

# Sacrococcygeal teratoma in the perinatal period

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Teratomas are formed when germ cell tumours arise from the embryonal compartment. The name is derived from the Greek word "teras" which literally means "monster". The ending "-oma" denotes a neoplasm.<sup>1</sup>

## Incidence

Sacrococcygeal teratoma is the most common congenital tumour in the neonate, reported in approximately 1/35 000 to 1/40 000 live births.<sup>2</sup> Approximately 80% of affected infants are female—a 4:1 female to male preponderance.<sup>2</sup>

The first reported case was inscribed on a Chaldean cuneiform tablet dated approximately 2000 BC.<sup>3</sup> In the modern era, the first large series of infants and children with sacrococcygeal teratomas was reported by Gross *et al* in 1951.<sup>4</sup>

## Embryonic origin

Sacrococcygeal teratomas have tissues derived from ectoderm, mesoderm, and endoderm.<sup>2</sup> Although their embryonic origin is still uncertain, they are believed to arise early in gestation (at around the late second or early third week) from the totipotential cells of Hensen's node (also called the primitive knot), a remnant of the primitive streak in the coccygeal region.<sup>5-7</sup> The primitive streak appears as a linear thickening in the ectoderm at the caudal edge of the bilaminar embryonic disc. It usually diminishes in size, eventually disappearing after undergoing degenerative changes. As the mesoderm rapidly proliferates, the primitive streak comes to lie more and more caudally, where the remnant of Hensen's node descends to the tip or anterior surface of coccyx.<sup>5-7</sup>

## Types

Sacrococcygeal teratomas may be classified as benign (mature) and malignant or immature (composed of embryonic elements).<sup>8</sup> Mature teratomas are most common in neonates (68%) and older children (73%). Immature teratomas are cystic, whereas malignant tumours are solid. Over 50% of sacrococcygeal teratomas have calcification and ossification.<sup>8</sup> Keslar *et al* reported that 69 (62%) of the 96 sacrococcygeal teratomas in their series were composed of both solid and cystic elements.<sup>8</sup> The cysts may be filled with serous fluid, mucoid, or sebaceous material and lined by true epithelium. Ein *et al* found cystic tumours that were filled with cerebrospinal fluid from choroid plexus present in the tumour mass.<sup>6</sup> Virtually any tissue can be present in a sacrococcygeal teratoma.<sup>8 10-13</sup> Neuroglial tissue, skin, respiratory and enteric epithelium, cartilage, smooth muscle, and striated muscle are the most common elements found.<sup>8</sup> Bone, pancreatic tissue, choroid plexus, and adrenal

tissues are less commonly identified.<sup>12</sup> An ocular lens present as lentiginoids (lens-like cells), as well as a completely formed eye, have been found within sacrococcygeal teratomas.<sup>10 11</sup> Parizek *et al* reported a mature teratoma containing the lower half of a human body in one of fraternal twins.<sup>13</sup>

## Size

Size of a sacrococcygeal teratoma (average 8 cm, range 1 to 30 cm) does not predict its biological behaviour.<sup>8</sup> Altman *et al* have defined the size of sacrococcygeal teratomas as follows: small, 2 to 5 cm diameter; moderate, 5 to 10 cm diameter; large, > 10 cm diameter.<sup>14</sup>

## Site

The sacrococcygeal region is the most common location. Less common sites are the mediastinum, testes, retroperitoneum, brain, head and neck, vagina, stomach, and pineal region.<sup>8 15</sup> Sacrococcygeal teratomas may continue to grow posteriorly to form an external protrusion, or dissect anteriorly, distorting regional organs (rectum, vagina, and bladder) without invading them.<sup>8 14</sup>

## Extent

The American Academy of Pediatrics' surgical section (APPSS) classification helps in grading the extent of sacrococcygeal teratomas, as follows<sup>16</sup>:

- Type I—Predominantly external with minimal presacral component.
- Type II—Present externally but with significant intrapelvic extension.
- Type III—Apparent externally but predominantly a pelvic mass extending into the abdomen.
- Type IV—Presacral with no external presentation.

## Histology

Sacrococcygeal teratomas are graded histologically as follows<sup>17</sup>:

- Grade 0—Tumour contains only mature tissue.
- Grade 1—Tumour contains rare foci of immature tissues.
- Grade 2—Tumour contains moderate quantities of immature tissues.
- Grade 3—Tumour contains large quantities of immature tissue with or without malignant yolk sac elements.

Grading of sacrococcygeal teratomas, unlike that of ovarian teratomas, does not seem to correlate directly with prognosis.<sup>8</sup>

## Fetal diagnosis

Though described as early as the 19th century, it was not until the advent of antenatal ultrasonography that sacrococcygeal teratomas

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Figure 1 Antenatal ultrasound (sagittal section) at 22 weeks of gestation, revealing a large sacrococcygeal teratoma (courtesy of Sinh Le and the Department of Radiology, Kirwan Hospital for Women).

were diagnosed in utero<sup>18</sup> (fig 1). The majority of sacrococcygeal teratomas present between the 22nd and the 34th week of gestation. Most are benign, resectable after birth without significant morbidity, and rarely associated with other congenital anomalies. There is thus the potential to lead a normal life without disability.<sup>18</sup> The diagnosis of sacrococcygeal teratoma on routine sonograms is associated with a greater than expected incidence of prenatal and perinatal complications.<sup>19</sup> Close antenatal follow up is needed to optimise patient counselling and treatment in the presence of a completely solid tumour and the onset of polyhydramnios. A poor outcome is usually correlated with placentomegaly, cardiomegaly, or non-immune hydrops fetalis.<sup>19</sup>

#### Associated malformations

The incidence of various congenital malformations associated with sacrococcygeal teratoma ranges from 5% to 26%.<sup>4 6 20</sup> Of these, anorectal and genital malformations are of prime concern. The association of sacrococcygeal teratoma with anorectal malformations was described as early as 1935.<sup>21</sup> During the third week of embryonic life, genital folds unite to form the genital tubercle by migration of mesenchymal cells from the primitive streak region around the cloacal membrane. During the fourth and seventh weeks, the cloaca is subdivided by the urorectal septum to form the anorectal canal and the primitive urogenital sinus.<sup>21</sup> A growing sacrococcygeal teratoma could thus encroach between the layers of the cloacal membrane and prevent descent and fusion of the urorectal septum to the cloacal membrane, resulting in a high anorectal malformation with a rectourethral fistula.<sup>21</sup> An anterior location of the tumour could thus result in the absence of rectum and anus.<sup>21</sup> The physical presence of a teratoma could also prevent fusion of the genital folds, resulting in a bifid scrotum and hypospadias.<sup>21</sup>

Other associated anomalies include spinal dysraphism, sacral agenesis, dislocation of the hips caused by a large tumour, and meningocele.<sup>8</sup> Rarely cardiac anomalies such as a ventricular septal defect or gastrointestinal

anomalies other than imperforate anus have been described.<sup>6 14</sup> Vogl and Riel reported a case with anorectal malformation, sacral dysplasia, and a presacral mass (Currarino's triad).<sup>22</sup> Lahdenne *et al* reported vertebral abnormalities in 80% of their 45 patients with benign sacrococcygeal teratoma.<sup>23</sup>

#### Perinatal management

The perinatal management of sacrococcygeal teratoma requires an interdisciplinary liaison between obstetricians, neonatologists, and paediatric surgeons. The high perinatal/neonatal mortality and morbidity rates associated with sacrococcygeal teratoma are attributed to preterm delivery and to complications such as malignant invasion, haemorrhage into the tumour, obstruction of umbilical flow, high output cardiac failure, hydrops fetalis, and bladder outlet obstruction.<sup>19 24–28</sup> Some of these complications can be detected prenatally and treated appropriately.<sup>29</sup> Garcia *et al* reported a case of decompression of a cystic grade IV sacrococcygeal teratoma in utero through an amniotic catheter to alleviate hydronephrosis and potential renal damage.<sup>30</sup> Bond *et al* postulated that the tumour acts as an arteriovenous fistula, resulting in high output cardiac failure with placentomegaly and non-immune hydrops. If this were true then intervention to control this circulatory steal may be the only way to salvage severely affected fetuses.<sup>26</sup> Close follow up with repeated ultrasound scans is necessary to monitor an increase in tumour size, extension into adjacent structures, tumour vascularity, and evidence of cardiac failure.<sup>18</sup> Volume reduction amniocentesis and tocolysis may be required to treat symptomatic polyhydramnios and prevent preterm delivery. Barring the development of hydrops or maternal complications, delivery should be delayed to allow for fetal maturation. If hydrops develops, close maternal–fetal observation is needed. Maternal corticosteroids are advisable to accelerate fetal pulmonary maturation.<sup>31</sup> Termination of pregnancy may have to be considered in cases with bad prognostic signs (for example, cardiac failure, placentomegaly, marked hydrops) or with associated major congenital anomalies. Kum *et al* described the following prerequisites for consideration of fetal intervention in preterm pregnancy<sup>18</sup>:

- accurate prenatal diagnosis and well defined natural history to allow the confident diagnosis of a correctable lesion that will otherwise prevent fetal survival;
- absence of other life threatening or debilitating anomalies;
- ability to perform the procedure without increased risk to the mother's life or her future fertility.

The combination of fetal hydrops and sacrococcygeal teratoma is considered to be lethal.<sup>32</sup> Nakayama *et al*, however, reported two such cases who survived after preterm delivery by caesarean section.<sup>32</sup> They found that the combination of sacrococcygeal teratoma and hydrops had a particular effect on pulmonary function and fluid balance. Removal of tumour led to an immediate improvement in respira-

tory function. They proposed the following tumour related factors contributing to respiratory insufficiency:

- the increased blood volume represented by tumour circulation may have exceeded the capacity of baby's lungs to arterialise blood;
- venous blood from the tumour may have contributed to a low mixed venous  $PO_2$  and pulmonary vasoconstriction; excision may have removed the source of desaturated acidaemic blood and relieved pulmonary vasoconstriction;
- fetal ascites and abdominal distension may have embarrassed lung development in utero, resulting in reduced lung volumes;
- ascites raised the diaphragm after birth and paracentesis thus improved pulmonary function.

### Investigations

Sacrococcygeal teratomas should be excised soon after delivery to avoid tumour ulceration and haemorrhage and to reduce the risk of malignant change.<sup>18</sup> The following investigations are recommended before surgery:

- x rays of spine, abdomen, chest, and pelvis to detect evidence of tumour or metastasis<sup>33 34</sup>;
- ultrasound to define the extent, nature (cystic or solid) of the mass, the presence and size of solid elements or calcification, and to recognise intrapelvic extension<sup>35</sup>;
- computed tomography to delineate the bony pelvic structures and to identify small areas of intrapelvic extension, but this involves radiation of the gonads; it is best performed following administration of a contrast agent<sup>8 35</sup>;
- magnetic resonance imaging (MRI) also delineates bony and muscular pelvic structures and can characterise the nature of the mass and detect intrapelvic extension<sup>8 36</sup>; Hata *et al* reported facilitation of antenatal diagnosis of sacrococcygeal teratoma by combined use of Doppler sonography and MRI<sup>37</sup>;
- angiography to demonstrate the main blood vessels supplying sacrococcygeal teratomas—the middle and lateral sacral and gluteal branches of the internal iliac artery and a branch from the profundus femoris artery; as hypervascularity, irregularly encased vessels, arteriovenous shunting, and serpentine dilated veins may be seen in malignant as well as in benign tumours, angiography is not conclusive in distinguishing benign from malignant disease<sup>8</sup>;
- Gross *et al* have discussed the role of myelography in evaluating the intraspinal extension of sacrococcygeal teratomas.<sup>38</sup>

### Management at delivery

The management of sacrococcygeal teratomas depends on fetal gestational age and wellbeing, the presence of associated anomalies, and tumour vascularity assessed by colour flow Doppler.<sup>18</sup> Highly vascular tumours should be delivered by caesarean section to avoid risk of haemorrhage during the second stage of

labour.<sup>18</sup> Robertson *et al* reported devascularisation of a sacrococcygeal teratoma to decrease the chance of spontaneous haemorrhage in a preterm neonate<sup>39</sup>; antenatal ultrasound had confirmed the highly vascular nature of tumour, with several large feeding vessels arising from the internal iliac arteries. Both internal iliac arteries and the small middle sacral artery were ligated, after stabilisation of respiratory status.<sup>39</sup>

Delivery should be conducted in a tertiary hospital. The size of a sacrococcygeal teratoma determines the mode of delivery. Non-vascular tumours and those less than 10 cm in diameter may be delivered vaginally.<sup>18</sup> However, Gross *et al* recommended caesarean delivery in all fetuses with sacrococcygeal teratomas of more than 5 cm diameter, to minimise the risk of rupture and haemorrhage.<sup>40</sup> Tumour size has been more than 10 cm in almost all reported cases with difficult deliveries.<sup>14 41–43</sup> Teratomas should be well protected after delivery because erosion of the surface may precipitate bleeding before surgical excision is undertaken.<sup>44 45</sup> Blood loss in large and even benign tumours has been reported to be substantial, sometimes equalling the patient's blood volume.<sup>46</sup> Most reviews report mortality ranging from around 5% to 9% following exsanguinating haemorrhage.<sup>14 34 47 48</sup>

Grosfeld *et al* reported disseminated intravascular coagulation in a neonate who suffered tumour rupture and haemorrhage during delivery.<sup>34</sup> The aetiology of such clotting abnormalities is probably complex and multifactorial.<sup>45</sup> Extensive disruption of large, defective tumour endothelium during labour and delivery could precipitate fulminant disseminated intravascular coagulation. Trauma to the sacrococcygeal teratoma during delivery may cause entry of tissue thromboplastin into the blood stream, resulting in activation of the coagulation cascade.<sup>45</sup> Good vascular access and adequate supply of blood products is thus necessary before surgery.

During surgery, precautions must be taken to prevent hypothermia, which is easily precipitated because of the large surface area and the vascularity of the tumour.<sup>44</sup> The treatment of choice for sacrococcygeal teratoma is early surgical resection with complete excision of the coccyx, because microscopic nests of neoplastic cells are commonly found in or immediately adjacent to the coccyx.<sup>5 15</sup> A recurrence rate of 37% was reported when the coccyx was not removed completely.<sup>51</sup> Patients with malignant sacrococcygeal teratomas are managed after surgery with irradiation if residual disease is present, and always with combination chemotherapy.<sup>1 15</sup>

### Postoperative complications

The main postoperative complication is wound infection because of the proximity to the anus of the surgical site and the skin flaps that may be needed.<sup>8 59</sup> Bladder dysfunction may occur.<sup>8 34 47</sup>

### Follow up/recurrence

The recurrence rate of sacrococcygeal teratomas varies between 7.5% and 22%.<sup>48, 49</sup> In contrast to previous reports,<sup>14, 50</sup> Bilik *et al* found a higher recurrence rate for primary sacrococcygeal teratomas with larger mean diameters (11.1 (3.2) cm with recurrence *v* 7.9 (4.4) cm with no recurrence (mean (SD));  $p = 0.07$ ).<sup>49</sup> They concluded that despite multiple histological sections in large primary tumours reported as benign, these large tumours may well harbour undetectable small foci of malignant endodermal sinus cells.

Serum  $\alpha$  fetoprotein in the first month of life and immunohistochemical markers are not reported to be of prognostic significance.<sup>50</sup> However, after total resection of primary sacrococcygeal teratoma together with the coccyx, a raised serum  $\alpha$  fetoprotein has been found to be a reliable marker of recurrence of poorly differentiated yolk sac tumours.<sup>51</sup> Bilik *et al* reported markedly raised serum  $\alpha$  fetoprotein in malignant recurrent compared with benign sacrococcygeal teratomas.<sup>49</sup> As none of the variables except tumour size was found to be a reliable predictor of recurrence, these investigators concluded that meticulous routine physical examination is essential every three to six months for at least the first three postoperative years to detect recurrence.<sup>49</sup> They also emphasised the importance of regular serum  $\alpha$  fetoprotein measurements, prompt radiographic investigations to detect possible recurrences (for example where there is raised  $\alpha$  fetoprotein but clinically undetectable tumour), and the need to regard every recurrence as potentially malignant.<sup>49</sup>

The potential for late occurrence of malignancy in mature sacrococcygeal teratoma is well documented.<sup>52, 53</sup> Lack *et al* reported infiltration of virtually the entire coccyx with an adenocarcinoma 40 years after the resection at two months of age of a congenital mature teratoma.<sup>52</sup> Lahdenne *et al* followed up 45 patients (aged 4–43 years, mean 21.5 years) for detection of late recurrences.<sup>53</sup> All had been operated on in infancy for benign sacrococcygeal teratomas. Three recurrences (two benign and one malignant) were diagnosed 21 to 43 years after the initial diagnosis and operative treatment; coccygectomy had not been performed as a primary procedure in patients with malignant recurrence. Rescorla *et al* reported recurrent disease in nine of 80 patients (11%) with mature teratomas, at between six and 34 months after resection.<sup>9</sup> Three had metastatic disease. Two were long term survivors after surgery alone. Six were alive at a mean follow up time of 114 months after chemotherapy (5) and pelvic irradiation (1). One patient with metastatic disease was lost to follow up. This series also included cases of immature teratoma and endodermal sinus tumour (EST). Of 24 children with immature teratoma followed up for an average of 39 months, one (4.2%) had a rising  $\alpha$  fetoprotein level 6 months after the initial procedure and required resection. The child was free of disease 33 months after the second procedure. Of the 13 neonates and infants with EST, nine under-

went early surgery with complete resection. Seven were long term survivors. Two died from non-disease-related causes. One child whose lesion was observed after birth presented with metastatic disease at 10 months of age and underwent incomplete resection at that time. She died despite adjuvant treatment. Three other patients presenting between seven and 30 months of age without evidence of metastatic disease underwent complete resection and were long term survivors.<sup>9</sup>

Schropp *et al* reported their experience with sacrococcygeal teratomas in children over four decades (1950 to 1990).<sup>54</sup> Of 73 teratomas, 57 (78%) were benign and 16 (22%) malignant. There were five recurrences in children with benign disease, only one of which did not have an initial coccygectomy. The average age at presentation of benign sacrococcygeal teratomas was 20 days *v* 468 days in those with malignant disease. Seven of the 16 children with malignant sacrococcygeal teratomas were long term survivors. The average survival was 9.3 months in the nine children who died (range 1 to 28 months). Only two of 10 cases (diagnosed between 1976 and 1991) undergoing chemotherapy and radiotherapy for malignant disease died with disease (80% survival). Three deaths occurred in those with benign sacrococcygeal teratomas (95% survival). One neonate died in the immediate postoperative period because of complications (rupture of hepatic subcapsular haematoma and disseminated intravascular coagulation with sepsis).<sup>54</sup> Maturation from malignant to benign sacrococcygeal teratoma after chemotherapy (pseudoretroconversion) has been reported.<sup>54, 55</sup>

### Sequelae of sacrococcygeal teratoma

The type of the teratoma and the surgical approach both contribute to functional sequelae. Tumours with a large presacral component may present with lower extremity weakness or paralysis, constipation, abdominal distension, and urinary tract symptoms secondary to bladder outlet obstruction.<sup>56–58</sup> Isolated cases of lower extremity weakness or paralysis and bladder dysfunction have been described postoperatively, particularly in association with malignant lesions. Many of the previously reported large series did not report long term faecal and urinary incontinence.<sup>34, 47, 56, 59–63</sup> Engelskirchen *et al* reported electromanometrically detected rectal and bladder dysfunction in 40% of children following excision of sacrococcygeal teratoma, although none had overt incontinence.<sup>63</sup> Malone *et al* reported on 27 patients followed up for a mean period of five years (range 2–12).<sup>64</sup> Eleven (41%) had some form of functional impairment. Faecal and urinary incontinence were present in nine. Two had lower extremity weakness because of sciatic nerve palsy.<sup>64</sup> Tumours with large intrapelvic extensions requiring an abdominoperitoneal approach for resection were associated with a higher incidence (67%) of functional sequelae. It was concluded that it was impossible to determine retrospectively whether dysfunction was caused by the tumour

itself or by the surgery.<sup>64</sup> Rintala *et al* evaluated quality of life and faecal continence in 26 adult patients (mean age 30 years) who had undergone surgery for benign sacrococcygeal teratoma in infancy.<sup>65</sup> Good faecal continence was reported by 88% of these patients. However, only 27% had completely normal bowel habits. Some faecal soiling was present in 27%. No correlation between the severity of anorectal malfunction and the degree of intrapelvic extension of the tumour was found. Other health problems, including urinary incontinence, were reported by 13 patients (50%). All control subjects (26 healthy patients of similar age and sex distribution) had good faecal continence; 77% had completely normal bowel habits.<sup>65</sup> Hypergonadotropic hypogonadism and sperm abnormalities have been reported in men born with benign sacrococcygeal teratomas.<sup>66</sup> Such patients may have Leydig cell dysfunction, abnormal spermatogenesis, or both. The association of these with sacrococcygeal teratomas may reflect a congenital germ cell defect.<sup>66</sup>

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