# Optimal Surveillance for Identification of Recurrence in Patients with Sacrococcygeal Teratomas

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## Background

- Sacrococcygeal teratomas (SCT), although rare, are the most common germ cell tumor of infancy/early childhood (see **Figure 1**).
- They are noted in approximately 1 in every 35,000 to 40,000 live births[1].
- Although there is an excellent prognosis for patients with this tumor, prediction and management of recurrence has been a challenge[1-4].
- There is inconsistency in the reported recurrence rate of these tumors, ranging from zero to over thirty percent[1].
- This is likely due to the limited number of studies reporting recurrence rates in large cohorts.
- The primary goal of this study is to evaluate the amount of imaging, tumor marker testing, and clinic visits done for patients with SCTs post-resection to see if they assist in identifying a recurrence, which would further assist in a more systematic surveillance process for these patients.

## Methods

- **Design**: retrospective cohort study.
- Data will be collected from multiple sites, as this study is part of the Midwest Pediatric Surgery Consortium (MWPSC), with the hope that each site will be able to identify 20-50 patients.
- At Cincinnati Children's (CCHMC), patients medical record numbers were obtained, guided by ICD9 and ICD10 codes.
- **Exclusion criteria**: patients > 18 years of age and patients without sacrococcygeal teratomas.
- **Inclusion criteria**: patients < 18 years of age with a diagnosed and resected, both pre and postnatally, sacrococcygeal teratoma.
- Data collected included baseline patient characteristics, operative characteristics, and outcome variables (e.g. amount of recurrent SCTs).
- **42 patients** were enrolled from CCHMC.
- Full statistical analyses will be obtained upon data being collected from all MWSPC sites.

## Results

- Total cohort = 42 patients
- **78.5%** female.
- **Figure 1** demonstrates the time of diagnosis of sacrococcygeal teratomas for patients in this study.
- **66.7%** had at least 1 prenatal imaging study for their SCT.
- **14.2%** had a fetal debulking procedure.
- Median age at the time of post natal resection was **3.5 days** (0 days-20 years).
- **90.2%** of patients had a coccygectomy.
- **Figure 2** demonstrates the distribution of different teratoma pathologies, with the most common being immature teratoma.
- Median length of stay post resection was **20 days** (0-127 days).
- The median number of **surveillance alpha-fetoprotein and beta HCG labs** drawn were **3.5 and 0**, respectively.
- The median number of **surveillance imaging studies** done for each patient was **2**.
- The median number **of follow up clinic visits** with pediatric surgery was **3** visits.
- The median length of follow up was **2.41 years** (0 days – 14.62 years).
- **4.8%** (n=2) died from their SCT.
- **2.4%** (n=1) patient had a recurrence of their SCT.

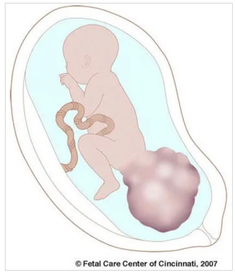


**Figure 1**

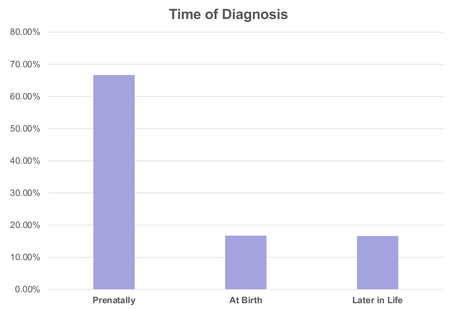


**Figure 2**

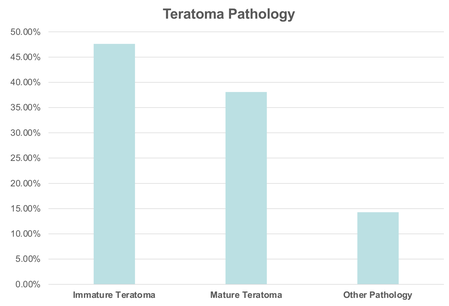


**Figure 3**

## Discussion

- A limitation of this study is its small sample size in addition to being an entirely retrospective review.
- It is known that coccygectomy is critical to preventing recurrence and was thought to be concerning that in this study that not all patients had a coccygectomy.
- The range of follow up length was also widely varied, representing the inconsistency with which these patients have been followed.
- The extremely low recurrence rate is a promising factor, although limited in this study, that could assist in the development of a protocol that is less burdensome on patients, families, and providers.

## Discussion Cont'd

- The widely varied number of imaging studies, labs, and clinic visits could also be brought into a narrower range if the low recurrence rate seen at CCHMC is true at other institutions as well.

## Conclusions

- Follow up is inconsistent across patients with sacrococcygeal teratomas at CCHMC.
- Patients are not being put on protocol for SCT management, depending on which department care was initiated through.
- This data demonstrates the need for further standardization of SCT follow up care and shows that a more conservative approach to follow up could be developed seeing that the recurrence rate is likely low.
- More conclusive results will come with the input of data from other MWSPC sites.

## References

1. Yao W, Li K, Zheng S, Dong K, Xiao X. Analysis of recurrence risks for sacrococcygeal teratoma in children. *Journal of Pediatric Surgery.* 2014;49(12):1839-1842.
2. Derikx JP, De Backer A, van de Schoot L, et al. Factors associated with recurrence and metastasis in sacrococcygeal teratoma. *Br J Surg.* 2006;93(12):1543-1548.
3. Padilla BE, Vu L, Lee H, et al. Sacrococcygeal teratoma: late recurrence warrants long-term surveillance. *Pediatric Surgery International.* 2017;33(11):1189-1194.
4. Wang Y, Wu Y, Wang L, Yuan X, Jiang M, Li Y. Analysis of Recurrent Sacrococcygeal Teratoma in Children: Clinical Features, Relapse Risks, and Anorectal Functional Sequelae. *Med Sci Monit.* 2017;23:17-23.

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