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Presacral masses and sacrococcygeal teratomas in patients with and without anorectal malformations: A single institution comparative study☆

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ABSTRACT

Background: Despite variability at presentation, presacral masses in patients with and without anorectal malformations (ARM) appear histologically similar. The purpose of this study was to identify differences in oncologic outcomes between these two groups.
Methods: A retrospective review was performed utilizing our institutional cancer and colorectal and pelvic reconstruction databases for patients with presacral masses and sacrococcygeal teratomas between 1990 and 2017. Data captured included age at surgical resection, type of ARM, tumor location within the pelvis, tumor histopathology, tumor size, adjuvant chemotherapy, recurrence, and follow-up.
Results: Forty-six patients comprised our cohort, of whom 12 had an ARM. The median age was older at resection for those with an ARM (1.4 years; range 1 day to 29.4 years) compared to those without an ARM (9 days; range 0 days to 6.9 years) ($p = 0.01$). The mean tumor size was 2.5 cm in patients with an ARM compared to 6.0 cm in patients without an ARM ($p = 0.036$). All patients with ARM had exclusively intrapelvic tumors, and histopathology included mature teratoma (8), yolk sac tumor (1), lipoma (1), and unknown (2). Tumor location for patients with sacral and presacral masses without ARM included exclusively extrapelvic (10), primarily extrapelvic with large intrapelvic component (7), primarily intrapelvic with extrapelvic component (1), exclusively intrapelvic (8), and unknown (8). Histopathology for patients with presacral masses without ARM included mature teratoma (20), immature teratoma (7), yolk sac tumor (3), ganglioneuroma (1), neuroblastoma (1), benign epithelial cyst (1), and unknown (1). Tumor recurrence rate was similar between patients with ARM ($n = 3$, 25%) and those without an ARM ($n = 5$, 15%) ($p = 0.41$). The 5-year event free survival was 65% (95% CI: 25%–87%) in the group with ARM and 81% (95% CI: 60%–92%) in the group without ARM ($p = 0.44$).
Conclusion: Sacral and presacral masses in patients with ARM are resected at a later age and are more likely to be intrapelvic. They appear histologically similar and have similar rates of recurrence and malignancy when compared to patients without ARM.
Level of Evidence: III
Type of Study: Retrospective comparative study.

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Tumors of the sacral and presacral space are rare in the pediatric population, occurring with an incidence of approximately 1 in 40,000 [1]. The most common of these lesions are histopathologically benign sacrococcygeal teratomas (SCT), although malignant tumors such as yolk sac tumors may arise in this location as well. The presacral space marks the site of fusion of all germinal layers in the developing fetus, predisposing the child to a wide histologic variety of tumors [2]. Tumors in this space are classified anatomically based on the relative intra- or extrapelvic location of the lesion as described by Altman et al. (Fig. 1) [3].

A subset of patients with presacral masses are found to have associated congenital anomalies. In 1981, Currarino et al. described the relationship of presacral masses, anorectal malformations, and sacral deformities, although the presentation is heterogeneous and all three features are not required for the syndrome [4–6]. The Currarino syndrome as it is known today is an autosomal dominant disorder [7]

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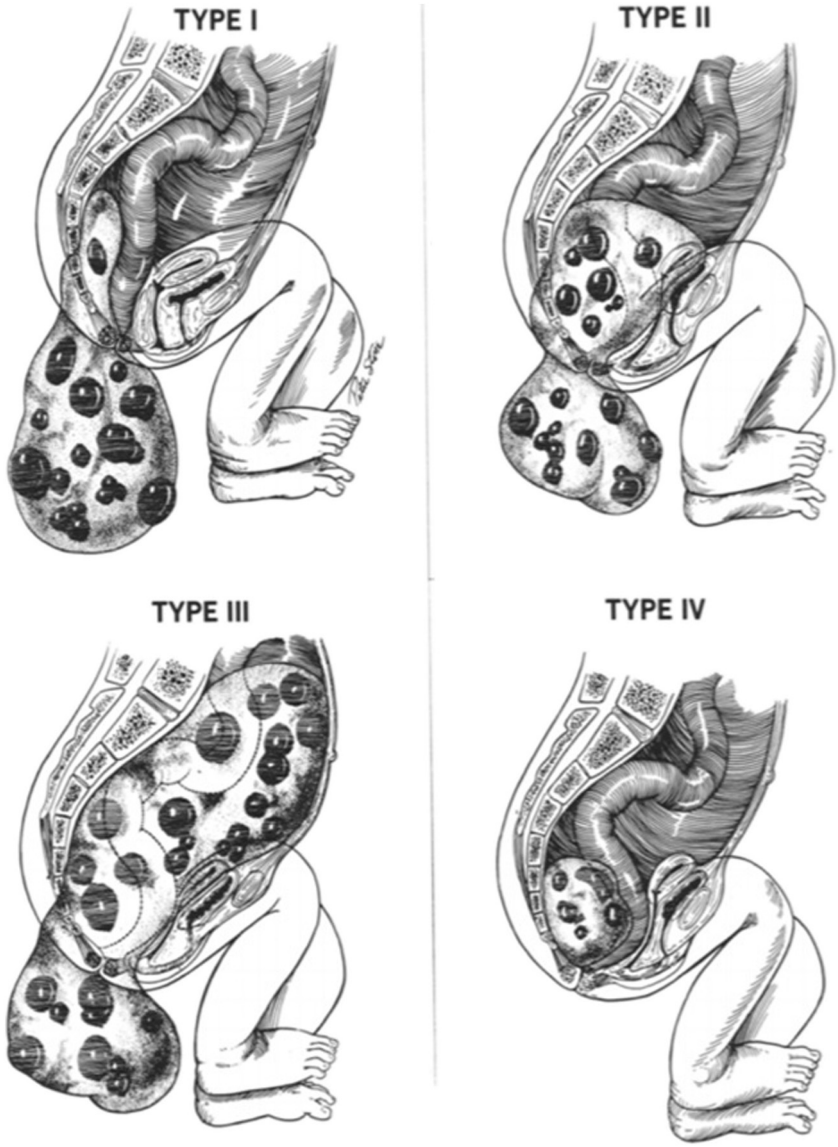

Fig. 1. The Altman classification of sacrococcygeal teratomas [1]. Reproduced with permission.

with a variable phenotypic expression, and screening of first degree relatives is to be considered. Most children are diagnosed early in infancy, although older children may not be diagnosed until after an evaluation for constipation reveals a narrowed anal canal or a presacral mass [5]. When correlated inversely, the incidence of anorectal anomalies in patients with a presacral mass has been reported at approximately 3%.

While the oncologic outcomes in pediatric patients with sacral and presacral masses have been reported previously [8–10], it is not well known whether such lesions in patients with ARM portend a worse prognosis than for patients without ARMs. The objective of this study was to evaluate our institutional experience with presacral masses in patients with and without ARMs to compare the oncologic outcomes of both groups.

1. Methods

1.1. Retrospective review

We performed a retrospective review utilizing our institutional cancer and colorectal and pelvic reconstruction databases for all patients with sacrococcygeal teratoma or tumors arising from the presacral space treated between 1990 and 2017. Patients with pelvic tumors arising from the paraspinal region, such as meningoceles, were excluded. Additionally, patients with incomplete records were excluded. This study was approved by the Institutional Review Board at Nationwide Children's Hospital (IRB17–00157).

Data captured included gender, age at surgical resection, location of the tumor within the pelvis, histopathology, chemotherapy received, duration of follow-up, time to tumor recurrence, and malignant degeneration. The Altman classification (Fig. 1) was used to describe the location of sacrococcygeal teratomas. For those patients with an ARM, the anatomic subtype was recorded. Operative reports were reviewed to determine the procedure performed and whether a coccygectomy was performed. Pathology reports were reviewed for the size and histologic subtype of the tumor.

1.2. Statistical analysis

Patient age at the time of surgical intervention, tumor location, coccygectomy, tumor size, and histopathology were compared between patients with and without an ARM using nonparametric statistical methods. The Mann–Whitney test was used for continuous variables and chi-square or Fisher's exact tests for categorical variables. Event-free survival (EFS) was calculated from the time of surgery until either

Table 1
Characteristics of patients with sacral and presacral masses of all histologic subtypes from 1990 to 2017.

	ARM (n = 12)	No ARM (n = 34)	p-Value*
Median age at resection (range)	1.4 years (1 day-29.4 years)	9 days (0 days-6.9 years)	0.01
Median follow up, years (range)	4.5 years (6 weeks-16.5 years)	5.6 years (0 days-37.3 years)	0.36
Sex			0.33
Male	5 (42%)	9 (26%)	
Female	7 (58%)	25 (74%)	
ARM Subtype			
Anal stenosis	6 (50%)	N/A	
Cloaca	2 (17%)		
Rectovaginal fistula	2 (17%)		
Perineal fistula	2 (17%)		
Sacral Anomalies			
Hemisacrum	3 (25%)		
Sacral dysplasia	7 (58%)	N/A	
None	2 (17%)		

* P-values result from nonparametric statistical methods.

Table 3
Tumor characteristics of patients with presacral masses of all histologic subtypes from 1990 to 2017.

	ARM (n = 12)	No ARM (n = 34)	p
Median Tumor Size (range)	2.5 cm (1.5–5.0 cm)	6.0 cm (0.6–20.0 cm)	0.036
Coccygectomy*	10 (91%)	21 (91%)	0.99
Histopathology			
Mature teratoma	8 (67%)	20 (59%)	
Immature teratoma	0 (0%)	7 (21%)	
Yolk sac tumor	1 (8%)	3 (9%)	
Lipoma	1 (8%)	0 (0%)	
Ganglioneuroma	0 (0%)	1 (3%)	
Neuroblastoma	0 (0%)	1 (3%)	
Benign epithelial cyst	0 (0%)	1 (3%)	
Unknown	2 (17%)	1 (3%)	
Recurrence	3 (25%)	5 (15%)	0.41
5 Year Event-Free Survival	65% (95% CI: 25% - 87%)	81% (95% CI: 60–92%)	0.44**
Adjuvant chemotherapy	1 (8%)	5 (15%)	0.66

* Coccygectomy information missing for 12 patients.
** Log-rank p-value comparing EFS.

tumor recurrence or malignant degeneration. Freedom from malignancy was compared between the ARM and non-ARM groups for sacrococcygeal teratomas using a log-rank test. Data is presented as median (range) or frequencies (percentages) accordingly. Analyses were conducted using SAS software, version 9.4 (Cary, NC). All p-values are two-sided and those <0.05 were considered statistically significant.

2. Results

2.1. Patient characteristics

Between 1990 and 2017, we identified a total of 50 patients with a diagnosis of a sacral or presacral mass. Four patients with incomplete operative and follow up data were excluded, resulting in 46 patients eligible for inclusion in the study. Twelve patients (26%) had an associated ARM while 34 (74%) did not. Males comprised 42% (5/12) of the ARM group and 26% (9/34) of the non-ARM group. The median age at the time of tumor excision was 1.4 years in the ARM group compared to 9 days in the non-ARM group (p = 0.01). The median time from diagnosis to resection was 18 days in the ARM group (range 1–54 days) compared to 2 days (range 0–17 days) in the non-ARM group (p = 0.03). The most common ARM subtype was anal stenosis, which was seen in 6 (50%) patients, followed by cloaca (n = 2, 16%), rectovaginal fistula (n = 2, 16%), and perineal fistula (n = 2, 16%). Sacral dysplasia was evident in 7 (58%) and hemisacrum seen in 3 (25%) patients with ARM. A summary of the patient characteristics can be found in Table 1.

Table 2 illustrates patient characteristics of those diagnosed with an SCT. Eight patients had an ARM compared to 27 without an ARM. Those with an ARM were diagnosed at a later date compared to those without an ARM (1.7 years vs. 2 days, p = 0.002). Males comprised 50% (n = 4) of the ARM group and 19% (n = 5) of the non-ARM group.

2.2. Tumor characteristics

Analysis of all histologic subtypes demonstrated that the median tumor size was smaller in patients with an ARM compared to those without an ARM (2.5 cm vs. 6.0 cm, p = 0.036). Histopathologically, mature teratoma was the most common tumor seen in patients with an ARM (n = 8, 67%), followed by yolk sac tumor (n = 1, 8%), lipoma (n = 1, 8%), and unknown pathology (n = 2, 16%). Similarly, mature teratoma was the most common tumor seen in the non-ARM group (n = 20, 59%), followed by immature teratoma (n = 7, 21%), yolk sac

Table 2
Characteristics of patients with sacrococcygeal teratomas from 1990 to 2017.

	ARM (n = 8)	No ARM (n = 27)	p-Value
Median age at resection (range)	1.7 years (1 day-29.4 years)	2 days (0 days-6.9 years)	0.002
Median follow up, years (range)	4.1 years (6 weeks-16.5 years)	6.1 years (0 days-37.3 years)	0.11
Sex			0.18
Male	4 (50%)	5 (19%)	
Female	4 (50%)	22 (81%)	
Altman Classification			<.0001**
I	0 (0%)	9 (33%)	
II	0 (0%)	7 (26%)	
III	0 (0%)	1 (4%)	
IV	8 (100%)	5 (19%)	
Unknown	0 (0%)	5 (19%)	
Median Tumor Size (range)	2.8 cm (1.5–5.0 cm)	6.0 cm (1.8–20.0 cm)	0.0493
Coccygectomy*	7 (88%)	16 (94%)	0.99
Recurrence	2 (25%)	3 (11%)	0.58
Malignant Degeneration	1 (13%)	3 (11%)	0.99
5 Year Event-Free Survival***	60% (95% CI: 13%–88%)	87% (95% CI: 63%–96%)	0.25
Chemotherapy	0 (0%)	3 (12%)	

* Coccygectomy information missing for 10 patients.
** Comparison is IV vs. I-III.
*** Log-rank p-value comparing EFS.

tumor (n = 3, 9%), ganglioneuroma (n = 1, 3%), neuroblastoma (n = 1, 3%), benign epithelial cyst (n = 1, 3%), and unknown (n = 1, 3%).

Similarly, the median size of sacrococcygeal teratomas in patients with an ARM was smaller than those with no ARM (2.8 cm vs. 6.0 cm, p = 0.049). All 8 patients with an SCT in the ARM group were found to have an Altman type IV mass (Table 2). Among patients without an ARM, 9 (33%) had Altman type I masses, 7 (26%) had type II, 1 (4%) had type III, and 5 (19%) had type IV. The Altman classification was unable to be determined from the medical record in 5 (19%) patients without an ARM. Table 3 describes the tumor characteristics of the cohort.

2.3. Oncologic outcomes

Chemotherapy was given to one patient in the ARM cohort and 5 in the non-ARM cohort. Recurrence was seen in 3 (25%) patients with an ARM and in 5 (15%) patients without an ARM (p = 0.41). The histologic subtype in patients who had a tumor recurrence was mature teratoma (n = 2) and yolk sac tumor (n = 1) in patients with an ARM, and mature teratoma (n = 2), immature teratoma (n = 1), ganglioneuroma (n = 1), and yolk sac tumor (n = 1) in patients without an ARM. One patient with a yolk sac tumor in the ARM had a recurrence after receiving adjuvant chemotherapy, whereas one patient with an immature teratoma in the non-ARM group had a recurrence after receiving adjuvant chemotherapy.

The 5-year (EFS) for all tumor types was 65% in the ARM group compared to 81% in the non-ARM group (p = 0.44) (Fig. 2). Similarly, the 5 year EFS in patients with a sacrococcygeal teratoma was similar between patients with and without an ARM (60% vs. 87%, p = 0.25) (Fig. 3). Malignant degeneration occurred within 2 years in all cases and were seen at a similar rate in patients with and without an ARM (13% vs. 11%, p = 0.99) (Fig. 4). The median age of patients who experienced a malignant degeneration of their SCT was similar between those who did not (1 day vs. 57 days, p > 0.05). Similarly, tumor size was not found to be associated with malignancy (4.0 cm vs. 5.5 cm, p > 0.05) in our cohort.

3. Discussion

The primary purpose of this study was to evaluate oncologic outcomes in patients with presacral masses with and without an associated ARM. We found that patients with presacral masses and an ARM presented at an older age compared to those who did not have an ARM. However, we did not find a significant difference in recurrence rates, 5-year EFS, or freedom from malignancy between the groups.

Despite the known associations between ARM and tumors of the presacral space, the diagnosis of the latter was made at a significantly older age compared to patients without an ARM (1.4 years vs. 9 days, p = 0.01). This finding is likely explained by several reasons. First, all presacral masses in patients with an ARM in this study were exclusively intrapelvic lesions, compared to only 24% in the cohort of patients without an ARM. The presence of the extrapelvic tumor in the remaining patients makes the diagnosis apparent on examination or during prenatal screening, whereas intrapelvic tumors require imaging for confirmation, and might not be investigated until after the onset of symptoms. This fact is another important reminder that all ARM patients should be screened as a newborn for a presacral mass, ideally at the time of the spinal ultrasound used to assess for tethered cord. In our cohort, intrapelvic tumors in patients without an ARM were also resected later than those with an extrapelvic lesions (2.4 years vs. 5 days) and

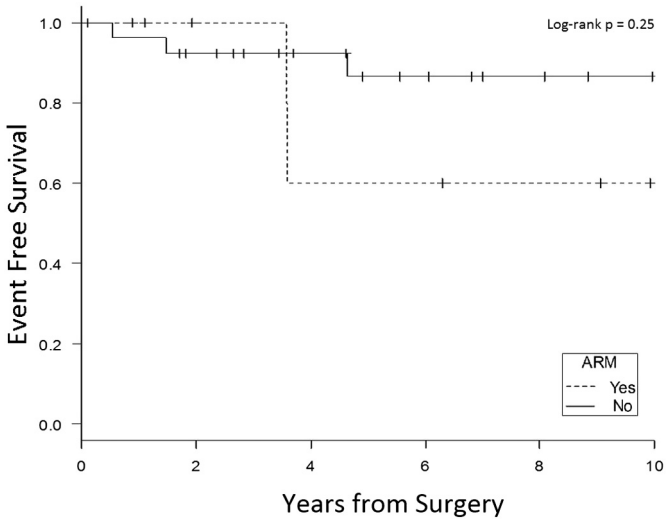

Fig. 3. Kaplan–Meier curve illustrating the event-free survival of patients with sacrococcygeal teratomas by the presence or absence of an associated anorectal malformation.

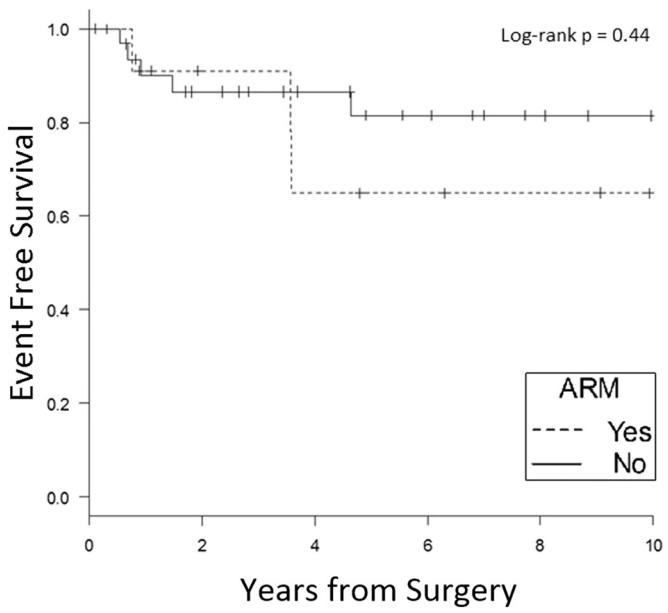

Fig. 2. Kaplan–Meier curve illustrating the event-free survival among patients with all tumor types by the presence or absence of an associated anorectal malformation.

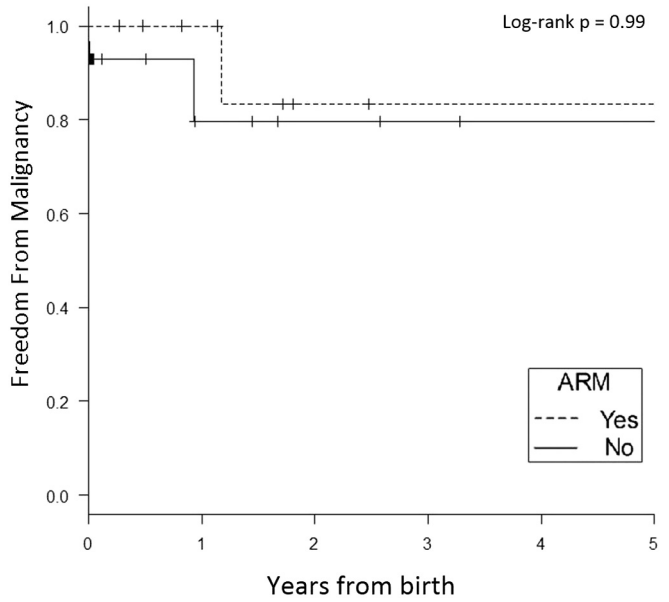

Fig. 4. Kaplan–Meier curve illustrating the malignancy-free survival of patients with sacrococcygeal teratomas by the presence or absence of an associated anorectal malformation.

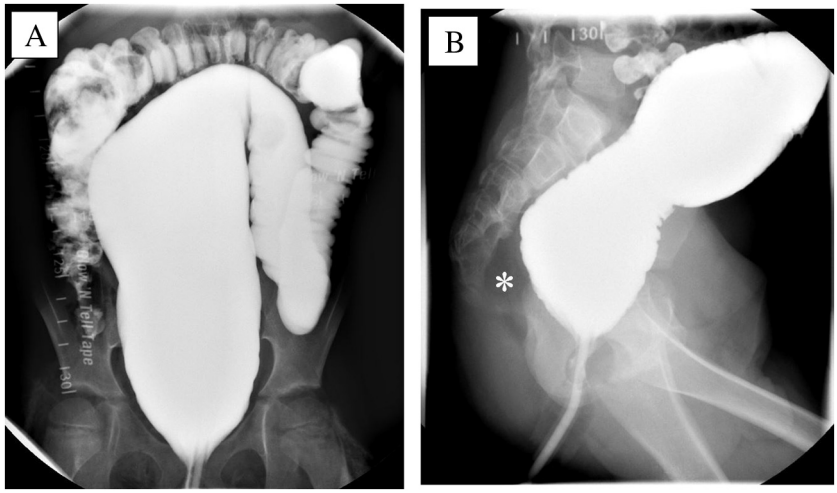

Fig. 5. Contrast enema in patient with severe constipation demonstrating A) dilated rectosigmoid and B) widening of the presacral space (*) caused by compression by a presacral mass.

at a similar age to patients with ARM, suggesting that this is not a feature unique to patients with ARM but rather a function of the tumor location.

Second, the delay in diagnosis of a presacral mass in patients with an ARM may be confounding, as the symptoms can be vague and often attributed to their ARM. Constipation is the most common symptom in patients diagnosed with presacral mass after the neonatal period, and is typically caused by mass effect of this lesion exerting pressure on the rectum [1,11]. Constipation however is not uncommon in

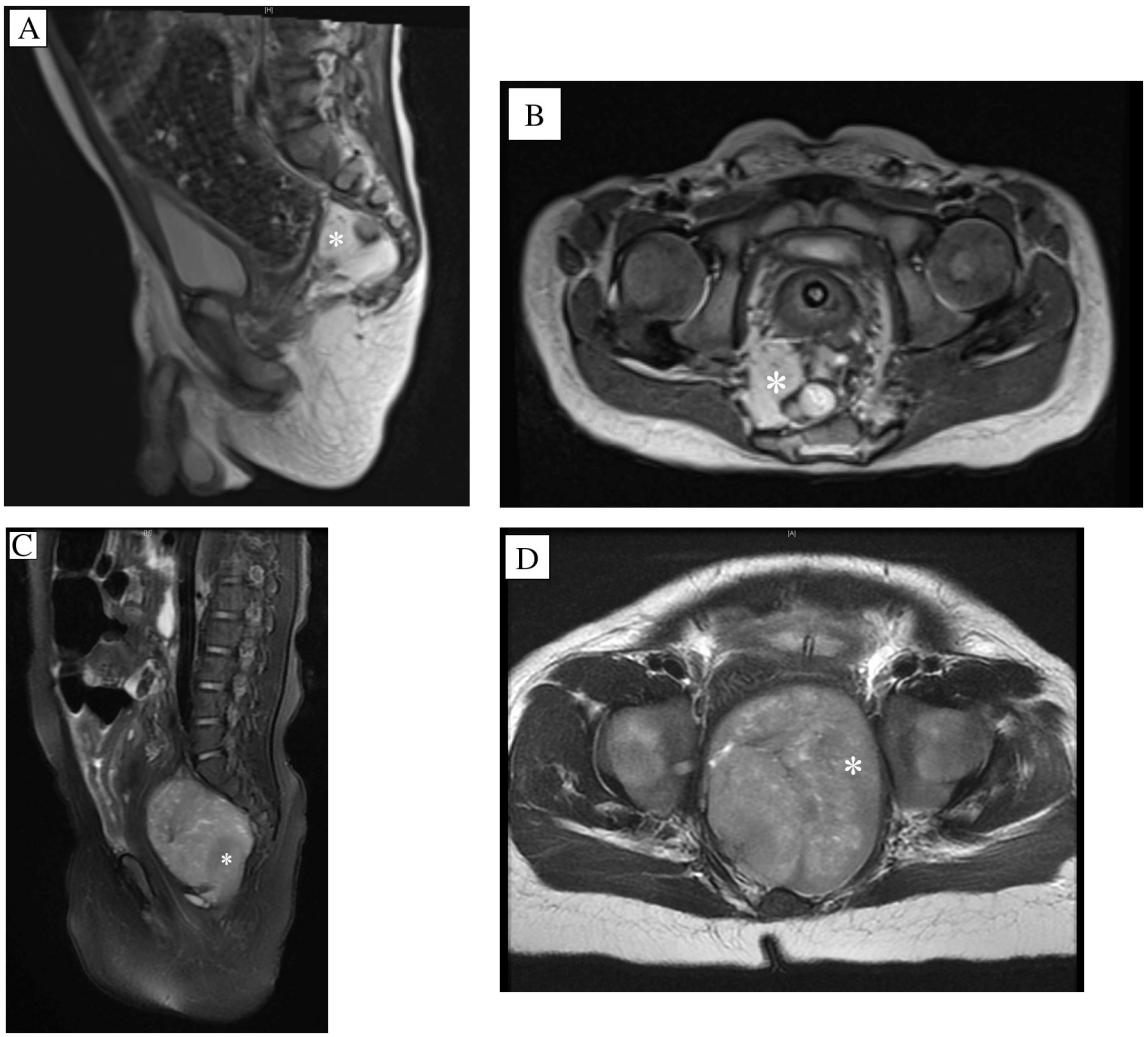

Fig. 6. T2 weighted pelvic magnetic resonance imaging demonstrating a presacral mass in a patient with (A and B) and without (C and D) an ARM. A) Sagittal and B) axial images of a patient with an ARM and presacral mass with fatty and cystic components projecting into the pelvis from the sacral canal. C) Sagittal and D) axial images of a patient without an ARM and a large solid presacral mass (mass denoted by * in all images).

children with ARM and may be caused by functional deficits related to poorly developed musculature and abnormal innervation. Thus, the existence of a presacral mass should be considered in any patient with an ARM and constipation, particularly when symptoms are new or evolving. The presence of a widening of the presacral space on lateral pelvic x-ray or on a contrast enema (Fig. 5) may suggest the presence of a mass, which can be confirmed with cross sectional imaging (Fig. 6) [12]. Furthermore, anal stenosis is often less apparent than other, more severe ARM subtypes, and as such can often be overlooked in the newborn period. It is important to avoid anal dilation in these patients as they may require surgical correction, and consider an MRI to rule out a presacral mass given their frequent association.

In addition to the later diagnosis of a presacral mass in the patients with an ARM, they also experienced a small delay in time to resection compared to patients without an ARM (18 days vs. 2 days, $p = 0.03$). Although statistically significant, this delay is unlikely to be of any clinical relevance, as the delay is small compared to what is likely years of asymptomatic tumor growth in patients with an ARM.

While benign SCTs comprise the most common tumors of the presacral space in children, malignant lesions are not uncommon. Overall, the distribution of benign and malignant lesions in this study were similar between patients with and without an ARM, consistent with prior histopathologic reports between the two groups [13].

Recurrence is a concern following resection of a presacral mass, as the risk of malignant degeneration of the lesion is not negligible [14]. Four of the 35 patients (11%) in our study experienced a malignant degeneration of their SCT, including 1 in the ARM group (13%) and 3 in the non-ARM group (11%), although the median tumor size and age of the time of resection was similar to those with benign teratomas. In their report of 205 patients with SCT and 16 patients with Currarino syndrome, Dirix et al. [13] reported 0/16 patients with an ARM as undergoing malignant transformation of their teratoma, similar to our rate of 13% (1/8). They also showed a nearly identical malignancy rate in patients without an ARM (13% vs. 11%) when compared to our study. Factors associated with tumor recurrence reported previously include incomplete resection of the tumor, intraoperative tumor spillage, immature pathology, and failure to perform complete coccygectomy [15,16]. The rationale behind coccygectomy includes complete removal of all primordial cell nests located at Hensen's node [17]. Of patients in the ARM group with a recurrence, coccygectomy was not performed in 2 patients, compared to 1 patient with recurrence in the non-ARM group.

In our series, 3 patients with ARM and a presacral mass were found to have hemisacrum. Of these patients, all 3 had an anal stenosis, which is the most common ARM subtype associated with presacral mass [18]. An additional 5 patients had severe sacral hypoplasia. Given the high association of anal stenosis with presacral mass, which may approach 30%, an MRI is recommended in patients with this ARM subtype to evaluate for both a presacral mass and tethered cord [19].

There is a paucity of data in the published literature on the long term functional outcomes of patients with Currarino syndrome [11,20]. This underscores the need for continued long-term evaluation through multi-institutional registries, and definitive management at centers with expertise in pediatric colorectal and oncologic surgery to maximize oncologic and functional outcomes in these complex patients. All patients undergoing resection of presacral masses with and without ARMs at our institution are referred to the bowel management program to achieve optimal functional results.

We recognize several limitations to our study primarily related to the retrospective design and small number of patients analyzed. The infrequency of these conditions complicates the study of presacral tumors in children and makes prospective, adequately powered studies difficult to perform, particularly within a single institution. Studies are currently ongoing to investigate outcomes of these patients using data from a multicenter registry comprised of high volume centers. Next, histopathology data was missing from 2 (17%) patients in the ARM group along with tumor location missing from 8 (24%) of the non-ARM group, both of which limits our ability to draw conclusions from these results, as they represent a significant subset of this small cohort. Similarly, missing coccygectomy data on 12 (26%) patients also limits the conclusions, as this is known to be a factor associated with SCT recurrence. As our institution serves as a referral center for patients with complex pediatric colorectal disease, we often see patients for evaluation of a previously repaired anorectal malformation or for bowel management after their tumor was addressed elsewhere. Thus, many operative reports or pathology data was not available for our independent review despite exhaustive attempts to retrieve these records from referring centers. Furthermore, our comparison includes extremely heterogeneous groups spanning a wide age range with a variety of ARM subtypes that could not be controlled for given the extremely small numbers. Additionally, although we believe the follow up with each patient was adequate to capture most recurrences, it is possible that significantly delayed recurrences might have not been diagnosed until after their care ended at our institution. Some patients might therefore have been treated for a recurrence by another (e.g. adult) provider, which would cause us to underestimate the actual recurrence rate in our cohort.

4. Conclusion

Presacral masses in patients with ARM are diagnosed and resected at a later age, are more likely to be intrapelvic in location, although they appear histologically similar and have similar rates of recurrences when compared to patients without ARM. Further study is needed to investigate and optimize the bowel management and functional outcomes for all patients with presacral masses.

CRediT authorship contribution statement

Devin R. Halleran: Data curation, Formal analysis, Writing - original draft, Writing - review and editing. **Alejandra Vilanova-Sanchez:** Conceptualization, Data curation, Formal analysis, Writing - original draft, Writing - review and editing. **Carlos A. Reck:** Conceptualization, Data curation, Formal analysis, Writing - original draft, Writing - review and editing. **Tassiana Maloof:** Data curation. **Laura Weaver:** Data curation. **Joseph Stanek:** Formal analysis, Writing - review and editing. **Marc A. Levitt:** Conceptualization, Formal analysis, Writing - original draft, Writing - review and editing. **Richard J. Wood:** Conceptualization, Formal analysis, Writing - original draft, Writing - review and editing. **Jennifer H. Aldrink:** Conceptualization, Formal analysis, Writing - original draft, Writing - review and editing.

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