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ORIGINAL PAPER

A retrospective analysis of newborns with congenital teratomas

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ABSTRACT

Introduction: The aim of this study was to analyse cases of neonates with congenital teratomas and to compare the accuracy of a prenatal teratoma suspicion with histological confirmation after birth.

Material and methods: The authors conducted a retrospective study including 9 patients with a prenatally suspected teratoma, born and treated in the period 2019–2023 at a tertiary care hospital. Medical records of all cases were analysed and the data of 8 patients, whose diagnosis was histopathologically confirmed after birth, were included in the analysis. Information such as gender, maternal morbidities, course of pregnancy and delivery, symptoms, results of diagnostic tests, histopathological reports and postoperative complications were analysed, compared and summarised.

Results: In all patients, the initial diagnosis was made prenatally. Each child was delivered by a caesarean section due to fetal anomalies and concomitant problems (premature rupture of membranes, mirror syndrome, chorioamnionitis, birth asphyxia). Three patients were additionally diagnosed with other congenital malformations: coarctation of the aorta, ventricular septal defect, hydrops fetalis. Six of the 8 children were operated on, and 2 died on the day of birth. A complete resection of the lesion was confirmed in 5 children, and a follow-up ultrasound was recommended for 1. A coccygectomy was performed in all patients with sacral teratomas. The average length of stay in the Neonatal Intensive Care Unit was 54 days. Of all surviving children, 1 required transport to another centre. The remaining patients were discharged home in overall good condition.

Conclusions: Surgical resection of fetal tumours is still extremely difficult. The only chance for the child's survival is a quick diagnosis, delivery at a tertiary referral hospital, stabilisation of the newborn at birth and surgery in the shortest possible time. If the course of pregnancy, the surgery and the postoperative period go well, the prognosis is favourable.

KEY WORDS:

prenatal diagnosis, newborn, teratoma, sacrococcygeal region.

INTRODUCTION

Sacrococcygeal teratomas (SCTs) are rare but the most common tumours in children under 1 year of age [1, 2]. These neoplasms derive from all 3 germ layers, and therefore can be composed of any tissue or cell mass without cellular differentiation [1, 3]. Morphologically, teratomas can be classified as solid, cystic or mixed. The most common tissues present within the lesion are: nerve, cartilage and bone tissues, skin, muscles, respiratory or

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intestinal epithelium. Although teratomas can be present in almost any part of the body, they are usually localised in distinctive regions: sacrococcygeal, gonadal, mediastinal, retroperitoneal and cervicofacial or intracranial. Extragonadal tumours are most common in neonates [4, 5]. Due to different locations of the lesions in relation to the sacrum and coccyx, a classification system for SCTs was created by Altman. Type I includes tumours with a minimal presacral component. Type II describes predominantly external lesions having a significant intra-abdominal component. Type III tumours are mainly located inside the pelvis and may have a small external component. Type IV includes entirely presacral lesions [3]. Teratomas detected in the perinatal period mainly originate from Hensen's node, located in the sacrococcygeal region, or caudal pluripotent primitive stem cells [1, 3, 5–7]. The size of the tumour and symptoms it causes during pregnancy may affect the course and the length of gestation [2]. Factors affecting an increased blood flow within the lesion (large tumour size, rapid growth and solid type) are the main causes of complications. The subsequent prognosis mainly depends on the type of the tumour. Ultrasonography is the most accessible method for diagnosing sacrococcygeal tumours [6, 7]. In cases of prenatally diagnosed teratomas, which account for half of cases, the risk of a fetal death reaches 16%, while the risk of a neonatal death is 24%. Arteriovenous shunting through the tumour can lead to heart failure, hydrops fetalis and death in extreme cases [1, 5].

The aim of this study is to analyse cases of newborns with congenital teratomas and to compare the accuracy of a prenatal teratoma suspicion with the histological confirmation after birth. The perinatal history, clinical course of a disease and treatment were described and compared. The authors present 9 newborns with a prenatal diagnosis of a teratoma.

MATERIAL AND METHODS

The study involved 9 patients with prenatally suspected teratomas, born and treated in the period 2019–2023 in a tertiary care centre. The analysed information included: gender, prenatal and postnatal diagnosis, symptoms induced by tumours, maternal morbidities, course of pregnancy and delivery, diagnostic test results, histopathology reports, serum levels of tumour markers (α-fetoprotein – AFP, the β subunit of human chorionic gonadotropin), surgery, postoperative complications and deaths. Medical records of all cases were reviewed and the data of 8 patients, whose diagnosis was histopathologically confirmed after birth, were included in the analysis. In 1 case, the histopathology report (vascular malformation) did not confirm the prenatal diagnosis of a teratoma and the patient's medical history was excluded from the study. The data were analysed, compared and summarised.

RESULTS

The gender distribution of patients included in the analysis was equal (4 girls, 4 boys). Seven newborns were born at our centre; 1 was transferred from another hospital. The course of pregnancy was complicated in 5 cases: hypothyroidism in 3, gestational diabetes in 2. In addition, 1 mother with gestational diabetes was diagnosed with a maternal mirror syndrome (a complication of non-immune foetal oedema; association of fetal and placental hydrops with maternal oedema and preeclampsia). Due to the deterioration of the woman's clinical condition, it was necessary to perform a caesarean section at 26 weeks of pregnancy. The child died on day 1 of life.

Three of 8 patients required an intrauterine laser ablation of tumour supplying vessels; moreover, 1 child underwent triple amnioreduction. Additional congenital anomalies were found in 3 neonates: coarctation of the aorta (1 case), ventricular septal defect (1), hydrops fetalis (1). A genetic disorder was suspected in 2 cases due to a facial dysmorphia.

All children were delivered by a caesarean section due to fetal malformations. Additional indications included: premature rupture of membranes (PROM) (4 cases), maternal mirror syndrome (1), chorioamnionitis (1) and birth asphyxia (1). The average gestational age at birth was 33 weeks (the shortest: 26 + 5/7, the longest: 39 + 0/7) and the birth weight with tumour was 2803.75 g.

After birth, 4 neonates required cardiopulmonary resuscitation and/or intubation. The remaining 4 were born in overall good condition, with a score of 9 or 10 on the Apgar score. No gastrointestinal disorders were observed in 6 neonates, although the anus was displaced by the tumour in 3 of them. In addition, dislocation of both femoral heads was observed in 1 case and a complete retraction of the anus by the lesion was diagnosed in 1 newborn. The urinary system functioned properly in 6 patients, and 1 neonate required the insertion of a Foley catheter due to difficulty urinating. One of the children, who died on day 1 of life, did not urinate. Imaging tests were performed after birth (Figures 1 and 2).

Serum levels of tumour markers were measured. Alpha-fetoprotein levels were determined in 4 children; the average was 42,618.25 μg/l preoperatively, and the highest level was 102,146 μg/l. Follow-up tests after an operation showed a decrease in the marker in all patients. The β subunit of human chorionic gonadotropin level averaged 7.3 IU/l (max. 16 IU/l).

With the exception of 2 patients with SCTs, who died on the day of birth, the remaining 6 were scheduled for surgery (3 with a sacrococcygeal, 2 with a lumbosacral and 1 with a craniofacial tumour). The surgery was performed on average on day 5 of the newborn's life (4.66; range 1–8). A coccygectomy was performed in all 5 patients with sacral teratomas (complete – 2 cases, incom-

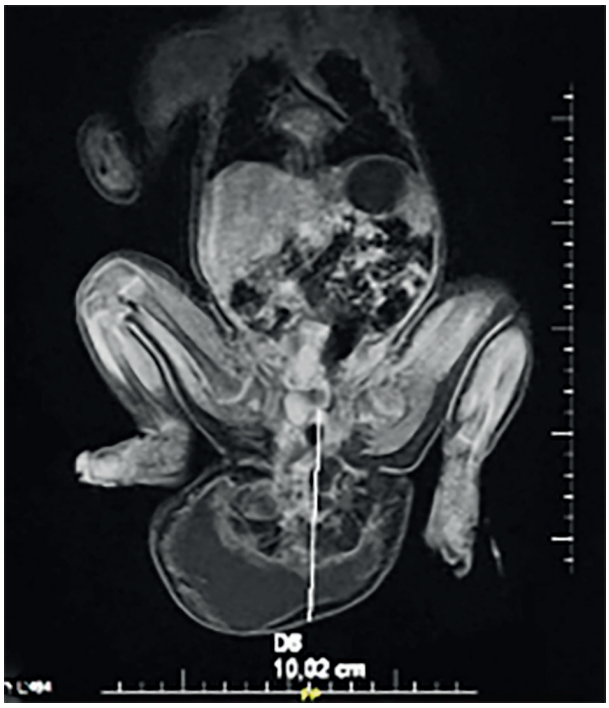

FIGURE 1. Magnetic resonance imaging of the abdomen and pelvis, lumbosacral teratoma

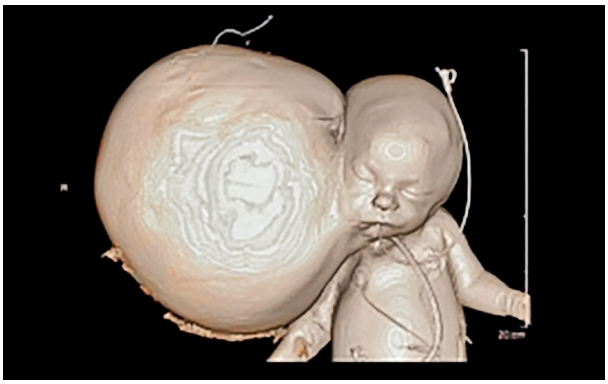

FIGURE 2. Head computed tomography scan with three-dimensional reconstruction, craniofacial teratoma

TABLE 1. Details of the analysed teratomas

Aspect	*n* (%)
Location (*n* = 8)	
Sacrococcygeal	5 (62.5)
Lumbosacral	2 (25)
Craniofacial	1 (12.5)
Morphological characteristics (*n* = 8)	
Solid, with internal calcifications	1 (12.5)
Cystic, with multiple septa	3 (37.5)
Mixed, with fluid compartments, disintegrating tissues, bleeding	4 (50)
Histopathology report (*n* = 6)	
Mature teratoma	4 (66.7)
Immature teratoma	1 (16.7)
Low malignant potential lesion	1 (16.7)
Type in the Altman classification for sacrococcygeal teratomas (*n* = 3)	
I	1 (33.3)
II	1 (33.3)
III	1 (33.3)
IV	0 (0)

plete – 3). Resected tumours were histopathologically examined; moreover, SCTs were categorised in the Altman classification system (Table 1).

The post-operative period was uneventful in 1 child; the others experienced: faecal incontinence (1), inflammatory infiltration in the wound area (1), elevation of inflammatory markers (1). Tumour rupture was noted during 3 operations. One patient developed a particularly severe condition with a respiratory failure, coagulation disorders, infection and impaired wound healing. The child with the craniofacial teratoma was found to have facial nerve palsy and intracranial haemorrhage. In the analysis we conducted, all tumours were diagnosed prenatally, at the earliest in the 13th week of gestation. They were characterised by their significant size, the smallest being 8 × 10 cm and the largest 25 × 30 cm (the craniofacial teratoma). Of the 9 children with a prenatal teratoma suspicion, the diagnosis was confirmed postnatally in 8 (88.89%).

The hospitalisation of 5 patients ended with discharge home in overall good condition. One child was transferred to a centre preparing for home respiratory ventilation. The average length of stay in the Neonatal Intensive Care Unit was 54 days (range 15–115).

DISCUSSION

The incidence of a teratoma is 1/35,000–40,000 live births with a predominance in women (2–4/1) [1–5, 7, 8]. In the group of patients analysed by the authors, the gender distribution was equal, in contrast to the distribution reported in the literature. Despite their infrequent occurrence, they are the most common tumours found in fetuses and neonates [1, 3, 5–10]. However, recent reports indicate that the incidence may be higher, with prenatally diagnosed lesions accounting for half of cases [1]. The majority of teratomas are located in the sacrococcygeal region, and other common locations include the head and neck, thorax and retroperitoneum [11]. A significant part of teratomas are benign tumours, but the risk of malignant transformation increases with age [5, 7, 8]. In the above analysis, a histopathological examination of the tumours was performed after their removal. In none of the newborns was a tumour biopsy performed preoperatively, due to a high risk of injury and haemorrhage.

Sacrococcygeal teratomas can be detected during a fetal ultrasound examination or at birth, as a mass at

the base of the coccyx [5]. With an ultrasound scan, not only can the tumour be visualised as early as the second trimester, but the pregnancy can be monitored for fetal cardiovascular endurance, and the observation of other congenital anomalies is also possible. A teratoma usually presents as a lesion with a heterogeneous echogenicity, containing solid and cystic structures, and may show varying degrees of blood supply [1, 7]. Most tumours are diagnosed prenatally, on ultrasound scans, as an intrauterine mass [9, 10]. However, when the tumour mass is completely within the child's pelvis, an SCT may be diagnosed late [12]. According to the Altman classification, in patients with external (or partly external) components (type I, II and III), the lesion may be noticed early. Type IV tumours (entirely presacral) might pass unnoticed [3]. In cases described by the authors, type I–III teratomas were also diagnosed prenatally, at the earliest in the 13th week of pregnancy. No type IV tumour was identified. When imaging findings are atypical or the clinical presentation is inconclusive, differential diagnosis is essential. Conditions that should be considered include: myelomeningocele, neuroblastoma, vascular malformations, sacral chordoma, sacral meningocele, terminal myelocystocele, enteric cysts in cystic types, meconium pseudocyst and obstructive uropathies [1, 5]. Accurate differentiation is crucial, as each of these cases requires different treatment. In our study, one case, initially suspected to be a teratoma, was ultimately diagnosed as a vascular malformation. It highlights the importance of postnatal histopathological verification.

The management of SCTs diagnosed at birth differs from management of those diagnosed prenatally, which are difficult to treat due to the highly unpredictable dynamics of development and the associated poorer prognosis [1, 2, 10]. Prenatal management methods include amnioreduction, cyst aspiration, radiofrequency thermal (or laser) ablation of the tumour's blood supply, tumour embolization and open surgical resection of teratoma [5, 6]. These interventions are associated with a high risk of complications. Ablation may result in a serious defect that requires subsequent surgical procedure. Fetal surgery may result in chorioamniotic separation, PROM or fetal pulmonary oedema [5].

Diagnosis during pregnancy is related to a higher risk of placentomegaly, hydrops fetalis, anaemia, polyhydramnios and circulatory failure, which results in a higher risk of preterm birth and high mortality [1, 5, 6]. Certain tumours do not increase in size, while others grow rapidly, leading to life-threatening complications, including deterioration of renal function and lung hypoplasia due to bladder compression and urinary retention. Teratomas characterised by a worse prognosis are greater than 10 cm in diameter or contain cystic structures [6, 10, 13]. All tumours described in this analysis were characterised by significant size, the smallest having dimensions of 8 × 10 cm and the largest 25 × 30 cm.

A prenatal ultrasound is an effective examination for detecting a teratoma, but magnetic resonance imaging is the tool with a greater sensitivity for assessing abdominal and pelvic organs [1]. Other factors, such as maternal obesity, the amount of amniotic fluid and fetal positioning, may influence the interpretation of the ultrasound image [6]. Magnetic resonance imaging is recommended when ultrasound findings are unclear and a precise imaging of the fetus is required [1]. It is also useful in determining the extensiveness of SCTs and differentiating them from other fetal tumours [12]. Assessment of the size of a teratoma is important in order to choose the optimal mode of delivery and the appropriate surgical planning after birth [1].

Solid tumours are characterised by hypervascularisation and malignancy, which increases the risk of bleeding and often requires intrauterine ablation of the feeding vessels [2]. This procedure reduces the size of the tumour, preventing fetal circulatory failure and improving the overall outcome [6]. In the present study, as many as 3 out of 8 patients required intrauterine laser ablation; moreover, 1 child underwent a triple amnioreduction. Additional complications associated with a worse prognosis include: polyhydramnios, cardiac dysfunction, hydrops fetalis and placentomegaly [2, 6]. In the analysed patients, heart defects were found: coarctation of the aorta in 1 case and ventricular septal defect in 1 neonate. Generalised oedema occurred in 1 patient; this child died on day 1 of life.

A proper evaluation of complications caused by the presence and size of the teratoma is possible after birth. Functions of the gastrointestinal and urinary systems, which are most often affected, are assessed. In most newborns, symptoms are equivocal and appear late, most commonly as urinary tract and/or bowel obstruction [12]. The tumour pressure on adjacent organs can lead to gastrointestinal abnormalities such as intestinal obstruction and faecal retention [1, 3]. According to the literature, up to 25% of infants with an SCT have additional birth defects: urethral or anal stenosis, rectal atresia or urinary tract obstruction [5]. In the majority of analysed children, no gastrointestinal disorders were observed, despite a displacement of the anus by the tumour mass in 3 patients. The urinary system functioned properly in 5 newborns; only 1 required the insertion of a Foley catheter due to difficulties in urination. In addition, 1 child was found to have dislocation of both femoral heads and complete retraction of the anus by the tumour.

The mode of delivery depends on the size and vascularisation of the tumour [1]. In most cases, when the teratoma is larger than 5 cm in diameter, a caesarean section is indicated. This is especially relevant for hypervascular tumours, which are more likely to rupture and bleed [2, 5, 6, 8]. Natural childbirth is associated with the risk of dystocia and trauma, which can result in rupture of tumour tissues, haemorrhage with anaemi-

sation of the baby and the necessity of a blood product transfusion [1]. All children analysed by the authors were delivered by a caesarean section due to fetal malformations; additional indications mainly included PROM. For a high-risk SCT, intrauterine surgery may be indicated. The aim of the treatment is to reduce the impact of the tumour mass on the fetal cardiovascular system and to allow the fetus to grow and mature properly [10]. There were no indications for prenatal teratoma surgery in any of the analysed children.

A procedure involving complete removal of the lesion (the traditional treatment approach for infants with SCT) should be performed as soon as possible after birth [1–3, 5, 10]. It reduces the risk of tumour rupture, bleeding and malignant transformation, which is 20% two months after birth and doubles after 4 months [3, 6]. In most cases, the coccyx is additionally removed, which is often an integral part of the surgery [1–3, 5, 6, 8–10]. Leaving the coccyx and tumour rupture during a surgery are the main risk factors for a tumour recurrence [3, 5, 6, 9]. During surgery, the possibility of scarring and deformities should also be taken into consideration, as well as proper nerve and muscle function. The gluteal region should be reconstructed with enhanced cosmetic closure, by gathering 4 skin flaps to leave a centrally located, hidden scar. Additional attention should also focus on the possibility of intraoperative blood loss in the case of highly vascularised lesions and urethral complications associated with SCT removal. Administration of blood products operatively and postoperatively may be necessary [5].

If malignancy is diagnosed, chemotherapy might be indicated [1, 9]. It should be performed prior to mutilating surgery in malignant cases and procedures requiring excision of vital organs in benign SCT cases [9]. Post-operative chemotherapy is not recommended – it is not effective in preventing recurrence. Chemotherapy may also be required in patients with recurrent SCTs, which tend to have a malignant histology [14]. These tumours respond well to this treatment modality, giving high cure and survival rates [3, 9]. In our analysis, none of the children required chemotherapy. Malignant teratomas can be classified into 4 stages. Stage I: tumours resected completely, with coccygectomy for sacrococcygeal site, clean margins. Stage II refers to the presence of tumour cells at the resection margin, without lymph node involvement. In stage III, full surgical removal is not done or only a diagnostic biopsy is performed. Tumour markers may be negative or positive in stages I–III. Distant organ metastases with liver involvement are included in stage IV [3].

Alpha-fetoprotein is the most reliable tumour marker for detecting malignant recurrences in paediatric SCTs, being a sensitive and early indicator of malignant transformation. It is typically elevated in all cases of malignant recurrence, with an increase observed 2–3 months before clinical diagnosis. However, due to physiological elevation during infancy, AFP interpretation requires age-adjusted reference values [15]. Immediately after birth, AFP level is in the range 17,200–44,300 μg/l. An average concentration of 158,125 μg/l can be found in prematurely born newborns, which is attributed to low body weight. Alpha-fetoprotein level gradually decreases in the first 12 months after birth and reaches values typical for adults (max. 10–15 μg/l) [16]. However, AFP level measurements are not fully used in the oncological diagnostics in neonates and infants, due to the inaccurately defined AFP norm ranges in these groups [17]. It is also worth noting that high AFP levels were briefly observed in approximately one-third of neonates without recurrence during the first 18 months of life. In addition, elevated cancer antigen 125 levels might be indicative of non-malignant recurrence. However, more cases need to be analysed to determine the value of this marker [15].

If an SCT is diagnosed prenatally, perinatal mortality reaches 50%. The most common causes of premature death are heart failure, perioperative bleeding and internal haemorrhage. An early resection of the lesion is associated with a better prognosis [10]. It depends on 3 factors: the time of the diagnosis, the tumour type and the surgical difficulty of resection [5].

FOLLOW-UP

Children diagnosed with an immature teratoma or malignant potential lesion remain under multispecialty care. Periodic follow-up imaging and laboratory tests show no recurrence of the tumour in any of the children to this day. Similarly, in a child with incomplete surgical resection of the tumour and a recommendation for frequent USG follow-ups every few months, there has been no sign of tumour recurrence. In one of the two children with suspected genetic disorder, Noonan syndrome was confirmed. In the second child, despite the presence of teratoma in the lumbosacral region, facial dysmorphia and coarctation of the aorta, no genetic disorder was found.

CONCLUSIONS

Despite developments in perinatology, a surgical resection of a fetal tumour is still extremely difficult. The only chance for the child's survival is a quick diagnosis, delivery at a tertiary referral hospital, stabilisation of the newborn at birth and surgery in the shortest possible time. This analysis has shown that if the course of pregnancy, the surgery and the postoperative period go well, the prognosis is favourable.

DISCLOSURES

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