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Case report

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PRIMARY RETROPERITONEAL TUMOR DURING PREGNANCY: CASE REPORT AND REVIEW OF THE LITERATURE

Tumor retroperitoneal primario durante el embarazo: reporte de un caso y revisión de la literatura

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ABSTRACT

Objective: To report the case of a pregnant patient diagnosed with a primary retroperitoneal tumor and to conduct a review of the literature pertaining to the diagnosis, treatment and maternal-fetal prognosis of this disease condition during pregnancy.

Materials and methods: A 19-year-old patient who presented with a retroperitoneal tumor identified on ultrasound. The results of the percutaneous biopsy showed a benign tumor. A healthy neonate was delivered by cesarean section. Surgical resection was performed four months later, and histopathology showed a mature cystic teratoma. A search was conduced in the Medline via PubMed, Lilacs, SciELO and ScienceDirect databases using the terms "pregnancy," "neoplasms," and "retroperitoneal neoplasms," including case reports and case series of retroperitoneal tumors during pregnancy. Diagnosis, treatment and maternal-fetal prognosis were identified.

Results: Overall, 1658 titles were identified. Of these, 34 case reports and 1 case series met the inclusion criteria. Of the cases, 62.9 % were benign. Diagnosis was made as an incidental finding in 63 % of cases, and 77 % of the masses were identified on ultrasound. Percutaneous biopsy was used in 8 % of cases, including the case reported here. Surgical treatment was used in 88 % of cases usually after delivery. Maternal mortality occurred in 8.5 % of cases. Fetal prognosis was good in 65 % of the pregnancies.

Conclusion: Despite being frequently benign, retroperitoneal tumors during gestation have a reserved maternal and fetal prognosis in a substantial number of cases. There is a need to assess the risks and benefits of percutaneous biopsy.

Key words: Pregnancy; neoplasms; retroperitoneal neoplasms.

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RESUMEN

Objetivo: reportar el caso de una paciente gestante con diagnóstico de tumor retroperitoneal primario y hacer revisión de la literatura del diagnóstico, tratamiento y pronóstico materno-fetal de esta entidad durante el embarazo.

Materiales y métodos: gestante de 19 años que consulta por tumor retroperitoneal identificado por ecografía, se realizó biopsia percutánea con resultado de tumor benigno; parto por cesárea con recién nacido sano. A los 4 meses se realizó resección quirúrgica, la histopatología mostró un teratoma quístico maduro. Se realizó búsqueda en las bases de datos de: Medline vía PubMed, Lilacs, SciELO y ScienceDirect, con los términos: "embarazo", "neoplasias" y "neoplasias retroperitoneales", incluyendo reportes y series de caso de tumores retroperitoneales en el embarazo. Se identificó el diagnóstico, tratamiento y pronóstico materno-fetal

Resultados: se identificaron 1.658 títulos, de los cuales 34 reportes de casos y una serie de casos cumplieron con los criterios de inclusión. El 62,9 % eran tumores benignos. El diagnóstico se hizo como hallazgo incidental en el 63 % de los casos, el ultrasonido identificó la masa en el 77 %, la biopsia percutánea se utilizó en el 8 % de los casos incluyendo el reportado. El tratamiento quirúrgico fue utilizado en el 88 % de los casos, generalmente después del parto. Hubo mortalidad materna en el 8,5 % de los casos. El pronóstico fetal fue bueno en el 65 % de las gestaciones.

Conclusión: los tumores retroperitoneales en la gestación, a pesar de ser principalmente benignos, tienen un pronóstico materno y fetal reservado en un importante número de casos. Se requiere evaluar los beneficios y riesgo de la biopsia percutánea.

Palabras clave: embarazo; neoplasias; neoplasias retroperitoneales.

INTRODUCTION

Primary retroperitoneal tumors are neoplasms that develop in the retroperitoneal space, arising from non-parenchymal structures such as adipose, muscle, connective, lymphatic, nerve and urogenital tissue, with wide histological variation (1) and their biological behavior is either bening or malignant they are divided into malignant or benign (2). They are extremely rare and close to 80% are malignant, the most frequent being sarcomas such as liposarcomas in 45% of cases (3). Benign tumors account for the remaining 20%, the most frequent being lipomas, fibromas and neurogenic tumors (4). Retroperitoneal tumors account for less than 0.2% of all malignant neoplasms (5).

They are usually diagnosed late and produce symptoms or become palpable once they are of significant size (2). Diagnosis is made by means of computed tomography (CT) or magnetic resonance imaging (MRI), as they provide information about their retroperitoneal location, size, relationship to adjacent organs and the presence or absence of metastasis (4). Percutaneous biopsy is performed in patients with tumors of uncertain radiological appearance or suggestive of a pathology in which neoadjuvant treatment may be appropriate as induction therapy (2). Treatment is based on surgical resection with curative potential and the likelihood of complete resection depends on tumor biology and invasion of adjacent organs and vascular structures. The specimen confirms the definitive histopathological diagnosis (2).

The frequency of retroperitoneal tumors during pregnancy is unknown; they are frequently mistaken for ovarian masses, renal cysts, adrenal tumors and lymphomas (6). Sarcomas are the most frequent tumors, and pregnancy does not affect prognosis or overall survival, although these tumors can grow at a very fast rate during gestation (7). There are case reports describing their association with maternal mortality (7,8). Special conditions in pregnancy include retroperitoneal hemangioma with potentially significant expansion probably due to vasoactive factors secreted during gestation resulting in peripheral vasodilation (9); and angiolipoma, also with significant growth, indicating hormonal dependence due to the presence

of estrogen and progesterone receptors in smooth muscle cells (10).

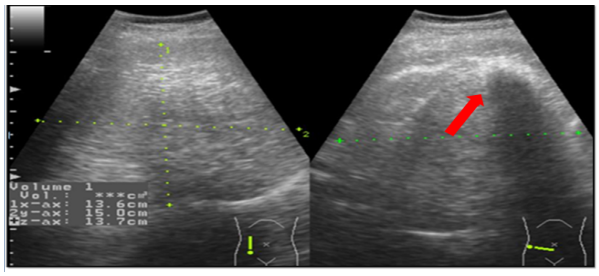
The infrequent occurrence of retroperitoneal tumors in pregnancy poses a diagnostic and therapeutic challenge for the obstetrician, the general surgeon and the oncologist (11), given the need to take into account both the mother and the fetus when considering treatment (12); compounded by the paucity of published information regarding fetal prognosis.

The objective of this study is to report the case of a patient with 19 weeks of pregnancy diagnosed with a primary retroperitoneal tumor managed with clinical monitoring and delayed surgical treatment, with adequate maternal-fetal outcome, and to conduct a review of the literature pertaining to the diagnosis, treatment and prognosis of this entity during pregnancy.

CASE REPORT

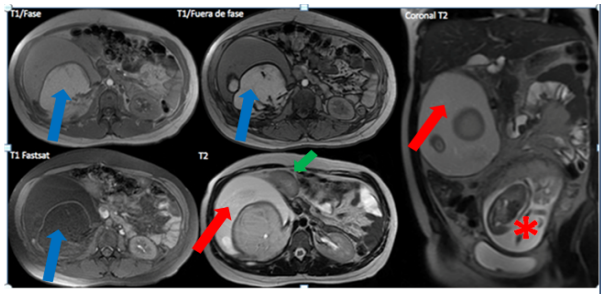
A 19-year-old patient with 19 weeks of gestation was seen in February 2017 at the emergency department of *Clinica Universitaria Colombia*, a high complexity center located in Bogota which serves patients under the contributive scheme of the Colombian General Social Security System. The main complaint was a first episode of heavy epigastric pain, intensity 4/10, lasting 10 days, accompanied by gastric content emesis. The patient had no pathological, pharmacological or surgical past history. Gynecological history included menarche at 15 years of age, regular menstrual periods, primigravida with desired pregnancy and three prenatal visits. On physical examination, blood pressure was 110/60 mmHG, heart rate 80 beats per minute (bpm), respiratory rate 16 breaths per minute, Glasgow scale 15/15 and temperature 36 degrees centigrade (°C); dry oral mucosa, epigastric and right upper quadrant tenderness with no peritoneal irritation; uterine height of 18 centimeters, longitudinal singleton fetus and fetal heart rate of 148 bpm; no vaginal discharge was observed on exploration, so no gynecological exam was performed. The patient was admitted for work-up for abdominal pain, dehydration and suspected cholelithiasis and in order to initiate parenteral crystalloids and conduct additional tests. Laboratory tests showed complete blood count without leukocytosis and normal leukocyte differential; normal liver and renal function tests and normal urinalysis. Obstetric ultrasound showed biometry for 19 weeks, estimated fetal weight of 264 g, in the 52nd percentile, normally attached placenta, abdominal fluid index of 13.5 cm. Abdominal ultrasound was used to rule out cholelithiasis, ruling out biliary disease and showing evidence of a right retroperitoneal solid tumor of regular contours 15 x 13 x 13 cm in size, with heterogeneous echogenicity probably due to fat component, and echogenic material projecting acoustic shadowing probably due to bone component (Figure 1). Magnetic resonance imaging was also performed showing an encapsulated retroperitoneal tumor 20 x 13 x 15 cm in size, with well-defined edges, hyperintense areas in T1, hypointense in FatSat sequences due to fat content, other hypointense areas in T1 but hyperintense in T2 due to cystic component of the tumor and multiple markedly hypointense areas in the adipose tissue, consistent with calcified-type tissue. The right adrenal gland was found to exert a compressive effect, displacing bowel loops and kidney but without invasion invasion of those structures. The potential diagnoses considered were malignant teratoma versus liposarcoma (Figure 2). Given the diagnosis of retroperitoneal tumor of unknown nature, cancer antigen 125 (CA-125) was requested, showing a result of 15.7 U/mL with a reference value lower than 35 U/mL. Ultrasound-guided percutaneous tru-cut biopsy was performed by the interventional radiologist retrieving two cylinders from the solid component of the tumor, without complications. On the second day of hospitalization, the patient reported improvement with resolution of the abdominal pain managed with parenteral metamizol and was discharged with tolerance of oral intake.

Figure 1. Ultrasound of retroperitoneal tumor in a pregnant woman seen at *Clinica Universitaria Colombia*. Bogota (Colombia)



Retroperitoneal mass 13.6 x 15 x 13.7 cm in size. The red arrow points an echogenic area, most probably bone component.

Figure 2. Magnetic resonance imaging of retroperitoneal tumor in a pregnant woman seen at *Clinica Universitaria Colombia.* Bogota (Colombia)

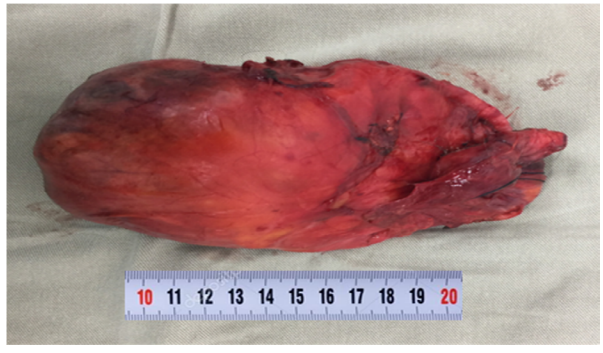


Retroperitoneal mass 20 x 13 x 15 cm in size. The red arrow points to cystic component, the blue arrow points to fat content and the green arrow points to displaced right kidney due to compression effect without tumor invasion. Fetus (*).

Two weeks later, the patient attended a multidisciplinary meeting with general surgery, gynecological oncology, oncology and maternal-fetal medicine. Having reviewed the histopathology report (hematoxylin-eosin staining) which described tissue consisting of keratinizing squamous epithelium, sebaceous glands, hair follicles and smooth muscle without immature tissue, consistent with mature teratoma, the team decided to follow the case clinically and to delay surgery after delivery at term because we found no indication for surgery. The patient remained on monthly outpatient prenatal follow-up by maternal-fetal medicine with monthly fetal ultrasound scans and had no complications. Cesarean section was performed at 39 weeks due to cephalopelvic disproportion and resulted in the delivery of a healthy neonate with a birthweight of 2,720 g, 48 cm in length and Apgar scores of 8, 9, 9 at 1, 5 and 10 minutes, respectively. To avoid increasing associated surgical risks, the retroperitoneal tumor was not resected at that stage. The postoperative period proceeded uneventfully, with uncomplicated puerperium.

Four months later, surgical resection was performed by laparotomy with the finding of a retroperitoneal tumor 25 cm in diameter, adherent to the caudate lobe of the liver and the diaphragm, with heterogenous fat, cystic and hair content, causing renal, right colon and caval displacement (Figure 3). Diaphragmatic violation occurred during tumor removal, requiring repair and tube thoracostomy. Estimated intraoperative blood loss was 1000 cc requiring transfusion of two units of packed red blood cells during the procedure. During observation in the inpatient unit, hemoglobin was 13.1 g/dL, requiring no additional blood product transfusions. The thoracostomy tube was removed on day 6 after adequate uncomplicated course and the patient was discharged on day 7. The definitive histopathological study (hematoxylin-eosin staining) showed a cystic lesion with mature teratomatous elements including epidermis, skin, bone and adipose tissue and

Figure 3. Retroperitoneal tumor specimen diagnosed during pregnancy at *Clinica Universitaria Colombia. Bogota (Colombia)*



no immature elements, with no tumor involvement of the margins and definitive diagnosis of mature cystic teratoma. The patient remains asymptomatic 24 months later.

MATERIALS AND METHODS

Based on the question "What is the diagnosis, treatment and maternal-fetal prognosis of retroperitoneal tumors in pregnancy?" a search of the literature was conducted in the Medline via Pubmen, Lilacs, SciELO and ScienceDirect electronic databases, using the terms "pregnancy," "neoplasm" and "retroperitoneal neoplasms." Case reports, case series and reviews of the literature in English and Spanish published between January 1998 and January 2019 describing diagnosis, treatment and maternal-fetal prognosis of retroperitoneal tumors during pregnancy were included; cases diagnosed 12 weeks after delivery were excluded. Two authors working independently selected articles by title and full texts were searched for the articles that met the population and design criteria. Compliance with inclusion criteria was verified and in cases of discrepancy, a third author made the decision of inclusion after analyzing the full text. The assessed variables were: age, nature of the tumor, symptoms, tumor markers used, diagnostic tests, non-surgical treatment, surgical treatment, timing of surgery, adjuvant therapy, fetal outcome, route of delivery, and antenatal, perinatal and postpartum maternal complications.

Ethical considerations. A written informed consent from the patient was obtained and authorization to publish the case was requested from Clinica Universitaria Colombia and Fundación Universitaria Sanitas through the Health Science Research Center. The necessary steps were taken to ensure information confidentiality and patient anonymity. The photographs were taken by the authors.

RESULTS

Overall, 1,658 titles were found of which 35 articles on pregnancy-associated retroperitoneal tumors that met the search criteria in the full text were included: 34 case reports (1,6-38) and one case series of primary mucinous retroperitoneal tumors which included 1 case during pregnancy (39) (Table 1). Of the 35 reported cases, 1 was a patient under 20 years of age (14), 13 patients were aged 20 to 29 years of age (1,6,13,20-22,25-28,30, 33, 37), 20 cases were patients between 30 and 39 years of age (7-12,15-19,23,24,29,31,32,34-36,38); there was no age reported in 1 case (39). According to their biologic behavior during pregnancy, 62.9% (22 cases) of retroperitoneal tumors were benign (1,6,9-11,14,20,22-24,27-32,34-39) and 37.1% (13 cases) were malignant (7,8,12,13,15-19,21,25,26,33).

Diagnosis: Diagnosis was made before pregnancy in three cases (7,9,36), in the first trimester in 6 cases (13,21,25,26,30,33), in the third trimester in 12 cases (1,6,8,12,15,17-19,22,24,28,37) and during puerperium in one case (10); there was no report of the time of diagnosis in two cases (32,34). Retroperitoneal tumors grow to a significant size before being diagnosed or producing symptoms (11). During pregnancy diagnosis is challenging due to abdominal distension and the fact that symptoms are attributed to gestation and, for this reason, early detection is infrequent (17,18). It was found that 22 cases were asymptomatic (1,9,12-14,16,18-20,22-26,28, 29,31-33,35,37,38), 6 presented with abdominal pain (6,10,11,21,30,39), 3 had abdominal mass sensation (15,17,27), and 1 patient complained of weight loss (8).

Tumor markers were requested in 13 cases (1, 6, 12-14,16,18,19,23-25,31,33) and, of those, five were normal (6,13,16,23,33), CA19-9 was elevated in three cases (12,19,24), alpha-fetoprotein was elevated in three cases (1,14,18) and CA125 was elevated in two cases (25,31). Some authors argue that these markers are not good for differentiating the nature of retroperitoneal tumors because values may be altered due to pregnancy and they are non-specific (14,19,20).

Ultrasound was carried out in 27 cases (1,6,7,10-14,16,19-21,23-33,35-38) with findings that

Table 1.
Included studies of case reports or case series of pregnant women with retroperitoneal tumors, 2000-2020

Author, year, country	Gestational age at diagnosis and parity	Symptoms	Diagnosis and testing during pregnancy	Type of delivery	Treatment	Recurrence or mortality	Pathology diagnosis
Subramony 2001 USA (37)	28 weeks, G3P2C1	Asymptomatic	Ultrasound	Term cesarean section	Surgery after cesarean surgery	N/R	Mucinous cystoadenoma
Molina 2001 Spain (30)	17 weeks, G1	Abdominal pain, fever	Ultrasound	Miscarriage, 17 weeks	Surgery after D&C	No	Angiomyolipoma
Sivarajan 2004 United Kingdom (21)	18 weeks, G3	Abdominal pain	Ultrasound, MRI, percutaneous biopsy	Cesarean section, 27 weeks IUGR	Non-resectable chemotherapy (cyclophosphamide), doxorubicin, vincristine, then cyclophosphamide replaced with ifosfamide, radiotherapy*	N/R	Primitive neuroectodermal tumor
Sonntag 2005 Germany (12)	30 weeks, G1	Asymptomatic	Ultrasound, MRI	Cesarean section, 38 weeks healthy	Surgery with Cesarean section	No	Mucinous adenocarcinoma
Jeng 2005 Taiwan (7)	12 weeks, G2P0	History of liposarcoma	Ultrasound	Cesarean section, 36 weeks	Surgery with Cesarean section, radiotherapy*	Yes, death at 8 months	Mixoid liposarcoma
Talwar 2005, India (6)	32 weeks, G2P1	Abdominal pain,	Ultrasound	Delivery, 32 weeks	Surgery after delivery	No	Mature cystic teratoma with abscess
Ibraheim 2005, United Kingdom (29)	12 weeks	Asymptomatic	Ultrasound	Cesarean section 38 weeks, healthy	Surgery after cesarean section	N/R	Schwannoma
Par veen 2007 United Kingdom (23)	13 weeks, G1	Asymptomatic	Ultrasound	Cesarean section, 39 weeks healthy	Surgery after cesarean section	N/R	Schwannoma

Continued Table 1

Author, year, country	Gestational age at diagnosis and parity	Symptoms	Diagnosis and testing during pregnancy	Type of delivery	Treatment	Recurrence or mortality	Pathology diagnosis
Rouskova 2007 Czech Republic (8)	34 weeks	Weight **loss**	N/R	Induced delivery	Surgery after delivery, chemotherapy	Yes, death after one month	Pleomorphic liposarcoma
Kashima 2008 Japan (33)	26 weeks, G2P2	Asymptomatic	Ultrasound, MRI	Parto, 38 weeks healthy	Surgery during gestation (31 weeks)	No	Mucinous cuystoadenoma
Yadav 2008 United Kingdom (32)	N/R, G3C2	Asymptomatic	Ultrasound, MRI	VTOP	Conservative management	Not applicable	Schwannoma
Ulker 2008 Turkey (13)	17 weeks, G2P1	Asymptomatic	Ultrasound, MRI	Delivery, 38 weeks healthy	Surgery during gestation (17 weeks)	No	Leiomyosarcoma
Haakova 2009, Czech Republic (35)	I trimester, G1	Asymptomatic	Ultrasound, MRI	Cesarean section, 37 weeks healthy	Surgery after cesarean section	N/R	Schwannoma
Lopes 2009 Brazil (16)	13 weeks, G2P1	Asymptomatic	Ultrasound, CT scan	Cesarean section, 37 weeks healthy	Surgery during gestation (13 weeks)	No	Liposarcoma
Roma 2009 EE.UU. (39)	12 weeks	Pelvic pain	N/R	N/R	Surgery	N/R	Mucinous cuystoadenoma
Li 2010 China (34)	N/R	N/R	N/R	Delivery	Surgery after delivery	N/R	Mature cystic teratoma
Ramírez 2010 Spain (10)	Postpartum day 10	Abdominal pain	Ultrasound	Delivery	Surgery after delivery	No	Angiomyolipoma
Acín-Gándara 2010 Spain (31)	2 weeks	Asymptomatic	Ultrasound, CT scan, MRI, percutaneous biopsy	VTOP, 2 weeks	Surgery	N/R	Ganglioneuroma

Continued Table 1

Author, year, country	Gestational age at diagnosis and parity	Symptoms	Diagnosis and testing during pregnancy	Type of delivery	Treatment	Recurrence or mortality	Pathology diagnosis
Dueñas 2011 USA (15)	36 weeks, G2C2	Abdominal mass sensation	Physical exam, during Cesarean section biopsy	Cesarean section, 36 weeks	Surgery after cesarean section	N/R	Liposarcoma
Sousa 2012 Portugal (20)	13 weeks	Asymptomatic	Ultrasound, MRI	Cesarean section, 38 weeks healthy	Surgery during gestation (18 weeks)	N/R	Ganglioneuroma
Wei 2013 China (1)	35 weeks	Asymptomatic	Ultrasound	Cesarean section, 39 weeks healthy	Surgery with cesarean section	No	Lipoma
Becerril 2013 Mexico (14)	10 weeks, G1	Asymptomatic	Ultrasound	Miscarriage, 10.6 weeks	Surgery with D&C	No	Mature teratoma
Chen 2013 China (38)	8 weeks, G5P2A2	Asymptomatic	Ultrasound	Delivery, 39 weeks healthy	Surgery during gestation (14 weeks)	N/R	Mucinous cystadenoma
Oh 2014 Korea (18)	28 weeks, G6A5	Asymptomatic	Physical exam, MRI, CT scan	Cesarean section, 29 weeks	Surgery after cesarean section, chemotherapy (doxorubicin, ifosfamide), radiotherapy*	No	Liposarcoma
Hanhan 2014 Turkey (19)	29 weeks, G5P5	Asymptomatic	Ultrasound, MRI	Delivery, 38 weeks healthy	Surgery during gestation (30 weeks)	No	Mucinous cystadenoma
Tan 2014 Singapore (9)	Prenatal	Asymptomatic	MRI	Cesarean section, 39 weeks healthy	Conservative management	N/A	Hemangioma
Yu 2014 China (17)	40 weeks, G1P1	Abdominal mass sensation	Physical exam after delivery	Delivery, 40.6 weeks healthy	Surgery after delivery, chemotherapy	Yes, death "short period of time"	Spindle cell rhabdomyosarcoma
Huo 2015 China (26)	16 weeks	Asymptomatic	Ultrasound, MRI	Cesarean section, 37 weeks healthy	Surgery during gestation (20 weeks)	No	Mixoid liposarcoma

Continued Table 1

Author, year, country	Gestational age at diagnosis and parity	Symptoms	Diagnosis and testing during pregnancy	Type of delivery	Treatment	Recurrence or mortality	Pathology diagnosis
Berczi 2015 Canada (11)	13 weeks	Lumbar pain, fever	Ultrasound, MRI	Delivery, 39 weeks healthy	Surgery during gestation (14 weeks)	No	Mucinous cystic adenoma
Dayan 2016 Israel (24)	36 weeks, G3P3	Asymptomatic	Ultrasound	Unknown	Surgery after delivery	No	Mucinous cystadenoma
Goel 2017 India (27)	6 weeks	Absominal mass sensation	Ultrasound, CT scan	VTOP, 6 weeks	Surgery	No	Mature cystic teratoma
Nithya 2017 India (28)	34 weeks, G1	Asymptomatic	Ultrasound	Cesarean section, 34 weeks PROM	Surgery after cesarean section	No	Schwannoma
Greimel 2017 Austria (36)	I trimester, G2C1	History of schwannoma	Ultrasound	Cesarean section, term	Conservative management	N/A	Schwannoma
Wang 2018 China (22)	33 weeks	Asymptomatic	CT scan	Cesarean section, term	Surgery after cesarean section	No	Ganglioneuroma
Paul 2018 India (25)	24 weeks, G3A2	Asymptomatic	Ultrasound, CT scan	Delivery, 28 weeks	Surgery during gestation (24 weeks), radiotherapy*	N/R	Mixoid liposarcoma

IUGR: intrauterine growth restriction. VTPO: voluntary termination of pregnancy. MRI: magnetic resonance imaging. CT scan: computed tomography. PROM: premature rupture of membranes. N/R: not reported. *Radiotherapy after gestation.

included abdominal or retroperitoneal cystic or solid masses with sizes ranging between 5 cm (12) and 32 cm in diameter (19), that persisted in follow-up scans. CT was performed in six cases (16,18,22,25,27,31) and MRI in 13 (9,11-13,18-21,26,31-33,35); the latter has been described as the best non-invasive method for radiological imaging of retroperitoneal tumors during pregnancy as patients are not exposed to ionizing radiation (16). On the other hand, added to ultrasound, these studies offer better characterization of the lesion (location, morphology, tissue composition, plane delineation) and assessment of surgical resection options (1,14,20).

As for percutaneous biopsy of the retroperitoneal tumors, it was performed at two different times: at 2 weeks (31) and at 18 weeks, with no complications.

Treatment: Surgical resection was performed in 31 of the reported cases (1,6-8,10-20,22-31,33-35,37-39): 1 case in the first trimester (16), 6 cases in the second trimester (11,13,20,25,26,38), 2 in the third trimester (19,33), 3 cases at the time of the cesarean section (1,7,12), and 18 after delivery (6,8,10,14,15,17,18,22-24,27-31,34,35,37). The time of the surgical procedure was not reported in one case (39). No surgical treatment was performed in four cases (9,21,32,36): non-resectable tumor in one case (21) and three cases of benign tumors that continued under clinical observation (9,32,36). During pregnancy surgery can be performed through the open approach or through laparoscopy (11), although the traditional and most frequent approach is laparotomy (14,24) performed with the aim of achieving complete tumor resection.

In terms of adjuvant therapy, four cases of retroperitoneal liposarcomas were documented, two of which received radiotherapy (7,25), one received chemotherapy (8) and one received radiotherapy plus chemotherapy (18). Chemotherapy during pregnancy was used in one case of primitive neuroectodermal tumor followed by postparturm chemotherapy plus radiotherapy (21) and chemotherapy was used in one case of spindle cell rhabdomyosarcoma (17).

Prognosis: In retroperitoneal tumors during pregnancy, prognosis is determined by the nature of the tumor and its complete resection (13,16). Prenatal maternal complications included two cases of febrile syndrome which resolved after surgical resection of the retroperitoneal tumors (11,30) and one case of an abcessed retroperitoneal mature cystic teratoma that required surgical management during gestation (6). A case of mechanical dystocia was reported as maternal complication during delivery due to the size and location of the retroperitoneal tumor requiring cesarean section (28). As for postpartum complications, there were three cases of maternal mortality (7,8,17).

Related to fetal outcomes, pregnancy was terminated in three cases in order to treat the retroperitoneal tumor (27,31,32). 17 cesarean sections were performed (1,7,9,12,15,16,18,20-23,26,28,29,35-37), 11 were vaginal deliveries (6,7,10,11,13,17,19,25,33,34,38) and route of delivery was not reported in two clinical cases (24,39) for a total of 23 healthy neonates (1,7-13,15-17,19,20,22,23,26,29,33-38).

Regarding obstetric complications, there were two miscarriages (14,30), two spontaneous preterm deliveries (6,25), one case of premature rupture of membranes at 34 weeks (28), and one preterm delivery at 27 weeks with intrauterine growth restriction (21). There was another preterm delivery at 29 weeks due to suspected malignant retroperitoneal tumor with early termination of pregnancy after fetal lung maturation was achieved in order to perform the surgical removal of the tumor (18).

CONCLUSIONS

Primary retroperitoneal tumors during pregnancy are extremely rare and diagnosis is based on ultrasound as the first approach to retroperitoneal masses, complemented by MRI which is the preoperative radiological test of choice. Percutaneous biopsy appears to be safe during pregnancy as an adjunct to determine the nature of the tumor preoperatively since histopathology provides the definitive diagnosis.

There is no unified therapeutic approach during pregnancy although surgical tumor removal with an individualized plan for each patient is considered to improve maternal and perinatal outcomes as well as good maternal prognosis which is determined by the nature of the tumor and its complete resection.

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