

Ovarian teratoma as a cause of abdominal pain in preschool children

Teratoma ovariano como causa de dor abdominal em criança na idade pré-escolar

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ABSTRACT

Introduction: Ovarian tumors, rare in childhood, have a higher incidence between 8 and 9 years, peaking at 19 years. The main symptomatology is manifested by abdominal pain, palpable mass, fever, constipation, and more rarely polaciuria and dysuria. Mature ovarian teratomas although they are mostly benign neoplasms, if poorly conducted, they can progress to surgical emergencies. **Objective:** To analyze mature ovarian teratoma as a possible etiology of chronic abdominal pain in pediatric patients. **Case Report:** Child, 2 years and 9 months, taken to medical care with report of chronic abdominal pain, three months of evolution, with periods of sharpness, of strong intensity, associated with dysuria and polaciuria and primary suspected urinary tract infection (UTI). From the compatible clinic, associated with leukocytes, though there is no bacterial growth in the cultures, due to the recurrence of the condition, appropriate propaedeutic investigation of repeat UTI was followed by screening for urinary pathway malformations. Total abdominal ultrasound with urinary tract dynamics was requested, which that found the presence of a large expansive formation probably originating in the left ovary. Extended diagnostic research, the computed tomography of the abdomen and pelvis was performed with findings suggestive of left ovarian teratoma exerting local expansive behavior with deviation and compression of surrounding anatomical structures. With the diagnostic hypothesis evidenced, she was referred to pediatric surgery and oncology for appropriate treatment with oophorectomy and clinical follow-up. **Conclusion:** Importance of adequate propaedeutics for cases of chronic abdominal pain in pediatric patients due to the extent of possible differential diagnoses.

Keywords: Teratoma; Abdominal Pain; Pediatrics.

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RESUMO

Introdução: Os tumores ovarianos, raros na infância, apresentam maior incidência entre 8 e 9 anos, tendo pico aos 19 anos. A sintomatologia principal manifesta-se por dor abdominal, massa palpável, febre, constipação e mais raramente polaciúria e disúria. Os teratomas ovarianos maduros embora sejam neoplasias majoritariamente benignas, se malconduzidas, podem evoluir para emergências cirúrgicas. **Objetivo:** Analisar o teratoma ovariano maduro como possível etiologia de dor abdominal crônica em pacientes pediátricos. **Relato de Caso:** Criança, 2 anos e 9 meses, levada ao atendimento médico com relato de dor abdominal crônica, 3 meses de evolução, com períodos de agudização, de forte intensidade, associada à disúria e polaciúria conduzida como principal suspeita infecção do trato urinário (ITU). A partir da clínica compatível, associado a leucocitúrias, mesmo não havendo crescimento bacteriano nas culturas, devido à recorrência do quadro, seguiu-se a propedêutica adequada de investigação de ITU de repetição, através do rastreio de má formações das vias urinárias. O ultrassom abdominal total com dinâmica do trato urinário constatou presença de volumosa formação expansiva originada provavelmente em ovário esquerdo. Estendida a pesquisa diagnóstica foi realizada tomografia computadorizada do abdome e pelve com achados sugestivos de teratoma ovariano esquerdo, exercendo comportamento expansivo local com desvio e compressão de estruturas anatômicas circunvizinhas. Com a hipótese diagnóstica evidenciada foi encaminhada à cirurgia pediátrica e oncologia para tratamento adequado, com realização de ooforectomia e seguimento clínico. **Conclusão:** Importância da propedêutica adequada para casos de dor abdominal crônica em pacientes pediátricos devido à extensão de possíveis diagnósticos diferenciais.

Palavras-chave: Teratoma; Dor Abdominal; Pediatria.

INTRODUCTION

Chronic abdominal pain is a recurrent complaint in pediatric patients presenting countless diagnostic hypotheses to be investigated. Diagnostic investigation should be initiated with a view to differentiating abdominal pain from organic causes to functional ones. Carrying out an incredibly detailed anamnesis, with a complete medical history, thorough physical examination and achievement of complementary exams lead to a better diagnostic clarification¹.

A diagnostic hypothesis to be considered in cases of chronic abdominal pain in pediatric patients, despite its low incidence, are the neoplasms, among them, the ovarian tumors, which have a higher incidence between 8 and 9 years, peaked at 19 years old. The main symptoms are manifested by abdominal pain, and may be accompanied by a palpable mass, fever, constipation, vaginal bleeding

and, more rarely, polaciuria and dysuria. Usually they are originating from germ cells, preferentially located in the gonads, dysgerminomas, tumors of the endodermal sinus, immature teratomas, mixed germ cell tumors and embryonic carcinomas are the most common types in pediatrics².

Mature ovarian teratoma or dermoid cyst is a benign tumor, most common in women of childbearing age, composed of tissues from the three germ layers (ectoderm, mesoderm and endoderm) which may also contain adipose tissue, hair, teeth, and other cells derived from other tissues. Usually it does not exceed 10 cm in diameter with clinical presentation as asymptomatic which had been detected incidentally on imaging studies³. They present in most cases unilaterally⁴. Symptoms such as pain, are more commonly found, when it presents torsions, rupture or infection. The compression of adjacent structures is more frequent when the mass can be found in large dimensions³.

CASE REPORT

Child, 2 years 9 months, accompanied by his mother, seeks medical attention with complaints of acute abdominal pain, in colic, of severe intensity associated with dysuria and polaciuria. In the clinical history, it is reported chronic abdominal pain, three months of evolution, with periods of exacerbation, and primary suspected urinary tract infection (UTI), without laboratory confirmation, as demonstrated in the tests attached to Table 1. On physical examination, a patient with good general condition, no fever, facial expression of pain, normotensive abdomen, absence of a palpable mass with diffuse painful deep palpation.

As shown in Table 1, despite presenting symptoms compatible with UTI and sedimentoscopy with positive leukocytes and presence of pyocytes there was no bacterial

growth in culture, thus not establishing the diagnosis of infection. However, due to the clinical manifestations suggestive and recurrence of the condition, the patient was, in some episodes, treated inappropriately, with empirical antibiotic therapy for UTIs and symptomatic, in their various interconsultations in emergency care. As the patient had been attended only in emergency services by different professionals adequate propaedeutics and chemoprophylaxis measures were not performed.

From acute abdominal pain, a clinic compatible with UTI, associated with the finding of piuria in 4 of the 5 urinary samples collected in the period of 3 months and, considering that the predictive value of this varies between 40 and 80%, may be absent in 23 to 50% of patients with bacteriuria and UTI, we chose to follow with the

Table 1. Laboratory tests performed in the investigation of UTI.

Date	Urine routine	Gout Gram	Uroculture	Urea	Creatinine	CBC	CRP
07/17/07	Negative nitrite Leukocytes ++ Countless pyocytes Bacterial flora greatly increased	Negative nitrite Leukocytes ++ Countless pyocytes Bacterial flora greatly increased	There was no bacterial growth	29,0 mg/dL	0,8 mg/dL	HGB 10,6 HCT 31,3% WBC 11.900 Band 238 Segmented neutrophils 5.235 PLT 220.000	96,0 mg/L
22/17/07	Negative nitrite Leukocytes + Pyocytes 10/ field Slightly increased bacterial flora	Rare Gram- negative rods and some Gram- positive coconuts	There was no bacterial growth	-	-	HGB 12,1 HCT 37,6% WBC 10.140 Band neutrophils 0,0 Segmented 6.084 PLT 327.000	48,0 mg/L
30/17/07	Negative nitrite Piocytes 6/field Normal bacterial flora	Absence of microorganisms cordoned by the method	There was no bacterial growth	-	-	HBC 12,6 HCT 37,8% WBC 13.100 Band neutrophils 0,0 Segmented neutrophils 4.061 PLT 312.000	< 6,0 mg/L
03/17/09	Negative nitrite Leukocytes ++ Numerous pyocytes/ field Slightly increased bacterial flora	Absence of microorganisms cordoned by the method	There was no bacterial growth	30 mg/ dL	0,47 mg/ dL	HBC 10,5 HCT 31,1% WBC 10.320 Band neutrophils 0,0 Segmented neutrophils 5.160 PLT 323.000	24,0 mg/L
14/17/10	Negative nitrite Negative leukocytes Pyocytes 4/field Slightly increased bacterial flora	Absence of microorganisms cordoned by the method	There was no bacterial growth	-	-	HBC 10,8 HCT 31,6% WBC 4.810 Band neutrophils 0,0 Segmented neutrophils 3.078 PLT 166.000	< 6,0 mg/L

Source: Prepared by the authors.

investigation propaedeutics of retry urinary infection, three episodes in 12 months or two or more in 6 months. It then followed with the screening for urinary tract malformations and ultrasound (US) with total abdominal dynamics with urinary tract dynamics is requested.

The US found kidneys with normal parenchyma, bladder with normal anatomical aspect, pielocalinitial systems and ureters with normal functional aspect, bladder with fullness capacity as expected for the child's age, negligible residual urine volume. There were no signs of increased intravesical pressure, ruling out a diagnostic hypothesis of malformation of urinary tract.

US was observed with the presence of a large expansive formation probably originating in the left ovary, heterogeneous, of etiology to be clarified, having ovarian teratoma as a suspected diagnosis, as can be seen in Figures 1 and 2.

Extended diagnostic research was performed by computed tomography (CT) of the abdomen and pelvis without oral or venous administration of water-soluble iodized contrast medium.

CT, the left ovary was not visualized with the usual topography, with a complex and grossly oval mass, with precise limits measuring 4.7 x 3.2 x 5.1 cm in their major antero-posterior (AP), lateral-lateral (LL) and longitudinal axes, respectively. Mass of heterogeneous density, highlighting ovoid and more hypoattenuating formations suggesting cystic nature and separated by non-uniform septa with soft tissue densities. More center-caudal region of the mass showed an oval image with precise limits, lobulated contours and fat density measuring 1.6 x 1.7 x 1.3 cm in the AP, LL and longitudinal axes with an image of a radiodense and malformed tooth in their intimacy.

All findings described by CT therefore characterize left, mature ovarian teratoma, exerting local expansive behavior, causing deviation and compression of surrounding anatomical structures, thus justifying the symptoms presented by the patient of colic abdominal pain, dysuria and polaciuria in addition to the recurrent urinary infections caused by the compression of the mass in the ureter.

CT, the right ovary had the usual topography, regular contours and anatomical dimensions of 1.5 x 1.3 cm in its major axial axes. It presented some small oval and hypodense formations in between, suggesting follicles, the largest being 9.5 mm (Figure 3). Bladder with good water repletion, presenting regular and normothick walls. Homogeneously hypodense content. The most postpartum-left-lateral part of the bladder is somewhat compressed by heterogeneous pelvic mass. Absence of lymphadenomegalias in the main abdominal and pelvic sites. Absence of ascites liquid.

Eutopic kidneys, of usual shape and dimensions, presenting regular contours and normodense parenchyma. There is no evidence of hydronephrosis or even images of calculations. Free peri-reinspaces (Figure 6). Visible ureters throughout its path, of usual caliber, without caulcic foci in



Figure 1. Abdominal US with urinary tract dynamics: expansive formation in left pelvis.



Figure 2. Abdominal US with urinary tract dynamics: expansive formation in left pelvis.

its interior. Other abdominal and pelvic structures showed no alterations on the examination. The tomographic findings described above can be seen in the figures below. Figure 3. Left ovary with unusual topography with complex and grossly oval mass, of heterogeneous density, stressing ovoid and more hypoattenuating formations suggesting cystic nature and separated by non-uniform septations with soft tissue density. Right ovary with usual topography, regular contours and anatomical dimensions.

With a diagnostic hypothesis of mature ovarian teratoma evidenced on CT patient was referred to elective pediatric surgery, by the absence of signs suggestive of acute abdomen, and oncology for appropriate treatment. The preoperative measurement of tumor markers was not performed in the event of unavailability of this test in the public health scenario in which the patient was inserted. It was then continued with the surgical approach by laparotomy oophorectomy, Pfannenstiel incision (Figure 7), no need for left salpingectomy, preserving the ipsilateral fallopian tube. No signs of tumor implantation were found in the rest of the patient's pelvis, followed by clinical follow-up.

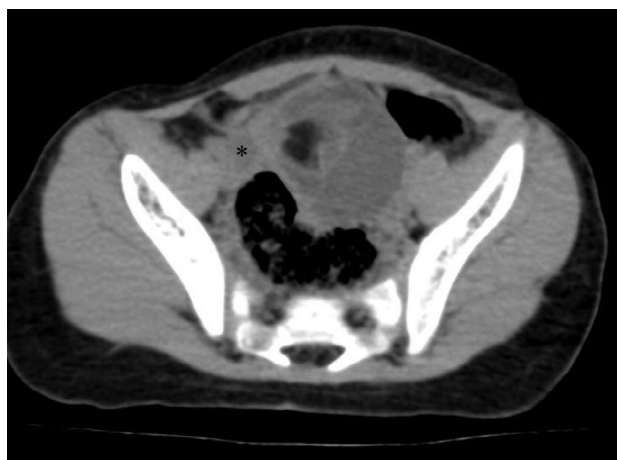


Figure 3. Left ovary with unusual topography with complex and grossly oval mass, of heterogeneous density, stressing ovoid and more hypoattenuating formations suggesting cystic nature and separated by non-uniform septations with soft tissue density. Right ovary with usual topography, regular contours and anatomical dimensions.



Figure 4. Most caudal region of the mass shows oval image of precise boundaries, lobulate contours and fat density with radiodense tooth image and poorly formed in its intimacy

The patient progressed without postoperative complications and was discharged after 5 days of hospitalization in good general condition with a scheduled return 10 days after the date of hospital discharge. Figure 8 shows the macroscopic aspects of the piece after surgical removal.

After diagnostic confirmation of mature ovarian teratoma by anatomopathological, without any evidence of malignancy and signs of immaturity, the patient is still under routine outpatient follow-up, asymptomatic.

DISCUSSION

The patient was diagnosed with mature ovarian teratoma, unilateral in the left ovary, exercising local expansive behavior causing deviation and compression

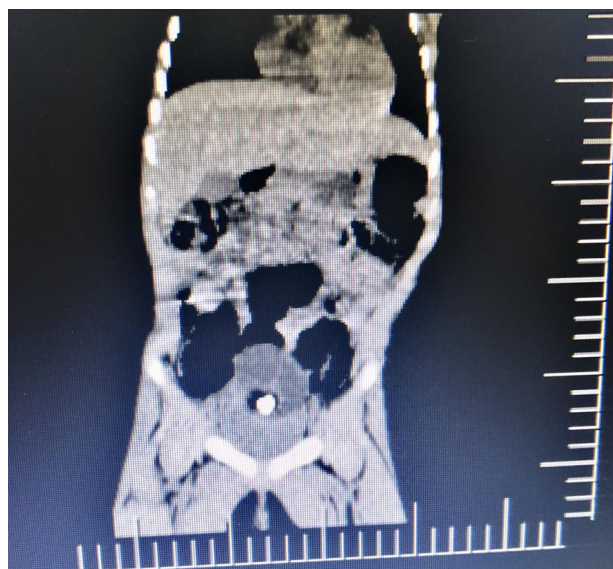


Figure 5. Bladder with good water repletion and regular and normothick walls, more posttherto-supero-lateral left part somewhat compressed by heterogeneous pelvic mass. Left ovary was not visualized with usual topography, and a complex and grossly oval mass was observed.



Figure 6. Eutopic kidneys, of usual shape and dimensions, presenting regular contours and normodense parenchyma, without evidence of hydronephrosis and images of calculations.

of surrounding anatomical structures which provoked symptoms of abdominal pain in severe colic and dysuria and poliuria, being initially confused with the diagnosis of recurrent urinary infections by similar clinical presentation. It is worth to mention that the patient was 2 years, 9 months and 22 days old at the time of presentation of the condition, an age which is not expected for the condition presented since this is a more common pathology in women of childbearing age³.

Leukocytes or piuria is characterized by an increased number of leukocytes and pyocytes in the urinary sample indicating, in most cases, inflammation of the urogenital tract. Despite presenting variations in reference values,

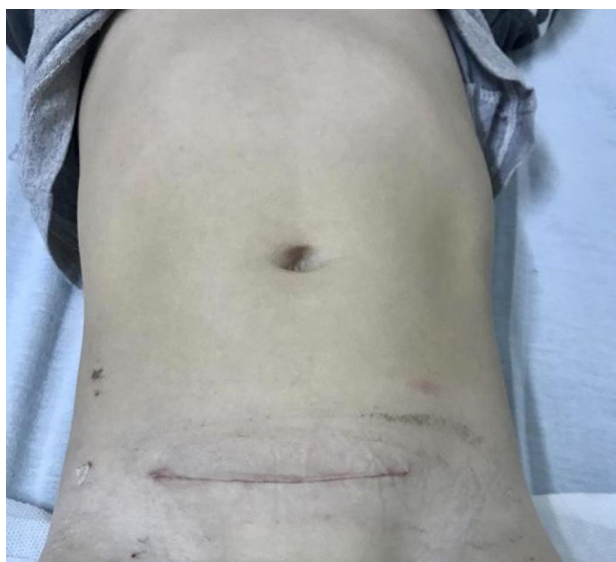


Figure 7. Pfannenstiel incision.



Figure 8. Macroscopic aspects of the piece after oophorectomy.

usually the presence of 5 leukocytes/field (400x) in the sample of medium jet urine, represents the upper limit of normality. Acute infection of the low or high urinary tract is the most frequent cause of leukocyturia however, other conditions can also cause this laboratorial change as any disease that is inducing inflammatory process in the urinary tract, such as tumors, urinary stones and foreign body^{5,6}.

In the laboratory tests of the patient in the case reported (Table 1) it is noticed that most of the samples have the finding of leukocyturia/piuria without the presence of bacterial growth in the Gram method and cultures characterizing then the finding of sterile piuria. Adding this finding to the elevated PCR test, in the absence of associated infectious condition, and the CT image mass in the left ovary exerting compressive process in the bladder it is concluded that piuria probably occurred due to the inflammatory process that the teratoma caused in the urinary tract structures, thus also responsible for the symptoms described by the patient,

who were confused with those of UTI by similar clinical presentation.

In the case of mature ovarian teratoma, ultrasound allows diagnosis in most cases, however, CT is essential to characterize large lesions and to exclude soft tissue components suggestive of malignancy. Visualization of fat-density tissue in a cystic adnexal lesion is diagnosed as ovarian teratoma³.

Although mature ovarian teratomas have a benign presentation, 1% of these ones have malignant transformation, thus justifying surgical intervention when it is detected. Furthermore, the assessment of the presence of symptoms, risk factors and desire for fertility are also decisive in the choice of the medical approach in face to the scenario. It rarely presents complication, though its occurrence is considered a surgical emergency⁷. The torsion or rupture of the cyst may evolve with a picture of chemical peritonitis. In addition, later on, mass content may drain into adjacent structures such as the bladder, intestine and vagina, thus leading to the formation of fistulas^{8,9}.

In unilateral tumors, annexectomy of the affected side is recommended, however, in pediatric patients or women of reproductive age, the preservation of the remaining normal ovarian tissue should be sought⁷. In cases of normal contralateral ovary on imaging exams, biopsy is not recommended, as this procedure is contraindicated to avoid adhesions or the formation of inclusion cysts that may interfere with the patient's fertility^{9,10}.

Tumorectomy is the surgical approach recommended for young women aiming to preserve the ovarian parenchyma and, consequently, hormonal actions and fertility since, the conservation of the remaining cortical tissue, however scarce, has follicles capable of maintaining these functions preserved. Although unilateral annexectomy may be preferred over tumorectomy or unilateral cystectomy in cases where it is not possible to individualize affected person's normal ovarian parenchyma, mainly in the case of large tumors¹¹.

In the face of recurrent UTI in pediatric patients, the Scientific Department of Nephrology of the Brazilian Society of Pediatrics (SBP), it is recommended the performance of ultrasound from the urinary tract and bladder in all patients who presented UTI in order to confirm/detect urinary tract malformations¹². USG is recommended as a screening method because it is a non-invasive procedure, does not expose patients to radiation and is comprehensive, allowing the detection of other pathologies¹⁰.

Based on the data presented, the importance of early abdominal USG is highlighted in the screening of different pathologies since a child complaining of abdominal pain needs to be evaluated by the paediatric specialist, who after anamnesis and complete physical examination will indicate the most appropriate propaedeutics. In the case described here, ultrasound was initially followed by tomography.

As neoplasms in the pediatric population have a different distribution from adults, radiologists need to be familiar

with ultrasound, tomographic and magnetic resonance imaging findings variable from ovarian neoplasms since imaging findings play an important role in the investigation of these conditions¹³.

Ovarian neoplasms are a rare condition in childhood and most often do not trigger clinical manifestations however, in some cases may present with abdominal pain, bloating, or palpable mass. Small ovarian physiological cysts are common findings in newborns, prepubescent and adolescents who, clinically, may be difficult to distinguish from ovarian tumors. Despite mature ovarian teratoma is not a common pathology in pediatric patients, the performance of an adequate investigation in cases of severe abdominal pain is extremely important for making an early diagnosis since, although they have mostly benign presentation, surgical treatment includes complete excision of the tumor, in order to preserve the fertility of patients whenever possible, is essential to avoid late complications that may bring risks to the patient and thus provide a better prognosis in these cases^{13,14}.

AUTHOR'S CONTRIBUTION

We describe contributions to the papers using the taxonomy (CRediT) provide above: Conceptualization, Investigation, Methodology, Visualization & Writing – review & editing: GRIBEL, Camile R.N.; GRIBEL, Navarro S.. Project administration, Supervision & Writing – original draft: GRIBEL, Camile R.N.; GRIBEL, Navarro S. Validation & Software: GRIBEL, Camile R.N. Resources & Funding acquisition: GRIBEL, Navarro S. Data curation & Formal Analysis: GRIBEL, Camile R.N.

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