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Case Report

Sacro-coccygeal teratoma: about an observation and diagnostic process

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Abstract

Sacro-coccygeal teratomas (SCT) are the most common benign fetal tumors with an incidence of about 1/3000 births. Currently, thanks to advances in imaging techniques, diagnosis can be made in the first trimester of pregnancy. We report the case of Mrs. JF, a 25-year-old primigravida female, with no significant pathological history, a primigravida who had consulted the radiology department of the maternity hospital of Kairouan, at 16 weeks of amenorrhea, to do a prenatal ultrasound. On examination, it was a progressive monofetal pregnancy of 16 weeks of amenorrhea, with discovery of a cystic formation about 3 cm in diameter, well limited, located in the sacro-coccygeal region. A complementary Magnetic Resonance Imaging (MRI) scan showed a well-defined mass measuring 24x17mm, which was hyper intense on T1, attached to the caudal end of the fetus, at the presacral space, with an extension downwards to the soft parts, with no endopelvic component or fat component. In the presence of these radiological data, a sacro-coccygeal teratoma was first suspected, but a meningocele could not be formally eliminated, due to the limits of these examinations. The collegial decision was therefore to authorize the continuum of the pregnancy. The patient was monitored on an outpatient basis with regular clinical and ultrasound check-ups. Pregnancy continued up to 40 weeks. The baby was operated on, at 2 weeks of age, with complete removal of the tumor. The pathological examination of the surgical specimen was in favor of a mature cystic sacrococcygeal benign teratoma, without signs of malignancy.

Keywords: Teratoma, sacrococcygeal region, prenatal diagnosis, ultrasound, MRI-differential diagnosis.

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1. Introduction

Sacro-coccygeal teratomas (TSC) are the most common benign fetal tumors with an incidence of about 1/3000 births [1]. They preferentially affect the female fetus. They develop at the expense of the totipotent embryonic cells of the sacrococcygeal region. Their excision must be performed during the first days of life in order to avoid degeneration [2]. Currently, thanks to advances in imaging techniques, diagnosis can be made in the first trimester of pregnancy [3]. The problem that arises during the prenatal detection of a pelvic mass is the question of the differential diagnosis between sacro-coccygeal teratoma and anterior pelvic myelomeningocele, which can involve both the vital and functional prognosis of the baby. Thus, its discovery in antenatal care requires a medical interruption of pregnancy. Herein, through this clinical observation, we investigated the radiological features of the sacrococcygeal teratoma, which characterize them from other pelvic masses and then we established a diagnostic approach.

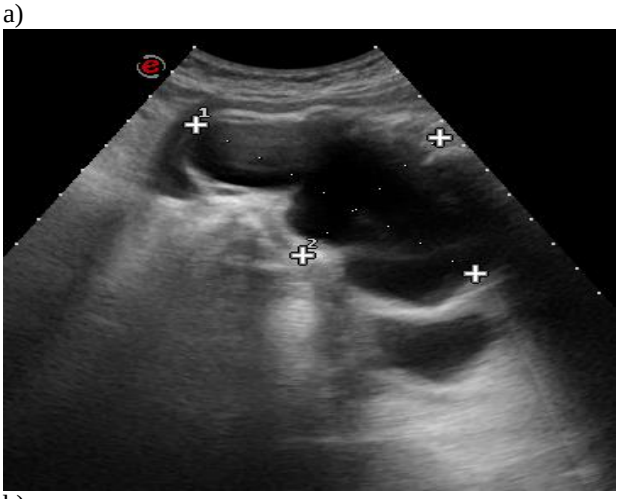
2. Case report

Mrs. J. F was a 25-year-old primigravida with no significant pathological history. She had consulted the Maternity Unit of the University Hospital of Kairouan, at 16 weeks of amenorrhea, to do a prenatal ultrasound. It was a progressive monofetal pregnancy of 16 weeks of amenorrhea, with discovery of a well-limited cystic formation about 3 cm in diameter, located in the sacro-coccygeal region (Fig.1a, b, Fig.2 a, b).

A complementary Magnetic Resonance Imaging (MRI) scan showed a well-defined mass measuring 24x17mm, which was hyper intense on T1, attached to the caudal end of the fetus, at the presacral region, with extension downwards to the soft parts, with no endopelvic component or fat component (Fig.3 a, b). In the presence of these radiological data, a sacrococcygeal teratoma was first suspected, but a meningocele could not be formally eliminated within the limits of these examinations. The collegial decision was therefore to authorize the continuum of the pregnancy. The patient was monitored on an outpatient basis with regular clinical and ultrasound check-ups. During the physical examination, the blood pressure was stable with negative proteinuria at each consultation. Besides, the uterine height was proportional to the gestational age. During the ultrasound monitoring: there was

an increase in the size of the mass arriving up to 9 cm at full term. In addition, there were no signs of hydrops foetalis or right heart failure. Pregnancy was continued up to 40 weeks.

a)



b)

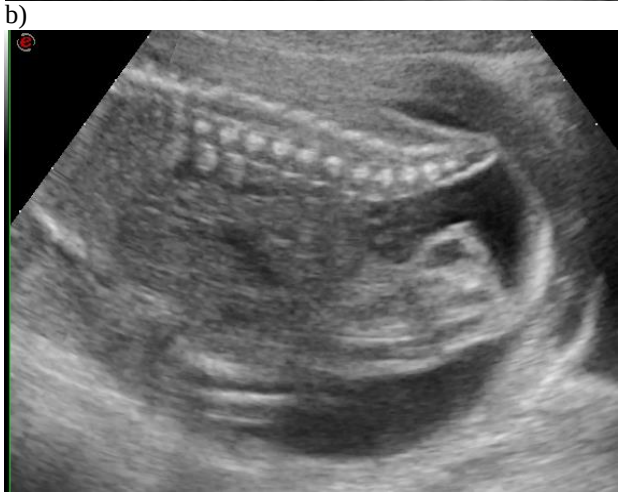
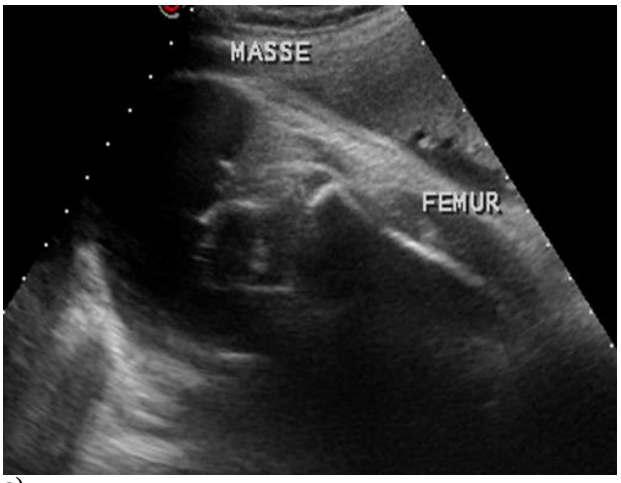


Fig. 1 a, b. Morphology ultrasound: Multilocular cystic mass about 3 cm in diameter, well limited, with respect to the sacrococcygeal region.

The delivery was scheduled by Caesarean section. On examination of the newborn: It was a boy weighing 3400g, with an Apgar score of 9/10. A tender oval mass of 12x9cm was found at the sacral region, with no skin abnormalities. At the rectal examination, we noticed that this formation compressed the rectum. Thus, the newborn was admitted to the neonatology ward. A soft tissue ultrasound was requested which showed a voluminous multilocular cystic mass with non-vascularized septa, located at presacral space, pushing the bladder up and forward, extending to the soft parts of the gluteal region mostly to the left. An abdominopelvic Computer Tomography (CT) scan showed an abdominopelvic mass of retro-rectal presacral fluid density with intrapelvic and exopelvic development measuring 80x46mm. This formation presented thin partitions that stood out, after injecting the contrast product. It does not have a fatty component but there are calcifications facing its lower pole. This mass pushes the bladder upwards and forwards without signs of invasion. It also represses the ureters laterally, with a slight bilateral uretero-pyelo-calyceal dilatation. The baby was operated on, at 2 weeks of age, with complete removal of the tumor. The pathological examination of the surgical specimen was in favor of a mature cystic sacrococcygeal benign teratoma, without signs of malignancy.



a)

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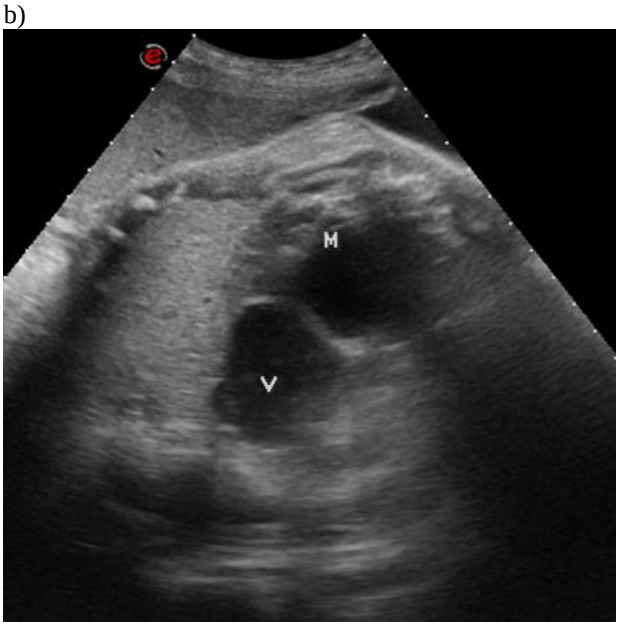


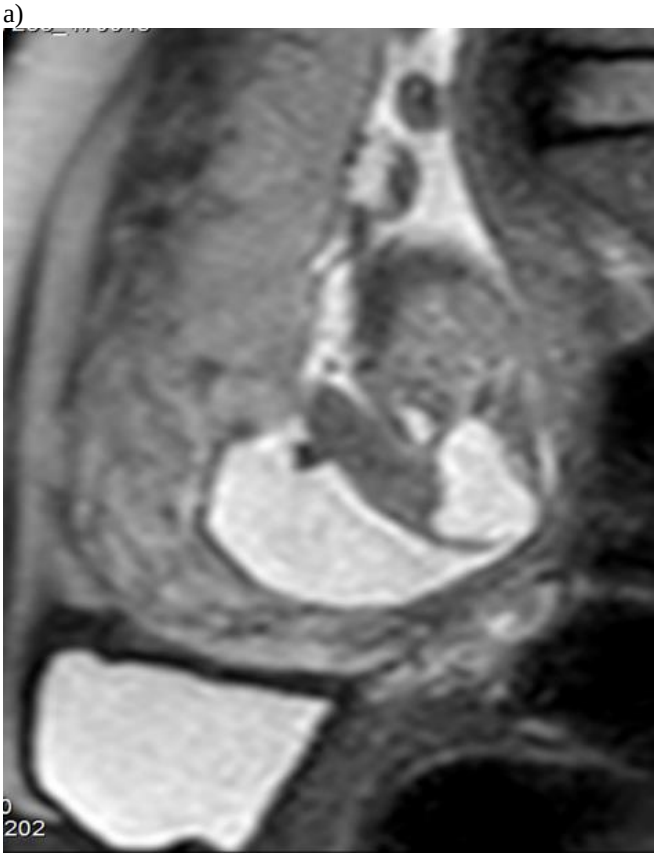
Fig. 2 a, b. mass extends towards the soft parts of the buttock with pelvic development.

4. Discussion

The sacro-coccygeal teratoma is a germinal congenital tumor that develops in the presacral region. It is a rare tumor with an overall incidence of 1/35000 to 1/40000 births [1]. It is the most frequent teratoma of the fetus, as it represents more than 50% of the teratomas of the newborn [2]. It is the second most common neonatal tumor after lymphangioma. Besides, it is the third largest retroperitoneal tumor in children after neuroblastoma and nephroblastoma [3]. The sacro-coccygeal teratoma is diagnosed more frequently in females, with a sex ratio of 4/1 [4]. In her study in Morocco in 2012, Nacireddine [5] found that 81.5% of newborns with sacro-coccygeal teratoma are female (30 of 35 cases). Similarly, in Niger, Sanoussi [6] found a female predominance with a frequency of 66.6%. There are no well-identified risk factors in the literature, but some authors suggest heredity and twin pregnancy. Since the emergence

of the ultrasound, the diagnosis would usually be made in the second trimester of pregnancy: between the 15 and the 32 weeks of amenorrhea.The second trimester of pregnancy:

a)



b)

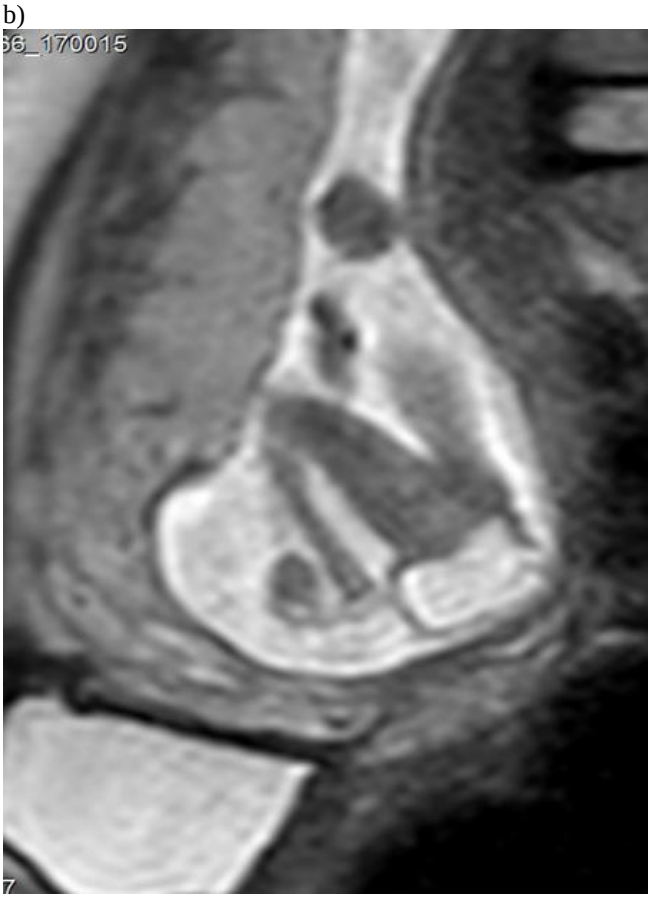


Fig. 3 a, b: T2-weighted sagittal MRI sections: T2 hyperintense cystic mass, well limited, attached to the caudal end of the fetus with extension to the soft parts.

between the 15 and the 32 weeks of amenorrhea. It would be either a fortuitous discovery, or more often during an assessment of an increase in uterine height. The mean age of an ultrasonographic discovery of a sacrococcygeal tumor, according to Makin [7] is 21 weeks of amenorrhea with extremes between 17 and 31 weeks of amenorrhea. According to Neubert [8], 23 weeks of amenorrhea is the average age of ultrasound discovery. In our observation, the mass was discovered at 16 weeks of amenorrhea. Macroscopically, the lesions may be cystic, solid, or mixed. Solid teratomas are the rarest, but they have a high potential for malignancy. However, cystic teratomas are generally benign and have a good prognosis. In the literature, the benign (or mature) form is the most frequent form. In fact, 75% of teratomas are mature according to Makin [7] and 79% according to Gabra [8]. It should be mentioned that most tumors that are benign at birth may become malignant after about 2 months, hence the value of early diagnosis and complete resection. In the case of an externalized tumor, the ultrasound would show a rounded multilocular mass, attached to the pelvic side of the fetus. The base would be located in the sacral or sacro-coccygeal region. It would usually be in the middle, thus pushing the anus forward, and sometimes it would be lateralized [9]. The walls are thin, clearly identifiable. The size of the mass varies from a few centimeters to more than 20 cm in diameter. The ultrasonographic structure of the tumefaction is variable. It is mixed in 75% of cases, with a solid echogenic component and fluid levels suggestive of cystic or hemorrhagic zones or necrosis. In 15% of cases, it could be liquid, transonic and thin-walled, with only a few solid elements found near the base.

The cyst would often be divided by some partitions. It is in this form that the differential diagnosis with spina bifida arises [10]. Sometimes, we can find highly echogenic areas, which, depending on the importance, can convey calcifications or even bone or osteocartilaginous structures. These calcifications, which are found in 35% to 45% of SCT, would be either small bone fragments or dystrophic calcifications in hemorrhagic or necrotic areas. The Doppler color mode would then look for tissue vascularization, arteriovenous fistula, and superficial hypervascularity [11]. Solid teratomas generally have a high malignancy potential. Ultrasound should always seek the existence of an endopelvic extension. Indeed, it would be necessary to analyze the position of each organ, its potential repression and a potential impact on structures above, such as bladder dilatation or ureterohydronephrosis [12]. The diagnosis of the non-externalized form with a unique intra-pelvic development is difficult. It depends on a perfect knowledge of the ultrasonographic anatomy of the normal fetal pelvis. It is extremely rare, since it only represents 10% of the sacro-coccygeal teratomas. However, its risk of degeneration is high: 2.4% at birth, 10% when diagnosed at 2 months and 60% to 4 months. Therefore, a Magnetic Resonance Imaging (MRI) scan is required at the smallest doubt of the diagnosis.

When such tumor is discovered, the search for associated malformations must be systematic in order to establish the post-natal prognosis. According to Altman [13], they are

found in 18% of cases with: skeletal muscular malformations (6%), malformations affecting the central nervous system (4%), malformations of the urinary tract (5%), heart defects (2%) and intestinal malformations (2%). The MRI aspect of the Sacro-coccygeal teratomas is typical; it is generally a large mass containing round well-defined areas, with intermediate signal intensity representing its solid, cystic or calcified components.

In conclusion, the antenatal diagnosis of sacro-coccygeal teratomas requires additional attention to be diagnosed even with advances in imaging techniques, regular ultrasound monitoring, probably weekly or bi-weekly in order to assess the evolution of the tumor and its potential impact on the fetus.

Consent of patient

Written informed consent was obtained from the patient for participation in this study.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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Conflict of Interest Disclosures

All authors declare that they have no conflict of interest.

Authors' contributions

Conception and design of the study: Marouen N, Chelly S, Soui S, Fatnassi R. Acquisition of data: Marouen N, Chelly S, Soui S. Analysis and/or interpretation of data: Marouen N and Chelly S. Drafting the manuscript: Marouen N, Bannour R, Haddad N, Merzougui L. Reviewing the manuscript: Bannour R, Haddad N, Merzougui L.

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