# Benign Sacrococcygeal Teratomas in Infants and Children

## A 25 Year Review

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**A review of 33 infants treated over 25 years for benign sacrococcygeal teratoma shows that this is predominantly a newborn tumour presenting mostly as an external mass and carrying an excellent prognosis provided surgical treatment is prompt and complete excision accomplished. Resectability was 100% in this series as is usually the case. This tumour must however, be differentiated from other masses presenting in the sacrococcygeal area by a careful histologic study since these other lesions generally require a different therapeutic approach.**

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THE PURPOSE of this paper is to review the records of 33 infants with benign sacrococcygeal teratomas who received treatment at the Hospital for Sick Children, Toronto between 1951 and 1976.

### Clinical Material

Twenty-five of the 33 infants were females and eight males. The diagnosis was made at birth in 32 of these babies and at six weeks in one. There was one twin and a family history of twinning in four.

Twenty-six babies were born full term and four at periods less than 38 weeks gestation. Delivery was normal in 24 and two were delivered by Caesarian section because of the sacrococcygeal mass. Birth weight was over 3 kg in 23 and less than 3 kg in seven.

Associated anomalies were an absent eye, sacral myelomeningocele, vaginal stenosis, VSD, spina bifida occulta and esophageal atresia with TEF.

Five of the tumors were small, 15 medium and 13 large. Preoperative x-rays showed calcification in 12. Five were totally cystic, three solid and 25 had both components.

The tumor was resectable in all 33 neonates through a sacrococcygeal (prone) approach. One baby also needed an abdominal exposure. The coccyx was removed along with the mass in 21 babies. There was one tumor recurrence. One newborn died of operative cardiac arrest due to anesthetic problems. Postoperative complications were wound infection (six), diarrhea (three), wound dehiscence (three) and draining sinus (two).

Twenty-four of the 33 babies were followed up to 1975: Three for less than one year, 16 for one to five years and seven longer than five years.

### Discussion

The benign sacrococcygeal tumor occurs much less frequently than other neonatal anomalies.[1,2] Its origin is controversial.[1–7] The fact that this tumor is benign

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and in the newborn contrasts with the malignant form which is almost always found in the older child. Seventy-six per cent of our infants were female and this coincides with the 80% average found in the literature.

The diagnosis of these tumors must be confirmed histologically since other masses presenting in the sacrococcygeal region may mimic them clinically. In our hospital between 1951 and 1976, the following infant sacral tumors were mistakenly diagnosed initially as benign sacrococcygeal teratomas: anterior meningocele, extraspinal glioma, presacral epidermoid tumor, neuroectodermal hematoma, CNS malformation and rectal duplication. Other conditions that may be similarly confused include: abscess, chordoma, neurofibroma, ganglioneuroma, ependymoma and cystic hemangioma.[2]

Although the incidence of twinning in these families appears to be small in our series, there was no mention of this fact being sought after in the majority of records. It may help in making the clinical diagnosis. It is enticing to speculate that a sacrococcygeal teratoma is one end of a spectrum that stretches from the sacrococcygeal teratoma through the neonate with a double pelvis and four limbs, fetus-in-fetu, siamese twins to the normal set of twins.

The presence of a sacrococcygeal mass does not appear to affect gestation, although delivery may be made difficult if the mass is large enough. This was the case in two newborns in our series who required Caesarian section for delivery on account of the large mass. In such instances, the tumor is usually the size of a baby's head.

Congenital anomalies are more frequent in infants with benign sacrococcygeal teratoma; no particular anomaly seems to be more frequently found than others. Altman reported an 18% incidence of newborn anomalies in these babies compared with 2.5% in the unselected population.[8] Cross[5] (5%) and Carvey[9] (26%) both found higher numbers of anomalies in their series. The incidence in our group was 18%.

Altman et al. classified these tumors into:

Type I: (totally external);
Type II: (almost completely external);
Type III: (almost completely internal) and
Type IV: (totally internal).

In our series there were 19 Type I, 10 Type II, three Type III and one Type IV. As with most other series, 87% of the newborns in our group had Type I and Type II tumors. This fact is obviously the reason why the diagnosis was made at birth or shortly thereafter. The larger the portion of the tumor inside the pelvis, the later the diagnosis is made. The only infant who had a benign tumor diagnosed outside the newborn period had a Type IV tumor. This baby was indeed very fortunate that its tumor was removed not much later than six weeks of age, because such delay results in malignant transformation with a completely different outlook. When this transformation exactly takes place is as yet an unanswered question. Most Type IV tumors are diagnosed at a much later age by which time malignant changes have taken place. Hence the importance of early diagnosis and treatment of any sacrococcygeal mass no matter how small or apparently insignificant. There is no rationale in watching and waiting as it will not go away.

As with most benign sacrococcygeal tumors, the majority seen in our series were of the mixed cystic and solid variety and quite large mainly due to the cystic component. This clear fluid is usually CSF not connected with the baby's own circulating CSF, but coming from the choroid plexus in the tumor mass. Although most newborn sacrococcygeal teratomas are for the most part large, cystic and benign, size or consistency does not have any clinical value in determining the prognosis. Furthermore, the presence or absence of radiologic calcification in the mass bears no significance as to whether it is a teratomatous mass or not, or whether the tumor is benign or malignant.[5,9,10] Only 36% of our neonates had radiologic calcification in their tumor.

The only absolute way to diagnose and treat the newborn with a sacrococcygeal mass is to completely excise it—the sooner the better! Since this tumor arises in the coccyx, excision of the coccyx along with the mass is necessary in order to accomplish total excision.[5,7] Twenty-one of our 33 babies were so treated. It is not intended to discuss the various surgical approaches of managing this benign tumor in this report, since others have adequately dealt with this aspect.[2,5,11,12] The only tumor recurrence in our series occurred in one of the 12 infants who did not have their coccyx removed.

Wound infection was the most frequent postoperative complication in our group (18%) as well as in others. This is hardly surprising considering that all but one of the babies in our series had their tumor removed by the perineal (prone) approach. The small incidence of temporary diarrhea (9%) as well as the occasional bladder dysfunction are also not beyond belief considering the major retrorectal and lower retroperitoneal dissection required for many of these tumors. Sometimes the retroperitoneal extension, while not always felt by abdominal examination (even under general anesthesia), can be quite large making

the external component of the mass resemble the tip of an iceberg. These few complications did not appear to pose much of a problem. Antibiotics were not used on a routine basis either prophylactically or therapeutically in this series.

The only death was related to anesthesia and occurred on the operating table. Although the follow-up of the babies in this series was only 80%, those that were followed remained well with normal perineal function in all respects.

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