Journal of Surgery
2019; 7(6): 202-206
http://www.sciencepublishinggroup.com/j/js
doi: 10.11648/j.js.20190706.20
ISSN: 2330-0914 (Print); ISSN: 2330-0930 (Online)

Case Report

Sacro-coccygeal Teratoma in a Child of 6 Months: A Case Report

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To cite this article:

Mfouou Azoo Frederic, Engbang Ndamba Jean Paul, Essola Basile, Chichom-Mefire Alain, Ngowe Ngowe Marcelin. Sacro-coccygeal Teratoma in a Child of 6 Months: A Case Report. *Journal of Surgery.* Vol. 7, No. 6, 2019, pp. 202-206. doi: 10.11648/j.js.20190706.20

Received: November 15, 2019; **Accepted**: December 20, 2019; **Published**: December 31, 2019

Abstract: Sacro-coccygeal teratoma (SCT) is a type of tumor which mostly occurs in newborn babies. The vast majority of this type of tumor is benign. That pathology can be diagnosed antenatally or at birth. Teratomas, especially those diagnosed in the neonatal period, occur most frequently in the sacrococcygeal region. Clinically in many cases, we can observe a large swelling (mass) in the sacrococcygeal region. In some cases, there are signs of compression following by the displacement of the rectum and pelvic organs. Alphafetoprotein is the main biological test used. In association morphologically by echography, CT-scan, MRI and others, the tumor is classified. The classification used is the topographical one of TSCs, which was proposed by Altman to the American Academy of Pediatric Surgery. The mainstay of therapy is the complete surgical removal. We present a case of a newborn (6 months), female, with teratoma sacro-coccygeal. At birth the baby was noticed to have large swelling (mass), without any sign of compression of others organs. Clinical and paraclinical signs permitted us to conclude of a SCT type II of Altman's classification. A surgery was performed with a sacral approach. Histologically, tumors were classified into good (mature) category.

Keywords: Sacro-coccygeal Teratoma, Child, Mature

1. Introduction

Sacro-coccygeal teratomas (SCT) are malformations multi-tissue tumors composed in proportion tissue variable from the three embryonic leaves (endodermic, mesodermal and ectodermal) [1]. That affection rate of about 1 / 35000-40000 live births, occurring 35-60% of these types of teratomas, tumors found in girl than in boy [2]. It can be diagnosed antenatally or at birth with a prevalence of 3 to 4 times in female children compared to males [3]. Benign SCT has an excellent outcome after early surgery, but the incidences of malignancy increase if resection is delayed [4]. The mainstay of therapy is the complete surgical removal and postoperative chemotherapy if the histopathology report suggests malignant variety or a high grade immature teratoma associated with high levels of AFP (alphafetoprotein) [5]. We are reporting a case of b Saco-Coccygeal teratoma in a child of 6 months. The informed consent from parents for this study was obtained.

2. Case Presentation

We report a newborn case (6 months), female, with teratoma sacro-coccygeal. The mother is a thirth gesture, thirth part with antecedent of one caesarean. That female baby

of 2.8 kg was born through normal vaginal delivery at 38 weeks of gestation, with normal Apgar score. At birth the baby was noticed to have large swelling (mass) in the sacrococcygeal region, with solid consistency which was approximately 35×30 cm (Figures 1 and 2) in size. Urinate, defecation were normal. The investigations included CT scan of the abdomen and swelling, blood examination for tumor markers (AFP). These permits us to conclude of a SCT type II of Altman's classification (Figure 3).

A surgery was performed with a sacral approach. An inverted V skin incision was performed. The tumor was dissected at it lateral parts by digital dissection. We made the dissection of the coccyx by placing the finger into the rectum, to avoid lesion of the rectum. The total resection of the tumor was done (Figures 4, 5).

On microscopic examination, tumors were classified into good (mature) category. The predominant component was ectodermal tissue.

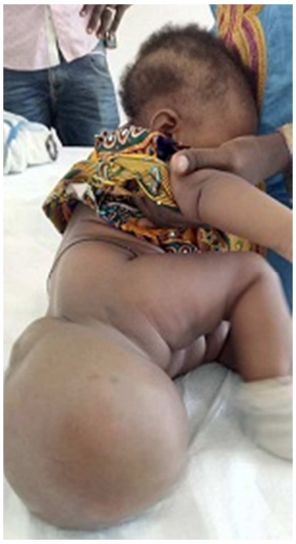

Figure 1. *Large swelling in Sacrococcygeal region.*

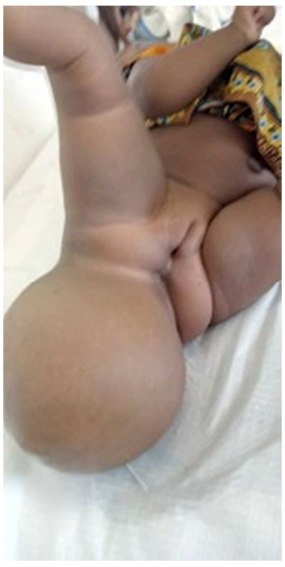

Figure 2. *Female baby.*

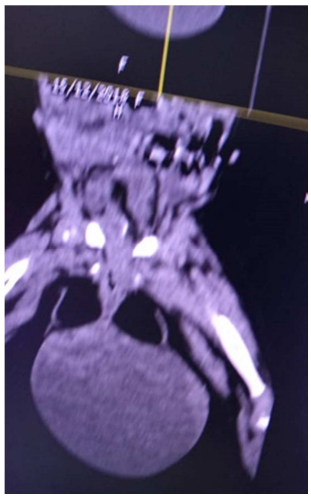

Figure 3. *Sacrococcygeal teratoma with mainly external development.*

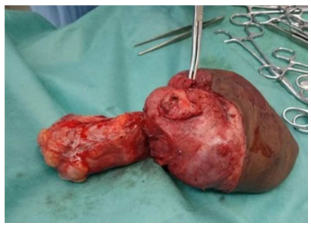

Figure 4. *Mass of 2.5 kg with its pelvic extension.*

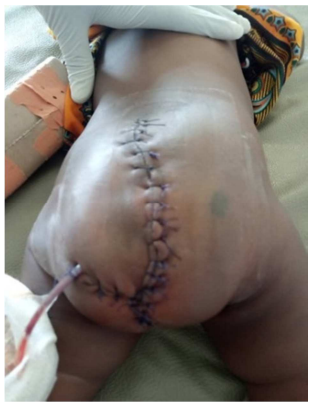

Figure 5. *Postoperative aspect.*

3. Discussion

Teratoma is the most common tumor in the neonatal period. It is a germinal tumor containing tissues from two or three embryonic leaves (ectoderm, mesoderm and endoderm). Although its etiology is not yet fully known, teratomas may result from the migration of totipotent cells near the Hensen node in the embryo. Teratomas, especially those diagnosed in the neonatal period, occur most frequently in the sacrococcygeal region. Nevertheless, they can be found in other locations depending on the location of migrating totipotent cells [1, 6].

Our case had a sacrococcygeal location of the mass. Many others found the same location. Out of 52 cases, Rattan et al found 50 children (96,15%) with saccrococcygeal teratoma [7]. Sathyanarayana et al in their study discussed of about 43%

of teratomas sacrococcygeal teratomas (far from the commonest), a relatively high incidence of retroperitoneal tumors occupying second place, constituting 25% (13 cases) of teratomas as against the other series which ranges between 5-10%; likewise Barksdale finds nearly 50% of cases with this position [5, 8].

By looking through the literature, it appears that most forms are diagnosed at birth. The average age of discovery of this pathology were different ranging from 4 days to 11 months with extremes ranging from 1 day to 5 years. Rescola et al. found an average of 11 months (extremes 1.5 days – 34 months), Kundal et al – 4 days (1 day – 4 years) [9, 10]. Rattan et al discored the age ranged from newborn to 13 years. Thirty-three cases presented in the neonatal period, 10 in neonatal to infancy period, 5 in 1-5 year age group, 1 in 5-10 year age group and 3 children were more than 10 years old. Authors thought this was a cause of poor antenatal supervision [7]. In our case, the tumor was discovered at birth. Advances in medicine, especially ultrasound, and fetal medicine allow not only the recognition of this pathology during pregnancy, but also specify some prognostic factors giving better information of the couple. The prenatal knowledge diagnosis of sacrococcygeal teratomas contributes to the improvement of obstetric follow-up and allows to predict the delivery in a specialized structure for a better pediatric and surgical management.

As in the case we present, many authors have discovered the predominance of the female sex. In the study performed by Rattan et al, there were 40 females and 12 males with age ranging from newborn to 13 years [7]. Altman and Rescorla, each, found 74% female in their study out of a total of 403 and 126 cases respectively [9, 11]. Higher proportions have been described by authors as Gabra et al (85%) et Schmidt et al (82,6%) [12, 13]. Balanced sex ratio was found in some series; familial cases of TSC have been described by Ashcraft and Holder; they suggested that the tumor is an inherited autosomial dominant. The forms of this family has several characteristics that distinguish it from the usual TSC (balanced sex ratio, low prevalence of malignant forms, more o less constant associated malformations with particular: sacral abnormalities, anorectal stenosis, vesico-ureteral reflux) [14].

Our case was noticed to have large swelling (mass) in the sacrococcygeal region, having solid consistency with normal urinate and defecation. Sharma et al described a case with large swelling in the sacrococcygeal region with an approximate size of 10*12 cm, with solid consistency and erythematous. There was scrotal skin thickening with hydrocele and anal opening was present in the center of the mass. There was differential edema with only right lower limb having pitting edema [15]. Rattan et al discovered many children with irregular mass with variable consistency in the sacrococcygeal, the size of the mass varied from 3 to 30 cm. About half of the cases had a vast mass of more than 15 cm [7]. More complex or even organoidal aspects have been described, evoking limbs of limbs and a cephalic extremity carrying a well-drawn lip, well-formed fingers with fingernails [16, 17]. The variable nature of TSC extension explains some late discoveries by signs of compression and at a stage where malignant transformation is not uncommon.

Non-externalized forms tend to extend upwards and forwards into the pre-sacral space, often producing signs of compression following displacement of the rectum and pelvic organs [17-19].

Several complementary examinations may be used for diagnosis and to evaluate the extension and thus classify the case. With Ultrasonography it is possible to specify the echo-structure of the mass (liquid, solid or mixed), to indicate the existence of calcification, to show the presence of hemorrhage, unilocular or multilocular cysts, greasy zones, to objectify the intra pelvic extension, to identify the relations with the bladder, the rectum, to explore the urinary tree in search of a possible repercussion with type of bladder dilation or ureterohydronéphrosis, to follow and to detect a possible recurrence [20-22]. Computed tomography determines the tumor composition (cyst, fat, and calcifications). This test is used to locate the teratoma with respect to the pelvic organs, in particular the rectum and the bladder, and to delimit the pelvic bone structures, thus showing the coccygeal attachment. The Scanner shows the endo-pelvic extensions of small sizes that could escape the ultrasound. Oral or intravenous administration of contrast enhances the chances of visualizing anterior repression of the rectum and bladder [23-25]. Concerning medical imaging, other diagnostic methods such as standard radiography, MRI, which we have not used in our case, are also very important for the diagnosis in such affection [26, 27].

Biological markers are of great interest in the diagnosis and the follow-up of this type of patients. Alfafoetoprotein (AFP) is one of the markers. Its elevation is the best indicator in the presence of an endodermal sinus tumor. But in the newborn and the infant, this rate can be physiologically high. The normal AFP level at birth can be 100,000 ng / ml. The interpretation of the AFP rate is only possible thanks to the Tsuchida chart [28, 29]. This marker is considered as an important key for the clinical differentiation between benign and malignant teratomas, as well as for evaluating the efficacy of treatment [30]. In addition to its prognostic value, the AFP assay is an excellent means for postoperative follow-up; as it falls after tumor resection, and rises in case of recurrence or metastases. According to some studies, normalization of AFP after tumor resection occurs between 6 and 12 months, however, its recommended dosage every 3 to 6 months for 3 years after surgery [28, 31].

the clinic and complementary examinations allowed us to classify our patient into the SCT type II of Altman's classification. A topographical classification of TSCs was proposed by 1974, Altman et al in their report to the American Academy of Pediatric Surgery. According to that classification, SCT is divided into 4 type [11]:

1) Type I: Tumors are external, with a small presacral component, and carry the best prognosis (46%).
2) Type II: tumors are predominantly external with a large intrapelvic portion (35%).

3) Type III: lesions are predominantly intrapelvic with abdominal extension with only a minor external component (9%).
4) Type IV: lesions are entirely intrapelvic and abdominal (10%).

Concerning our case, a surgery was performed with a sacral approach. Tumor excision is the only effective treatment for TSC. It is indicated as soon as the diagnosis is made, even in the first days of life. The objectives of the intervention varies, such as the fast and complete resection of the mass to avoid the risk of malignant transformations, which is increasing with time, the removal of the coccyxid recurrence, which is more readily in a malignant form, even if the tumor was initially benign. The reconstruction of the perineal muscles to ensure anal continence. The restoration of a perineum and a gluteal region of normal appearance. Different types of approaches are used, including abdominal and sacral [32-35]. Other surgical techniques have been proposed. Jan et al. proposed a sagittal incision to limit tissue damage, due to the sagittal orientation of muscles in this region, and to achieve better esthetic results [35]. Laparoscopy offers an excellent vision of this narrow pelvic space, thus preserving the sphincter and nerve structures, guaranteeing a good functional result later. Solari et al. reported the case of a newborn with a large, ulcerated TSC who underwent surgery on the first day of life. The ligation of the middle sacral artery was performed successfully by laparoscopy, avoiding laparotomy and transfusion [36].

The diagnosis of certainty of sacro-coccygeal teratomas is based on the histological examination of the operative specimen. Three histological types of prognostic interest are to be distinguished: Type 1 (mature teratoma or dermoid cyst of benign nature); Type 2 (mixed teratoma of questionable malignancy); Type 3 (immature teratoma of certain malignancy) [37]. For our case, on microscopic examination, tumors were classified into good (mature) category. The predominant component was ectodermal tissue. 70% of benign tumors are composed of mature tissue structures. The different samples can show various well-differentiated tissues, organoids [38]. Malignant tumors, which account for approximately 13.2% of sacrococcygeal teratomas, are essentially solid tumors with intratumoral calcification, criteria for maturation. Tumors with liquid contents would have a lower risk of malignant transformation [39]. Some authors believe that differentiation into mature and immature does not correlate with the prognosis of SCT [40]. Risk of malignancy is associated with large tumor size (>10 cm) in type III and IV due to delay in diagnosis and when the presentation is beyond the neonatal period [11]. Donnellan and Swenson showed in their study a percentage of malignancy of 91.7% after the age of 2 months [41].

4. Conclusion

Sacro-Coccygeal Teratomas are rare congenital tumors, most often diagnosed in the neonatal period, but sometimes the discovery is made after birth. The variable clinical presentation is most often in the form of a sacral mass or gluteal at birth, with or without signs of urinary or digestive compression later. Complete radical tumor excision is the only effective treatment for TSC. It prevents recurrence, which is more readily in a malignant form, even if the tumor was initially benign. The anatomopathological study confirms the diagnosis.

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