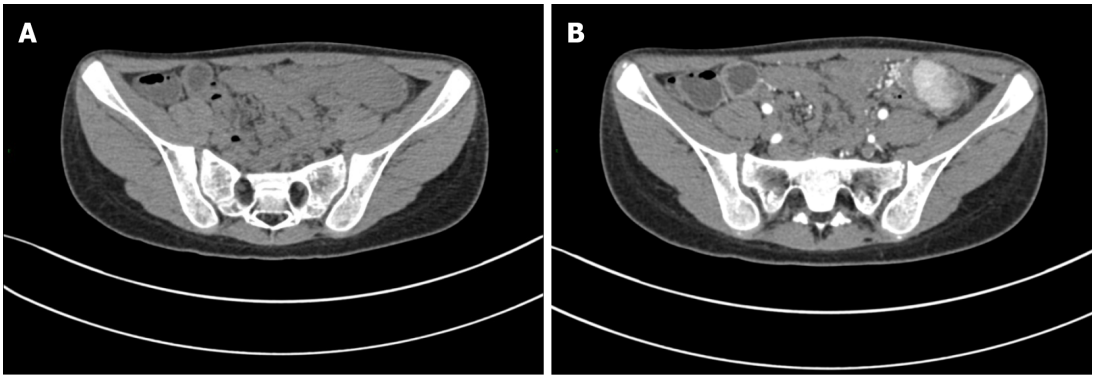

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Figure 1 Abdominal computed tomography results. A: Plain scan showed a transverse colonic mass; B: Space-occupying lesion showed obvious enhancement.

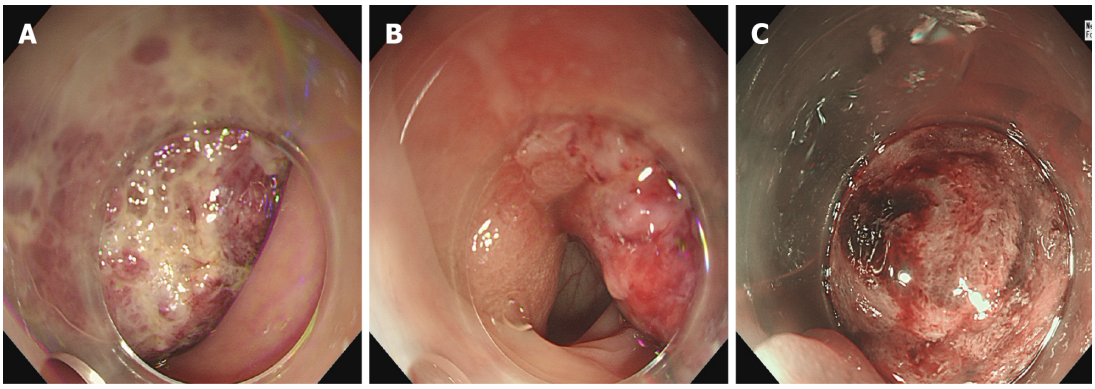

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Figure 2 Colonoscopy results. A: The tumor is spherical, with a diameter of about 5 cm, a surface that is congested and eroded, and with formation of local ulcers; B The root of the tumor has a thick pedicle, with rough surface mucosa and covered with leukoplakia; C: Narrow band imaging showed that the glandular ducts had disappeared and the presence of vasodilation.

Imaging examinations

Contrast-enhanced CT scan showed abnormal enhancement on the left side of the transverse colon with intussusception, which was considered as polyps (Figure 1). Supplementary colonoscopy showed a spherical protuberance of 5 cm in diameter in the transverse colon (Figure 2).

MULTIDISCIPLINARY EXPERT CONSULTATION

Combined with the microscopy findings and considering the high risk associated with endoscopy, after discussion with the pediatric surgeons, pediatricians, pathologists, and ultrasonographers, we decided to remove the tumor *via* general surgery.

FINAL DIAGNOSIS

PEComa was diagnosed by immunohistochemistry.

TREATMENT

We performed surgery on the patient, a tumor was found in the transverse colon near the spleen, of about 6 cm × 4 cm × 3 cm in size, with a wide pedicle connected to the bowel. It had good mobility, a hard texture, and a rich blood supply. Edema of the surrounding bowel wall and mesentery was found, separating the mesentery in turn and being ligated to the affected mesenteric vessels. We completely

removed the tumor, in addition to about 3 cm of the affected bowel (Figure 3).

Postoperative pathology showed that the tumor volume was 5.0 cm × 4.5 cm × 3.0 cm and the tumor tissue was located in the submucosa of colon, arranged in an acinar shape with mild cell morphology, no tumor necrosis, and rare instances of mitosis. The immunohistochemical staining results were as follows: Human melanoma black 45 (HMB-45) (+), cluster of differentiation 31 (CD31) (+), cytokeratin (-), melanoma-associated antigen recognized by T cells (-), smooth muscle actin (-), molleya (-), desmin (-), S-100 (-), CD117 (-), and Ki67 (hot spot positive rate < 5%) (Figure 4).

OUTCOME AND FOLLOW-UP

The patient recovered well after the operation, and no abnormalities were found at the 6 mo follow-up.

DISCUSSION

PEComa represents a group of mesenchymal tumors characterized by perivascular epithelioid cells[1]. The etiology is still unclear, and some scholars consider it to be related to the gene mutation of the tuberous sclerosis complex[2]. Histologically, it is mainly composed of blood vessels, spindle cells or epithelioid cells, and fat. The proportion of the three components varies, which leads to large differences in imaging manifestations; it can manifest as poorly differentiated soft tissue tumors or as sclerosing tumors. Its density or signal performance is also closely related to the tumor cell components, but most of these tumors are characterized by a soft tissue mass with a regular shape, clear boundary, high density, and low signal intensity[3].

PEComa diagnosis depends on the pathology and immunohistochemistry findings. According to the World Health Organization classification of digestive system tumors published in 2019, the basic and ideal diagnostic criteria of PEComa are: epithelioid cells and (or) spindle cells in tissues, eosinophilic granular or transparent cytoplasm; nestlike, trabecular or lamellar structure; and co-expression of melanocytes and smooth muscle markers[4]. At present, there is no definitive standard for the diagnosis of benign and malignant PEComa. Folpe *et al*[5] divided the tumors into benign, malignant, and undetermined malignant potential. The malignant features included: tumor size > 5 cm, marginal infiltration, atypical nuclear, mitotic image ≥ 1/50 high-power field, tumor necrosis, and vascular invasion. Benign tumors are considered malignant when they have more than two of the aforementioned features; cases where the diagnosis of malignant potential is uncertain and there is tumor necrosis, including obvious nuclear atypia and high proliferation index, need close follow-up[4]. Considering the pathological results of this case, we considered the tumor to be benign; however, due to the patient's young age and large tumor volume, close follow-up is still needed.

PEComa is rarely reported. Cecal PEComa was first reported by Birkhaeuser *et al*[6] in 2004. Since then, a total of 30 cases (Table 1)[6-32] of colorectal PEComa have been reported (as determined upon performance of a detailed PubMed search), including 18 females and 12 males, of ages ranging from 5.5-years-old to 69-years-old; most of these patients were adults, and only 7 (23%) were younger than 15-years-old. There was a significant sex difference among the adults but no significant sex difference among the children, consistent with the findings reported by Fadare[33], who proposed that PEComa may be a hormone-dependent tumor. PEComa can occur in all parts of the colon, although they occur more often in the left colon (9 cases in the sigmoid colon[11,12,15,16,18,21,22,26,31], 5 in the rectum[6,7,13,28,29], 3 in the descending colon[9,16,27], 4 in the ascending colon[16,19,30], 7 in the cecum[8,14,20,23-25,32], 1 in the transverse colon[10], and 1 in the right colon[17]).

The diameter of the reported tumors have ranged from 0.8 cm to 8.0 cm. There are no specific symptoms. The tumor can manifest abdominal pain, diarrhea, abdominal distension, hematochezia, or other symptoms of gastrointestinal tumors[19]. The most common clinical manifestation is abdominal pain (44%). Two intussusception cases have been reported. Among the 30 cases, 23 patients underwent surgery and 4 of them given postoperative adjuvant chemotherapy[14,19,26,31]. In total, 5 underwent endoscopic mucosal resection[10,20,24,25,30], and 1 patient underwent endoscopic mucosal dissection after pathological diagnosis. No recurrence was found during follow-up. There have been 10 malignant PEComa cases reported[8,9,11,14,15,19,23,26,28,31]; among them, 2 patients died[9,23] and 2 were lost to follow-up but involving the pancreas and liver metastasis respectively[28,31]. Combined with limited case analysis, the prognosis of malignant PEComa is poor.

Ileocolic intussusception is one of the most common abdominal emergencies involving children who are less than 3-years-old[34]. The pathophysiology underlying the majority of pediatric intussusception cases is thought to be secondary to a transient viral illness[35]. In adults, 70%-90% of intussusception can be found to have a clear cause, and about 40% are caused by a primary or secondary malignant tumor[36]. Here, we have reported the first pediatric case of benign PEComa in the transverse colon with intussusception, tumor prolapse, and incarceration outside the anus.

Table 1 Review of case reports of colorectal perivascular epithelioid cell tumor

Ref.	Age (yr)	Sex	Symptom	Location	Size (mm)	Metastasis	Treatment	Follow-up
1 Birkhaeuser *et al* [6]	35	F	Bleeding	Cecum	35	No	SR	NER at 5 yr
2 Genevay *et al*[7]	36	F	Anemia and rectorrhagia	Cecum	35	No	SR	NA
3 Evert *et al*[8]	56	F	Rectal obstruction loss	Rectum	80 × 50	Lung metastasis	NA	NA
4 Yamamoto *et al* [9]	43	F	Abdominal pain	Descending	80	No	SR	DOD at 38 mo
5 Baek *et al*[10]	16	F	NA	Transverse	25	No	ER	NER at 24 mo
6 Pisharody *et al* [11]	11	M	Bleeding	Sigmoid	30	Lymph node metastasis	SR	NER at 5 mo
7 Righi *et al*[12]	11	M	NA	Sigmoid	35	NA	SR	NA
8 Qu *et al*[13]	43	F	NA	Cecum	20	No	SR	NER at 25 mo
9 Ryan *et al*[14]	15	F	Bleeding	Rectum	37	Lymph node metastasis	SR and AC	NER at 5 mo
10 Tanaka *et al* [15]	14	F	Physical examination	Sigmoid	40	No	SR	NA
11 Shi *et al*[16]	38	F	Abdominal pain	Ascending	60	No	SR	NER at 8 mo
12 Shi *et al*[16]	42	M	Abdominal pain	Sigmoid	45	No	SR	NER at 15 mo
13 Shi *et al*[16]	36	M	Abdominal pain	Descending	48	No	SR	NER at 32 mo
14 Shi *et al*[16]	45	F	Abdominal pain	Ascending	35	No	SR	NER at 36 mo
15 Gross *et al*[17]	5.5	M	Abdominal pain and fever	Right	50	No	SR	NER at 15 yr
16 Freeman *et al* [18]	17	F	Bleeding	Sigmoid	NA	No	SR	NA
17 Park *et al*[19]	7	M	Abdominal pain and bleeding	Ascending	37	No	SR and IFN therapy	NER at 26 mo
18 Mar *et al*[20]	11	F	Prolapsed mass	Rectum	20	No	ER	NA
19 Lee *et al*[21]	62	F	Abdominal pain and melena	Sigmoid	50	NA	NA	NA
20 Cho *et al*[22]	62	F	Bleeding	Sigmoid	50	No	SR	NER at 16 mo
21 Scheppach *et al* [23]	23	M	Abdominal pain and bleeding	Rectum	NA	Lymph node and liver metastasis	SR and AC	DOD at 23 mo
22 Im *et al*[24]	17	M	Bleeding	Rectum	30	No	ER	NER at 10 mo
23 Kanazawa *et al* [25]	55	F	Physical examination	Rectum	25	No	ER	NER at 12 mo
24 Cheng *et al*[26]	40	M	Dyschezia	Sigmoid	70 × 60	No	SR	Pancreatic metastasis at 27 mo
25 Iwamoto *et al* [27]	42	F	Physical examination	Descending	NA	No	SR	NA
26 Lin *et al*[28]	28	M	Abdominal pain and bleeding	Cecum	88	No	SR	Liver metastasis at 49 mo
27 Iwa *et al*[29]	69	M	Physical examination	Cecum	41 × 32	No	SR	NA
28 Bennett *et al* [30]	67	F	Physical examination	Ascending	80	No	ER	NA
29 Cheng *et al*[31]	17	M	Bleeding	Sigmoid	NA	Lymph node metastasis	SR and AC	NER at 24 mo
30 Yeon *et al*[32]	45	F	Physical examination	Rectum	20	No	SR	NA

AC: Adjuvant chemotherapy; DOD: Died of disease; ER: Endoscopic resection; F: Female; M: Male; NA: Not available; NER: No evidence of recurrence; SR: Surgical resection.

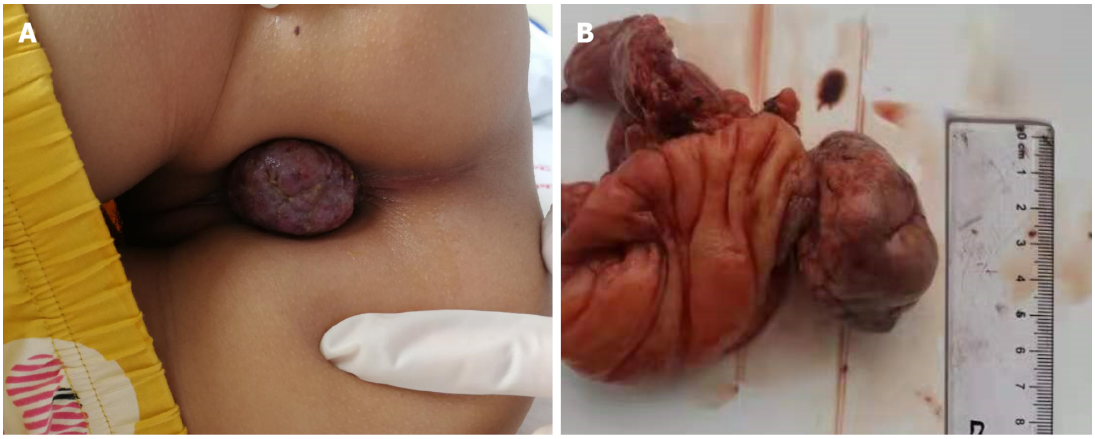

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Figure 3 Tumor. A: The tumor was outside the anus; B: The tumor was removed surgically, in addition to about 3 cm of the affected bowel.

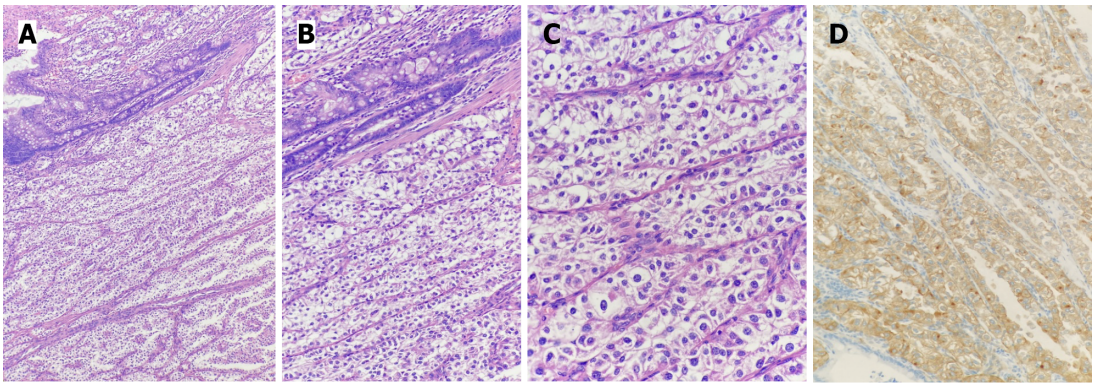

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Figure 4 Pathology and immunohistochemistry results. A: 40 × magnification showing that the tumor was located in the intestinal wall, and the tumor cells were arranged in nests or acini; B: 100 × magnification showing that the tumor cells were transparent or eosinophilic granular; C: 200 × magnification showing abundant capillaries in the interstitium; D: Human melanoma black 45 (+) detected by the EnVision method.

At present, benign PEComa has no adjuvant drug treatment. The main treatment for colon PEComa is radical resection, with a good prognosis. Long-term clinical and CT follow-up is recommended. Scheppach *et al*[23] administered sirolimus, doxorubicin, ifosfamide, citabine and docetaxel successively after surgery, which had no obvious effect. Park *et al*[19] reported on a 7-year-old boy with poorly differentiated PEComa in the ascending colon, who received adjuvant interferon-alpha for 1 year after surgery. There was no recurrence after 26 mo of follow-up. That was the first report of interferon-alpha for the treatment of PEComa in the colon. In recent years, an increasing number of targeted drugs have been used in PEComa. Studies have shown that mechanistic target of rapamycin inhibitors are the most effective drugs for the treatment of advanced/metastatic PEComa[37].

CONCLUSION

PEComa is a special type of mesenchymal tissue tumor, which is rarely encountered in the clinic and lacks specific clinical manifestations. The diagnosis depends on pathology and immunohistochemistry findings. Radical resection is the preferred treatment method, and there is no standardized treatment for postoperative adjuvant therapy. Targeted drug application is gradually increasing and has achieved certain results but still needs further research.

FOOTNOTES

Author contributions: Kou L analyzed the data and wrote the paper; Zheng WW, Jia L, and Gao FY revised the paper; Wang XL, Zhou JH, Hao JR, and Liu Z collected the patient's clinical data; Gao FY designed the report.

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