

Unilateral Cystic Teratoma of the Ovary in Children: A Case Report

¹Lilia Dewiyanti, ^{2*}Cipta Pramana, ¹Muhammad Akbar, ¹Felisitas Sinta, ¹Laurensius

¹Department of Pediatrics, KRMT Wongsonegoro General Hospital Semarang, Faculty of Medicine Tarumanagara University, Jakarta, Indonesia

²Department of Obstetrics and Gynecology, KRMT Wongsonegoro General Hospital Semarang, Faculty of Medicine Tarumanagara University, Jakarta, Indonesia

ABSTRACT

Teratoma is a type of extracranial germ cell tumor that often occurs and correlates with age and anatomical location. Mature Cystic Teratoma (MCT) is the most common ovarian tumor in children and adolescents. The most common sites for teratomas are the sacrococcygeal region and the ovaries. Laparoscopy is by far the surgical treatment of choice for teratomas. We report a case of a 9-year-old girl with a chief complaint of left lower quadrant abdominal pain 1 week before hospital admission. The patient appears weak and has decreased appetite, accompanied by weight loss, and painful urination. Physical examination of abdominal distension (+), tenderness, and release in the left lower quadrant (+). The patient had not menstruated, and a history of vaginal discharge was denied. On MSCT-scan examination of the abdomen with contrast, a cystic mass was found that adhered to and pushed the surrounding bowel structures in the lower abdominal region. On histopathological examination taken from the left ovary preparation, a macroscopic examination found cyst tissue of 9.5 x 6.5 x 5 cm. The cyst is gray black, filled with blood and a white pulp mass with hair. On microscopic examination found cysts with fibrous connective tissue stroma containing vascular. The treatment is in the form of laparoscopy and after surgery, currently, the patient is in good condition and will be followed up again every 3 months.

Corresponding Author: pramanacipta@yahoo.com

How to cite this article: Dewiyanti L, Pramana C, Akbar M, Sinta F, Laurensius. Unilateral Cystic Teratoma of the Ovary in Children: A Case Report. Journal of Complementary Medicine Research, Vol. 13, No. 4, 2022 (pp. 28-31).

INTRODUCTION

Teratoma is a type of extracranial germ cell tumor that often occurs and correlates with age and anatomical location. Teratomas are composed of layers of endodermis, mesoderm, and ectoderm which can be mature (malignant) and immature (benign or malignant). Teratomas usually occur in the coccyx or sacrum in neonates, testes, or ovaries in early puberty, and in rare areas such as the mediastinum, retroperitoneum, liver, and heart (National Cancer Institute, 2021; Rescorla, 2012). The prevalence of germ cell tumors is about 1% of malignancies in children with a prognosis that varies according to the initial diagnosis and management (Permono, Sutaryo, Windiastuti, & Abdulsalam, 2010). The most common teratoma in children is ovarian teratoma, and about 80-90% of the tumor mass is benign (Rescorla, 2012). We report a case of a 9-year-old girl who had ovarian teratoma and underwent laparoscopic surgery (Beger, Karaman, Kızılyıldız, Şimşek, & Düz, 2020).

KEYWORDS:

Extracranial germ cell tumor,
Laparoscopy
Mature Cystic Teratoma,
Teratoma.

ARTICLE HISTORY:

Received : Apr 15, 2022
Accepted : Jun 10, 2022
Published: Sep 11, 2022

DOI:

10.5455/jcmr.2022.13.04.06

CASE PRESENTATION

A 9-year-old girl came to Wongsonegoro General Hospital with the complaint of abdominal pain in the left lower quadrant 1 week before the hospital admission. The patient also had difficulty defecating 5 days ago and he had a history of eating spicy food before the complaint arose. The patient appears weak and has decreased appetite, accompanied by weight loss, and dysuria. The patient had been treated previously and given medicine for the gastric but there's no significant difference. The patient didn't have fever, nausea, vomiting, coughing, runny nose, diarrhea, or bloody stools. The patient had not menstruated, and a history of vaginal discharge was denied. The patient had a history of gastritis several years ago. The results of physical examination, the patient looked weak, compos mentis consciousness, GCS 15 (E4M6V5), pulse 113 x/minute, temperature 37°C, respiratory rate 20 x/minute, Spo2 99%. On generalized status examination, abdominal distension (+), tenderness and bound tenderness in the left lower quadrant (+), Mcburney sign (-), Rovsing sign (-) Muscular defense (-), Obturator sign (-), Blumberg's sign (-), no lumps or organomegaly were seen. On hematological examination, renal function, and electrolyte levels were all within normal limits. HCG and AFP tests were not performed.

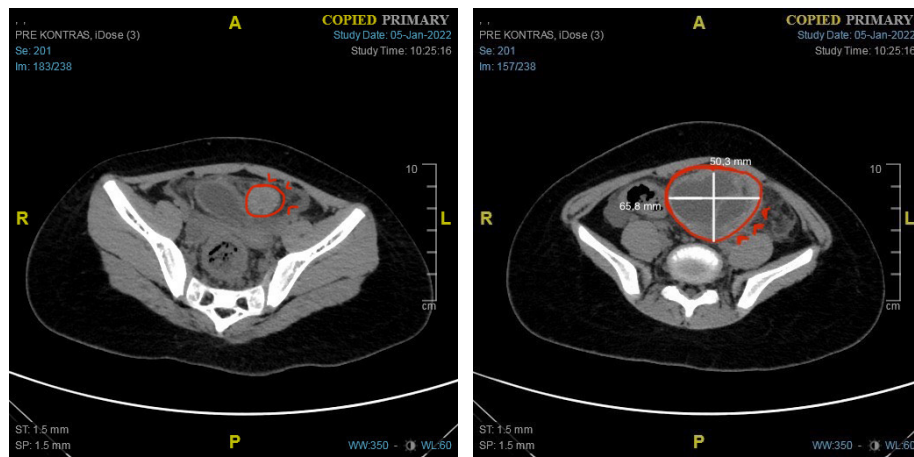


Fig.1: On the MSCT-Scan with contrast, a cystic mass with a fatty component and calcifications in it is lobulated with a partially defined border, and irregular edges.

An abdominal x-ray examination revealed a meteorism with prominent fecal material and an abdominal MSCT scan with contrast found a cystic mass was found with a fat component and calcifications in it in the form of lobulated, partially defined irregular edges accompanied by septation (size \pm AP 5.41 CC 8.6 cm LL 9.26 cm) that adhered and forced the surrounding bowel structure in the lower abdominal region, tended to be teratoma. Multiple lymphadenopathies in interaortocaval, right and left mesentery (largest size \pm 2.15 cm x 1.04 cm) (Figure 1).

On histopathological examination taken from the left ovary preparation, on macroscopic examination found cyst tissue of 9.5 x 6.5 x 5 cm with a tube length of 6 cm, a diameter of 0.3 cm. Cysts are gray black, on cutting in the form of bleeding areas and extensive infarcts are red black, filled with blood and a white pulp mass with hair. On microscopic examination found cysts with fibrous connective tissue stroma containing vascular. Most of the area within the cyst has been infarcted and hemorrhaged, making it difficult to analyze. Areas with keratinized structures and fatty tissue are seen. Unclear immature/embryogenic germ layer. There is no sign of malignancy in this preparation. The diagnosis in this patient was left ovarian cystic teratoma. The patient's therapy is surgery to remove the mass through laparoscopy. No chemotherapy or radiotherapy was performed on the patient. After follow-up currently, the patient's condition is improving, and there are no complaints of abdominal pain.

DISCUSSION

The most common germ cell in the pediatric population is a teratoma. The gonads and extragonadal are the most common sites for these neoplasms, and these locations are thought to correspond to the resting sites of primordial totipotent germ cell embryos. The incidence of this teratoma is closely related to age and anatomic location. In infancy and toddlerhood, teratomas generally appear in the extragonadal site, whereas in childhood, they usually occur in the testes or ovaries (Grosfeld, O'Neill, Fonkalsrud, & Coran, 2006). Mature Cystic Teratoma (MCT), or dermoid cyst (predominant ectodermal element), is an ovarian tumor that occurs in infancy and toddlerhood. It is most common in children and adolescents, accounting for approximately 50% of all pediatric ovarian neoplasms. MCT is bilateral in up to 10% of pediatric cases (Laufer & Goldstein,

2005). In a study conducted at a hospital in Indonesia in 2020, it was found that the prevalence of ovarian cysts in children was around 20% and the majority occurred in the left ovary (Cahyani, Sriwidyani, Mahastuti, & Saputra, 2022).

Another type of teratoma, namely immature teratoma usually affects younger age groups; the younger the patient, the more likely the teratoma to be an immature germ cell type (Laufer & Goldstein, 2005). Immature teratomas are common in the 10- and 20-year-old population, with a mean age of 17 years, representing 10%-20% of all ovarian malignancies in patients less than 20 years. This type of neoplasm tends to be more aggressive, and the prognosis is usually worse than mature teratomas (Gershenson et al., 1986; Heo et al., 2014).

The most common sites of teratoma are the sacrococcygeal region with an incidence of about 45-65%, anterior mediastinum 10-12%, gonads (ovaries and testes) 10-35%, retroperitoneum 3-5%, cervical area 3-6%, pre-pregnancy area. sacral 3 - 5%, central nervous system 2 - 4%, quite rare places, such as liver, kidney, vagina, stomach that is < 1%. Approximately 75% of teratomas occur in the sacrococcygeal area in prepubertal children (Shamberger, 2004). In this case, a gray-black cyst was found in the left ovary area with a size of 9.5 x 6.5 x 5 cm. In several studies, the ovary is one of the most common locations, which is about 10 - 50% of all neoplasms that occur in pediatrics (Grosfeld et al., 2006; Laufer & Goldstein, 2005).

The cause of ovarian teratoma is due to migration of primordial germ cells from the yolk sac to the gonads during the embryogenesis process (Morrow et al., 2017). The most common symptoms experienced by patients are abdominal pain, palpable mass, and abdominal distension. Other symptoms that can arise are fever, malaise, weight loss, nausea, vomiting, uterine bleeding, and urinary tract disorders (Łuczak & Baglaj, 2018) their appropriate treatment in this age group still remains unclear. Paucity of research dedicated exclusively to both mature and immature teratomas of the ovary, contribute to decision making difficulties. Therefore, we decided to review retrospectively our experience in treatment of pediatric ovarian teratomas in order to assess the epidemiology, presenting features, and diagnostic as well as surgical management of these lesions. Results: The study comprised 58 patients. Fifty percent of patients were between 9 and 15 years old. Mature teratoma was diagnosed

in 55(94.83%. In some cases, with small teratoma, symptoms can be asymptomatic, therefore comprehensive evaluation is needed. In this case, the patient complained of abdominal pain that felt like being stabbed all the time, pain was also felt on palpation, abdominal distension, painful urination, and weakness. At the time of taking a history on a patient with complaints of chronic progressive abdominal pain which gradually worsened, with normal ultrasound results and abdominal x-rays, as clinicians we can suspect a tumor in the intra-abdominal or pelvic area.

In diagnosing ovarian teratoma, radiological examination in the form of ultrasound or abdominal CT scan can be carried out (Budiananti, Budiananti, & Matulatan, 2021). CT scan has high sensitivity in tracking teratomas. The features that can be found on a CT scan of mature teratoma are fat inside the cyst or the presence of Rokitanski nodules (dermoid plugs that protrude into the cystic space, in which there can be hair, teeth, and bone), well-defined. On histopathological examination can be found a unilocular cystic cavity, a tumor cavity filled with sebaceous-like oily fluid, made of keratin, sebum, or hair surrounded by capsule), the squamous epithelium lining the tumor wall, presence of ectodermal elements (nerve tissue & skin), endodermal (bone tissue), fat, muscle, cartilage), and mesodermal (gastrointestinal and thyroid) (Saba et al., 2009; Sahin, Abdullazade, & Sancı, 2017) each radiological feature reflects a specific pathologic equivalent that forms because of diverse compositions of histological components. Understanding uncommon findings as well as the classic signs with basic knowledge of pathological equivalents permits a more accurate diagnosis and guides adequate treatment. In this review, radiological features of MCT in different imaging modalities (US, CT, MR imaging. In this case, we found a cystic mass with a fatty component and calcifications within and in a lobulated form with partially defined borders, irregular edges on a CT scan with contrast, and a gray-colored cyst, blackish ash, filled with blood and white pulp mass with hair, visible areas with keratin structure and fatty tissue, with no clear immature/embryogenic germ layer.

The difference between benign and malignant tumors is that benign tumors have characteristics of slow growth, flat edges, and do not invade surrounding tissues and other organs, whereas malignant tumors have fast growth, irregular edges, often invade surrounding tissues, and can metastasize to other organs. Treatment for benign tumors can be done by monitoring and performing surgery, while in malignant tumors because they can spread quickly and are detected early, the treatment may be surgery with the possibility of chemotherapy or radiotherapy. If the tumor has spread, treatment tends to be systemic, such as chemotherapy or immunotherapy (Patel, 2020).

The most effective treatment of ovarian teratoma in children is surgery. Indications for surgery include persistent or intermittent abdominal pain and a mass greater than 5 cm in size. Laparoscopy is the gold standard in ovarian teratoma surgery. Laparoscopy minimizes the occurrence of severe intraoperative bleeding, minimizes pain and hospitalization, and in terms of cosmetics the resulting scar is relatively small (Beger et al., 2020). On the macroscopic examination, we found a cyst tissue 9.5 x 6.5 x 5 cm with long tubes 6 cm, 0.3 cm

in diameter, and intermittent abdominal pain, so laparoscopic surgery was performed.

Torsion is a complication of an ovarian tumor that causes its rotation around the pedicle (supporting ligament), interfering with its blood flow and leading to gangrenous or hemorrhagic infarction. The risk of infection in cystic teratomas is about 1 to 4%. The route of infection can be hematogenous, lymphomatous, or by direct extension from adjacent structures such as the gut. Adhesions cause the cyst to depend on the blood supply of the omentum or surrounding structures by establishing a contralateral circulation and completely detaching from the pedicle. Anti-N-methyl-D-aspartate receptor (NMDAR) encephalitis is a rare autoimmune complication with neuropsychiatric symptoms (Mobeen & Apostol, 2022). Complications experienced after laparoscopic procedures include incisional hernia, infection, peritoneal irritation, and peritonitis. Infection is caused because laparoscopic surgery is generally performed at the umbilicus, which is the site of skin infection (Selva, 2016; Parker, Childers, Canis, Phillips, & Topel, 1996).

Mature cystic tumors provide a better prognosis than immature cystic tumors (Donnellan & Swenson, 1988). However, because all GCTs have a risk of local recurrence and distant recurrence, monitoring or follow up at 3-month intervals for 3-4 years is necessary. According to the findings reported by Rescorla et al., a Children's Cancer Group study recorded a tumor recurrence rate of 11% with mature teratomas and a recurrence rate of 4% in immature teratomas. Although 43% to 50% of these recurrences are malignant, chemosensitivity of yolk sac (endodermal sinus) tumors results in a high survival rate. Serum AFP levels should be monitored, and a physical examination performed at each check-up. Abdominal ultrasound should be performed when AFP levels do not fall properly. When there is an index of suspicion, an abdominopelvic CT or MRI is performed, as well as a CT scan of the lungs. Recurrent tumors may be benign but must be re-excised to minimize the risk of long-term malignant transformation (Rescorla, Sawin, Coran, Dillon, & Azizkhan, 1998).

CONCLUSION

Teratoma is a GCT that is quite common in the pediatric population with a common location in the ovary and sacrococcygeal. Teratoma was diagnosed based on history, clinical symptoms, radiological and histopathological examination (gold standard). Currently, the management of teratoma is quite developed and laparoscopy is one of the options that is often done, with average satisfactory results, risk of complications, and minimal recurrence. The patient's current condition after laparoscopy has improved and no chemotherapy or radiotherapy was performed, but monitoring is still done every 3 months.

Acknowledgment : Thank you to the director of Wongsonegoro Hospital Semarang for allowing us to conduct research and to the patient's family for giving permission to publish the case.

Ethical Approval: This case report has received approval from the ethics committee of the KRMT Wongsonegoro Hospital Semarang

Informed consent: This study has obtained the consent of the patient's parents

Authorship contributions: All the listed authors have made substantial, direct and intellectual contributions to the work and approved it for publication.

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