

# Early Outcomes of the Surgical Treatment of Sacrococcygeal Tumors in a Tertiary Level Government Hospital

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## ABSTRACT

**Objectives.** Sacrococcygeal teratomas (SCT) are the most common extragonadal tumors of early childhood. Their clinical characteristics and outcomes of patients with sacrococcygeal tumors who underwent excision in the Philippines has never been described, while numerous retrospective studies have been conducted in other countries.

**Methods.** This was a retrospective, descriptive study over a four-year period (December 2014 to November 2018). The study described the patients' demographic data, manner of delivery, clinical presentation, prenatal diagnosis of tumor, Altman classification, and alpha fetoprotein levels. These information were obtained from the medical records of the patients. Additional data from the operative technique include the surgical approach, size of the mass, and gross involvement of adjacent structures and the final histopathologic results. Outcomes include the 30-day mortality and morbidity, and tumor recurrence.

**Results.** A total of 29 patients were included in the study with 22 females (75.86%) and seven males (24.14%). Twenty-five out of the 29 (86.21%) had a sacral or gluteal mass at birth while other presenting factors include a palpable abdominal mass (1), constipation (1), difficulty in urination (1), and an elevated AFP in one postoperative patient. Even if 27 out of the 29 patients underwent a maternal ultrasound, only three patients (10.34%) had a correct ultrasound interpretation of sacrococcygeal teratoma. Age at presentation was problematic, with 12 presenting at greater than one year of age while 10 were brought for consultation at greater than one month old. Only seven presented at the neonatal period. CT scan was the most common imaging tool utilized (37.93%), followed by ultrasound (27.59%). AFP was elevated in ten patients (34.48%). Six of the patients with elevated AFP had mature teratoma, two had yolk sac tumor, one had fibroepithelial polyp, and one was post chemotherapy but had mature teratoma based on the final histopathology report. Fifteen out of the 29 patients had Altman type I tumors (51.72%), seven (24.14%) had type II tumors, six (20.69%) had type III tumors, and only one patient had type IV tumor. Sacral approach in the excision of the sacrococcygeal tumor was performed in 25 patients (86.21%). There was no reported perioperative mortality for patients who underwent surgery for SCT during the study period. Twelve out of the 29 had postop morbidities, three with surgical site infection and three with rectal or vaginal perforation. Five patients had tumor recurrence occurring from two months to three years postoperatively.



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Conclusion. Early detection of sacrococcygeal teratomas even in the prenatal period is the norm in certain areas of the world, but in our country, prenatal detection is still a challenge. Even if the majority of the patients presented with a gluteal mass at birth, less than a third were brought to our tertiary government hospital in neonatal life. The sacral approach for SCT excision was employed

**Conclusion.** Early detection of sacrococcygeal teratomas even in the prenatal period is the norm in certain areas of the world, but in our country, prenatal detection is still a challenge. Even if the majority of the patients presented with a gluteal mass at birth, less than a third were brought to our tertiary government hospital in neonatal life. The sacral approach for SCT excision was employed

for the great majority of our patients, but due to the advanced age at diagnosis and locally advanced disease, morbidities occurred in about a third of the patients. Therefore, early detection prenatally and early referral to a pediatric surgical center should be achievable goals for physicians dealing with these patients.

*Keywords: sacrococcygeal teratoma, extragonadal germ cell tumor, alpha feto protein, mature teratoma, Philippines*

## INTRODUCTION

Sacrococcygeal teratoma (SCT) is the most common extra gonadal tumor of infancy and early childhood accounting for 40% to 70% of teratomas.<sup>1,2</sup> Several retrospective studies on the clinical profiles and outcomes of SCTs have been conducted in Europe, United States, India, Australia, and Asia. In the Philippine General Hospital (PGH), a tertiary level government hospital, surgical treatment for SCT has been performed on 92 patients for the last 10 years. However, there is still no study describing the demographics, clinical characteristics, and immediate perioperative outcomes of patients who underwent excision of SCT in the Philippines.

In previous studies, factors affecting outcomes of patients were identified. Among the variables examined were age at presentation and diagnosis, histopathology, and completeness of excision. As high as 90% of SCTs are now detected antenatally compared to 11% in the 1980's.<sup>3-5</sup> Early diagnosis enables counselling of parents regarding recommended manner of delivery and earlier referral to centers with a pediatric surgery section.<sup>1,3,6</sup> Patients with SCT more commonly present as a palpable external mass and less frequently with compression symptoms like urinary incontinence, constipation, or a palpable lower abdominal mass for entirely pelvic masses.<sup>1,4,7</sup> In addition to age at diagnosis, the histopathology also had a significant impact on the prognosis. Immature and malignant histology were associated with recurrence, but the most important predictor of recurrence after SCT excision is the presence of residual tumor.<sup>8,9</sup>

The diagnosis and treatment of SCT has been standardized in most institutions. Diagnostic tests for SCT include abdominopelvic ultrasound, serum alpha feto protein (AFP) and xrays.<sup>3,10</sup> A contrast enhanced abdominopelvic CT scan is warranted in some cases for better preoperative planning, because of the ability to describe the tumor's location under the Altman classification. The Altman classification of SCT subdivides tumors based on the anatomic location and pelvic extension: Type I describes tumors that are predominantly external with minimal pre-sacral component; Type II are tumors that present externally but with significant intrapelvic extension; masses that are predominantly pelvic and extending into the abdomen but are still appreciable externally are classified as Type III; and Type IV masses are pre-sacral with no external component.<sup>11</sup>

Early and complete surgical excision remains to be the mainstay of treatment with adjuvant chemotherapy indicated in certain patients resulting in an overall survival of greater than 75% with multimodal therapy.<sup>2,12,13</sup> Through the years, there has been significant improvement in patient outcomes. In one series done by Mahour et al., in 1975, all patients with local recurrence and/or metastasis died regardless of the therapy given. At present, SCT has an overall survival of 90% after early surgery in most centers but this drops to 50% for recurrent cases.<sup>7</sup>

Aside from recurrence and survival rates, perioperative and long-term complications are presented by most authors. Massive intraoperative hemorrhage, surgical site infection, urinary retention, urinary tract infection, and fecal soiling are among the immediate complications noted with wound infection as being the most common.<sup>3,4,9,10</sup>

There is currently no published literature on sacrococcygeal tumors in the Philippines. The objective of the study is to look at the clinical experience with sacrococcygeal tumors in the PGH. The specific objectives aim to describe the demographic profile of patients, including clinical characteristics, surgical intervention, morbidity and mortality of this group of patients who underwent excision at the PGH. The results of this study will be used in finalizing the Sacrococcygeal Tumor Treatment Protocol, which will be conducted by the pediatric tumor board composed of the Division of Pediatric Surgery, Departments of Pediatrics (Hematology-Oncology), Radiology, and Pathology.

## METHODS

This is a retrospective descriptive study, a review of in-patient medical records. The charts of patients below 19 years old who underwent excision for SCT in the PGH from December 2014 to November 2018 were made available from the medical records section. Originally 40 patients were to be included, but only 29 patients' records fit the inclusion criteria.

The exclusion criteria of the study were the following:

1. Patients above 19 years old
2. Patients who did not undergo excision at the Philippine General Hospital.
3. Patients who underwent surgery outside and transferred to our institution for another surgical intervention were also not included
4. Patients whose charts were unavailable from the record section.

The list of in-patient charts included in the study was based on the operations performed by the fellows and consultants under the Division of Pediatric Surgery logged as excision of sacrococcygeal teratoma performed from December 2014 to November 2018 at the PGH. The demographic data, manner of delivery, clinical presentation, prenatal diagnosis of tumor, alpha fetoprotein levels, and

Altman classification were obtained from the medical records of the patients. This patient list was assigned codes to maintain patient privacy

The operative technique record was reviewed to collect data on the surgical approach, size of the mass, and gross involvement of adjacent structures. This information combined with the final histopathologic results that was obtained from the Department of Pathology was used to determine the completeness of resection. Lastly, data on the primary outcomes which includes mortality and morbidity in the immediate perioperative period was also based on the patients' charts while admitted. A detailed description of the demographics and clinical characteristics of the charts of patients meeting the inclusion criteria was performed. The percentages under each category listed above were computed.

It must be emphasized that this study underwent ethics approval by the research board of the hospital.

## RESULTS

From December 2014 to November 2018, a total of 40 pediatric patients underwent surgery for a sacrococcygeal mass at our institution. Of the total 40 patients, one patient had surgical excision in another institution while ten patients' charts could not be retrieved. Thus, only 29 patients met the inclusion criteria. Table 1 shows the clinical manifestations of these 29 patients who met the inclusion criteria.

There was a female predominance (75.86%) noted among those included in the study. Eighty-six percent of them presented with a gluteal or sacral mass at birth. Other presenting factors were palpable abdominal mass (1), constipation (1), difficulty in urination (1) and an elevated AFP in one postoperative patient.

These set of sacrococcygeal teratoma patients were occasionally diagnosed prenatally as seen in Table 2 via prenatal ultrasound. In 27 of the mothers (93.10%), a prenatal ultrasound was performed. However, it was not indicated in the charts at what age of gestation it was done. The maternal ultrasound was normal in 21 mothers (72.41%). Among those with abnormal findings, only three (20.70%) were interpreted as sacrococcygeal mass. One ultrasound was read as a lumbosacral meningocele, another was interpreted as an enlarged scrotum and there was one patient diagnosed to have a parasitic twin which eventually turned out to be consistent with the final histopathologic result.

Twenty-one out of the 29 patients (72.41%) were delivered vaginally while the remaining eight (27.59%) were delivered via caesarian section. The reason for the caesarian section was indicated in two of the eight patients. One was due to a twin gestation and the other was due to failure of descent.

Only three of the 29 (10.34%) had associated anomalies present. One had patent ductus arteriosus, another had an imperforate anus with perineal fistula, and one was diagnosed with G6PD deficiency.

**Table 1.** Clinical Manifestations

Signs and Symptoms	Number of Patients	Percentage (%)
<i>Sacrococcygeal mass</i>	25	86.21
<i>Lower abdominal mass</i>	1	3.45
<i>Urinary tract obstruction</i>	1	3.45
<i>Gastrointestinal obstruction</i>	1	3.45
<i>Others: elevated AFP</i>	1	3.45
<b>Total</b>	29	100

**Table 2.** Antenatal Diagnosis of SCT

Prenatal UTZ Details	Number of Patients	Percentage (%)
<i>SCT</i>	3	10.34
<i>Enlarged scrotum</i>	1	3.45
<i>Parasitic twin</i>	1	3.45
<i>Lumbosacral meningocele</i>	1	3.45
<i>Not seen on prenatal UTZ</i>	21	72.41
<i>Not done</i>	2	6.90
<b>Total</b>	29	100

**Table 3.** Age at Diagnosis

Age	Number of Patients		Total	Percentage (%)
	M	F		
<i>≤1 month</i>	1	6	7	24.14
<i>&gt;1 month to 1 year</i>	4	6	10	34.48
<i>&gt;1 year</i>	2	10	12	41.38
<b>Total</b>	7	22	29	100

**Table 4.** Preoperative Imaging

Imaging Study	Number of Patients	Percentage (%)
<i>Ultrasound</i>	8	27.59
<i>CT scan</i>	11	37.93
<i>MRI</i>	2	6.90
<i>None</i>	8	27.59
<b>Total</b>	29	100

Despite an obvious tumor at birth, Table 3 shows the age of the patients upon correct diagnosis of a sacrococcygeal teratoma. Seven of the 29 (24.14%) patients were diagnosed at less than one month of age. Ten patients (34.48%) were diagnosed at more than one month but less than one year, while twelve (41.38%) of them were operated on at more than one year of age.

Preoperative imaging was not performed on all patients as shown in Table 4. CT scan was the most common imaging tool utilized in 11 (37.93%) out of the 29 patients followed by ultrasound (27.59%) and two (6.9%) even underwent MRI.

Aside from preoperative imaging studies, serum AFP was also measured. AFP was elevated in ten patients (34.48%). Six of the patients with elevated AFP had mature teratoma, two had yolk sac tumor, one had fibroepithelial polyp, and

one was post chemotherapy but had mature teratoma based on final histopathology report.

Based on the Altman classification, fifteen out of the 29 of the patients had type I tumors (51.72%), seven (24.14%) had type II tumors, six (20.69%) had type III tumors, and only one patient had type IV tumor.

All twenty-nine patients underwent excision of the tumor as shown in Table 5. Sacral approach in the excision of the sacrococcygeal tumor was performed in 25 (86.21%) out of the 29 patients while an abdominosacral approach was done on three patients (10.34%). A purely abdominal approach was performed on one patient with recurrence at the pelvic and presacral area.

Four patients had incomplete excision of the mass. It was indicated in the operative technique of one patient that there was partial excision of the presacral recurrence but the final histopathology showed mature bone and cartilage only. In another patient, tumor spillage was reported with gross residual tumor adherent to the pelvic sidewalls. This case was signed out as rhabdomyosarcoma. Two patients had incomplete resection based on microscopic findings of inked tumor margins and cauterized tumor margins.

**Table 5. Surgical Approach**

Surgical Approach	Number of Patients	Percentage (%)
Transsacral	25	86.21
Abdominal	1	3.45
Abdominosacral	3	10.34
<b>Total</b>	<b>29</b>	<b>100</b>

**Table 6. Histopathologic Result**

Histopathology	Number of Patients	Percentage (%)
Mature	20	68.97
Immature	1	3.45
Malignant	1 - rhabdomyosarcoma 3 - yolk sac tumor foci	3.45 10.34
Others	1 - lipoma 1 - fibroepithelial polyp 1 - mature bone and cartilage 1 - parasitic twin	3.45 3.45 3.45 3.45
<b>Total</b>	<b>29</b>	<b>100</b>

**Table 7. Morbidity**

Morbidity	Number of Patients	Percentage (%)
Hemorrhage		
SSSI	3	10.34
Urinary retention		
Others	1 - wound dehiscence 5 - recurrence 3 - rectal perforation/ vaginal perforation	3.45 17.24 10.34
<b>Total</b>	<b>12</b>	<b>41.38</b>

On the review of the final histopathologic reports in Table 6, 20 out of the 29 patients had mature teratoma, and four had malignant tumors (3 – yolk sac tumor foci, 1 – rhabdomyosarcoma). The other histopathologic results were fibroepithelial polyp, lipoma, parasitic twin, and mature bone and cartilage.

There was no reported perioperative mortality for patients who underwent surgery for SCT during the study period. The morbidities reported are listed in Table 7. Wound infection was noted in four patients, one of which had wound dehiscence. Three patients had rectal perforation; one patient also had a concomitant vaginal perforation. Two of these were managed with a colostomy. The other one was repaired but one week after the surgery, there was dehiscence of the repair hence a colonic diversion was also performed. Five patients had tumor recurrence occurring from two months to three years postoperatively. Tumor recurrences were initially monitored postoperatively with alpha feto protein levels or physical examinations at the outpatient clinics. If abnormalities were detected, the patients then underwent radiologic imaging to document recurrence.

## DISCUSSION

Sacrococcygeal teratomas are one most common tumors affecting neonates and younger children.<sup>1,2</sup> They occur in approximately 1 in 30 000 to 40 000 live births. In our institution, 40 patients underwent surgery for SCT in four years, however only 29 were included in this study. A female predominance was noted, with a female to male ratio of 3:1 which was consistent with the ratio observed in previous studies.<sup>3,4,14,15</sup>

SCTs are usually diagnosed within the first month of life but with the improvement in imaging studies and widespread use of routine obstetric ultrasound, more SCTs are being diagnosed antenatally. As high as 90% of SCTs are now detected antenatally.<sup>3,5,6</sup> In a study performed at a tertiary hospital in Udaipur, India, a prenatal diagnosis rate of only 22% was described while in another retrospective study by Sayed et al., none of the patients with SCT were detected on prenatal ultrasound.<sup>2,3</sup> They attributed these numbers to the greater percentage of patients with tumors which were predominantly intraabdominal. In addition, they have also considered the impact of lack of awareness of families in their regions regarding antenatal care and screening due to lower educational attainment.<sup>2</sup> Prenatal ultrasound was performed in 27 of the 29 patients included in our study, but only three were correctly diagnosed. Contrary to the aforementioned reasons for low detection rate and screening, majority of the patients with normal prenatal ultrasound had predominantly external tumors and only two patients had no prenatal imaging done. Further investigation should be done regarding the age of gestation when the ultrasound was performed as well as whether a radiologist or obstetric

sonologist did the imaging to identify factors that may help in improving prenatal detection rates.

Early diagnosis enables counselling of parents regarding recommended manner of delivery, possible fetal intervention if warranted, and earlier referral to centers with a pediatric surgical unit because this contributed to better outcomes.<sup>6</sup> Caesarian delivery is recommended for patients with tumor size larger than 5 cm to avoid dystocia and tumor rupture.<sup>1,3</sup> Majority of the patients in this study (72.41%) were delivered vaginally while 27.59% were delivered via caesarian section. It would have been helpful if the reason for a caesarian delivery was available in their medical records.

As mentioned above, majority of patients are diagnosed within the first month of life. In one study, SCT was diagnosed within the neonatal period in 69.44% while 5% presented after one year of life. Age at presentation is a very important prognostic factor in SCTs because late diagnosis translates to a higher incidence of malignancy with metastasis.<sup>3,14</sup> Twenty-four out of the 29 patients presented with an external mass at birth. Only one patient presented after one year of age with obstructive urinary symptoms. Out of the 29 patients, 24.14% underwent surgery at less than one month of age and 41.38% of them were operated on at more than one year of age. The delay in the timing of consult and surgery may possibly be related to the lower socio-economic status, lack of awareness, and the scarcity of tertiary hospitals in the provinces as mentioned in a study by Kundal et al.<sup>10</sup>

The diagnostic evaluation of SCT involves imaging studies and serum tumor markers. CT scan was the most common imaging tool utilized (37.93%) in our study, followed by ultrasound (27.59%) and only 6.9% patients underwent MRI. AFP was also measured and it was elevated in 34.48% of patients. It is important to note that an elevated AFP was observed in benign and malignant cases. Surveillance using serum AFP for three years after excision is recommended. Unfortunately, a number of our patients are lost to follow-up when they go back to the provinces.

With the use of the aforementioned imaging studies, the Altman classification of SCT could be determined. It subdivides tumors based on anatomic location and pelvic extension: Type I describes tumors that are predominantly external with minimal pre-sacral component; Type II are tumors that present externally but with significant intrapelvic extension; masses that are predominantly pelvic and extending into the abdomen but are still appreciable externally are classified as Type III; Type IV masses are pre-sacral with no external component.<sup>2,11</sup> In three studies, it was noted that Type I was the most common type occurring in 36-41% of patients followed by Type II, with Type IV being the least common.<sup>6,8,10</sup> This distribution was also observed in our study where 51.72% of the patients had type I tumors. The Altman classification of the tumor usually influences the surgical approach employed. The sacral approach was performed in 86.21% of patients while an abdominosacral approach was done on three patients.

Aside from the surgical approach, the completeness of the excision was also assessed in this study based on the operative findings and histopathologic reports. Four out of the 29 patients had incomplete excision of the mass. Two had gross residual tumor while the other two had inked and cauterized tumor margins. One of the patients with partial excision of the presacral mass had no evidence of tumor based on the final histopathologic report. According to Yao et al., the histopathology showed mature teratoma in 69.2% of patients, immature teratoma in 5.6%, and malignant germ cell tumor in 15.9%.<sup>4</sup> Immature and malignant histology were associated with recurrence, with higher incidence in patients with immature teratoma.<sup>9</sup> The higher recurrence rate observed among patients with immature histology was attributed to the adjuvant chemotherapy received in the malignant group.<sup>2,14,15</sup> The recurrence rate based on histology are as follows: 0% to 26% for mature, 12% to 55% for immature, and 0% to 36% for malignant tumors.<sup>4,15</sup> But the most important predictor of recurrence after SCT excision is the presence of incomplete resection.<sup>9</sup> Patients who underwent incomplete resection had a recurrence rate of 69.2% compared to 8.33% among those who had complete resection. Recurrences usually occurred within three years after the surgery. This has become the basis of the recommendation in most institutions that a minimum of a three-year follow-up should be done to catch the recurrences earlier.<sup>2,4,14</sup>

There were five recurrences recorded during the study period. These were detected from two months to three years after the initial surgery. Four of them had complete excision at the initial surgery. Based on histopathology, two had yolk sac tumor foci, one had mature teratoma while the result of the other patient was not available. One patient had incomplete excision because the patient had bradycardia intraoperatively due to an anesthetic complication causing the surgical team to end the operation immediately.

Early and complete surgical excision remains to be the mainstay of treatment with adjuvant chemotherapy indicated in certain patients.<sup>2,12</sup> SCT has an overall survival of 90% after early surgery but this drops to 50% for recurrent cases. The overall recurrence of SCT has been observed to be between 2% to 35% in previous reports.<sup>4</sup> In addition to immature and malignant histology, and incomplete excision, De Backer et al. also mentioned that failure to detect the presence of malignant foci within the mature tumor may contribute to recurrence.<sup>15</sup>

Aside from recurrence and survival rates, perioperative and long-term complications are presented by most authors. Massive intraoperative hemorrhage, surgical site infection, urinary retention, urinary tract infection, and fecal soiling are among the immediate complications noted with wound infection as being the most common. This has been attributed to the proximity of the wound to the anal opening.<sup>3,4,9,10</sup>

Three patients developed wound infection and another patient had wound dehiscence, the latter was referred to the plastics service for co-management. Aside from wound

infection, three patients had rectal perforation, one was noted intraoperatively with associated vaginal perforation and was addressed with colonic diversion. The other perforation was primarily repaired but leak was noted one week after the surgery. The last patient with perforation was noted 12 days postoperatively and was also managed with a colostomy.

Other complications include long-term functional deficits like need for scar revision, fecal, and urinary incontinence. However, due to lack of a good follow-up database and tools to assess the said complications, these were not included in this study.

The limitations of this study include the fact that 40 patients had sacrococcygeal teratoma during the specified time period but only 29 patients were included. Unsuccessful chart retrieval of ten patients might shed more light on sacrococcygeal tumors seen at the PGH. Due to small sample size of 29 patients, generalizations cannot be made about SCT as a whole.

## CONCLUSION

We reviewed a total of 29 charts of patients who underwent excision of SCT for a period of four years. Most of the results based on the parameters measured were consistent with international studies.

However, we noted a low prenatal ultrasound detection rate. Coordination with the obstetric and radiologic societies for the improvement and standardization of prenatal ultrasound may be done to address this problem.

Also, despite presentation at birth, most patients were diagnosed and operated on beyond one month of age. Early diagnosis, complete surgical excision, a multidisciplinary management, and long-term follow-up are key in the management of SCT. Good parental counselling and availability of subspecialty clinics in the provinces or endorsement of patients to pediatricians in their localities may help in ensuring regular follow-up.

## Recommendations

The creation of a Sacrococcygeal Tumor Treatment Protocol will greatly enhance data collection, including the long-term outcomes like the sequelae of SCT surgery on bladder and bowel function, and more importantly, tumor recurrence. This will give a clearer picture of important aspects of patient outcome in the PGH.

## Statement of Authorship

Both authors certified fulfillment of ICMJE authorship criteria.

## Author Disclosure

Both authors declared no conflicts of interest.

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