Is intrapelvic tumor height correlated with long-term functional outcomes after surgical treatment of sacrococcygeal teratoma?

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Research Article

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Abstract

Background: Sacrococcygeal teratoma (SCT) is considered the most common congenital neoplasm in neonates. This study aimed to assess long-term impacts on urinary and anorectal functions and quality of life (QoL) in patients who underwent surgery for SCT and identify predictive factors for these outcomes.

Methods: Medical records of all patients who underwent surgery for SCT at the Armand Trousseau Hospital and had a minimum follow-up of 4 years were retrospectively reviewed. Age-appropriate questionnaires were used to evaluate urinary and anorectal functions and QoL. Urodynamic studies and radiological images were also retrospectively reviewed.

Results: Twenty-six patients (median age=17 years) were identified. Among them, 12 patients had functional impairments: seven had isolated anorectal dysfunction and five had both urinary and anorectal dysfunctions, including neurogenic bladder (n=2) and/or detrusor sphincter dyssynergia (n=3). Anorectal dysfunction (n=12) included chronic constipation (n=11) and/or fecal soiling (n=8). A significant correlation was observed between adverse outcomes and intrapelvic tumor height (ITH) (median was 27 mm and 15 mm with and without functional impairments, respectively; $p<0.05$). Of 12 questionnaire respondents, two patients experienced uro-anorectal dysfunction and five experienced isolated anorectal dysfunction. Most patients reported having a good QoL, and only one patient reported a severe impact on daily life.

Conclusions: Urinary and anorectal dysfunctions are non-negligible long-term outcomes in SCT patients that are significantly related to ITH. Postoperative follow-up enables early detection of bladder and anorectal impairment and treatment with conservative measures that can improve QoL.

Level of evidence: III

INTRODUCTION

Sacrococcygeal teratoma (SCT) is a rare congenital tumor with an incidence of 1 in 14,000–28,000 live births and a female predominance of 4:1 [1]. However, it is the most common neonatal solid tumor [2]. SCT may be diagnosed during antenatal, neonatal, infancy, or early childhood periods [3].

The treatment of SCT involves surgical resection of the coccyx and tumor, which extends a variable distance into the presacral space and pelvis. The causes for morbidity following surgery for SCT are likely to be multifactorial due to local compression on pelvic nerves and pelvic floor musculature caused by the original tumor and possible iatrogenic injury during surgical resection or associated urogenital anomalies, such as urethral stenosis or urethrovaginal fistula [4].

Few studies have evaluated the long-term impacts on urinary and anorectal functions or the quality of life (QoL) of patients treated for SCT and were either based on medical records alone or questionnaire-based surveys. Several factors such as the Altman classification, tumor size, abdominoperineal resection, and tumor histopathology have been heterogeneously reported as risk factors for unfavorable outcomes [5-8]. However, the intrapelvic tumor height (ITH) in relation to the sacro-lumbar vertebrae was not among the predictive factors considered. Therefore, this study aimed to assess long-term impacts on urinary and anorectal functions and QoL in patients who underwent surgery for SCT at our institution and identify predictive factors for these outcomes.

METHODS

Data collection

In this retrospective study, we included all patients who underwent surgery for SCT from 1984 to 2017 at Armand Trousseau Hospital. We reviewed their medical records to identify demographic, clinical, radiological, operative, and histological data to determine long-term functional outcomes and associated predictive factors.

Urinary and anorectal functional outcomes were assessed based on data in the medical records and by questionnaires sent to patients if they were more than 18 years of age or to their parents if they were younger. Urological outcomes were assessed through the validated questionnaire developed by Shalaby et al. [9]. Anorectal outcomes were based on the international Krickenbeck classification for the assessment of postoperative results following surgeries for anorectal malformations [10]. The QoL of patients treated for SCT was assessed using the pediatric incontinence questionnaire (PinQ) designed by Bower et al. [11]. Written informed consent was obtained from the patients or their parents beforehand.

Urodynamic studies (UDSs) performed for all suspected cases of bladder dysfunction following surgical treatment of SCT were reviewed and analyzed with the help of the physical medicine and rehabilitation department.

We investigated potential predictive factors associated with functional impairments following SCT resection by analyzing patients' demographic and clinical information, such as sex, age at diagnosis, Altman classification, radiological measurement of ITH relative to the sacro-lumbar vertebrae, radiological and histological characteristics of the tumor, operative approach, whether a complete resection was performed, and whether radiotherapy or chemotherapy was required.

The ITH was evaluated retrospectively by reviewing all preoperative radiological images obtained using magnetic resonance imaging (MRI) or computer tomography (CT). Radiological images were reviewed by our radiologist blinded from long-term outcomes of the patients. ITH was measured from the pubococcygeal line to the maximum height of the tumor relative to the sacro-lumbar vertebra.

Data analysis

Statistical analysis was conducted using GraphPad Software version 8.4.3 (San Diego, California, USA). Data are reported as median, interquartile range (IQR), and percentage values. The Mann-Whitney U test, The Fisher's exact test and Student's t-test were used to analyze the data. p-values <0.05 were considered statistically significant.

RESULTS

Entire cohort

We identified 26 patients (21 female and 5 male [4:1 ratio between sexes]) who met the inclusion criteria. The patients had a median follow-up of 68 months.

Sixteen patients had a prenatal diagnosis, while 10 patients were diagnosed postnatally. The Altman classification could be determined from radiological and/or operative reports. Seven patients underwent surgery for type I SCT, 10 for type II, six for type III, and three for type IV. All patients underwent surgical excision: six patients who were diagnosed prenatally were operated on the days immediately following birth, while six other patients had a late diagnosis and were operated on between 11 and 36 months of age. The perineal approach was adopted in 20 patients and the abdomino-perineal approach in six patients. Pathological examination showed that 21 patients were diagnosed with a mature teratoma, four with an immature teratoma, and one with a localized malignant mixed germ cell tumor. We observed four cases of recurrence. One patient initially had an immature teratoma that recurred in the form of a vitelline tumor and was treated by chemotherapy alone. Another patient had local recurrence of an immature teratoma with an intramedullary extension and received another operation and follow-up chemotherapy. The two other patients with initial mature teratomas received subsequent operations, and one of these two patients received follow-up chemotherapy for incomplete resection (Table 1).

Long-term functional impairments were observed in 12 patients (Tables 1 and 2). Seven patients had isolated anorectal dysfunction and five patients had both urinary and anorectal dysfunctions. Anorectal dysfunction ranged from chronic constipation in 11 patients to fecal soiling in eight patients. Urinary dysfunction manifested as voiding difficulty, urinary incontinence, and recurrent urinary tract infections (UTI). All patients with urinary dysfunction underwent a UDS, which identified neurogenic hypersensitive and hypocompliant bladder (n=1), hyperactive bladder associated with detrusor sphincter dyssynergia (n=1), or isolated detrusor sphincter dyssynergia (n=2). One case manifested with recurrent UTI and urinary incontinence with normal UDS findings. Vesicoureteral reflux (VUR) was also observed in three out of these five patients (Table 3). They were treated with clean intermittent catheterization, antibiotic prophylaxis, and anticholinergic medication or bladder augmentation in one severe case. Recurrent UTI was observed in nine patients, which included all patients with urinary dysfunction (n=5) and four other patients. There was no statistically significant difference between patients who did or did not have functional impairments in their sex distributions, Altman classifications, surgical approaches, histological characteristics of tumors, recurrence, or treatment with chemotherapy.

The ITH was measured in 18 patients with preoperative MR and/or CT images. Six patients were operated on immediately after birth based on antenatal ultrasonography findings or due to a large tumor found postnatally and did not have an ITH measurement. Two patients underwent preoperative ultrasound alone and the ITH measurement was not well-defined, therefore these two patients were not included in the statistical analysis. ITH was significantly correlated to the functional outcome regardless of the Altman classification (median ITH was 27 mm and 15 mm in the group with and without functional impairment, respectively; $p<0.05$) (Table 4).

Patients responding to questionnaires

Twelve patients completed the questionnaires. The demographic and clinical characteristics of the patients are shown in Table 5. The median age of respondents was 13 years (range=4–37 years). Five of 12 respondents experienced no urinary or anorectal dysfunction. Recurrent UTI was noted in five patients, among whom two had UTIs that manifested as urinary incontinence and voiding difficulty due to underlying neurogenic bladder or detrusor sphincter dyssynergia (Table S1). Anorectal complications occurred in seven patients and included chronic constipation (n=7) that ranged from grade I in 71% of patients treated conservatively by modification of dietary regimens to grade III in 29% of patients who required disimpaction or colonic irrigation. Fecal soiling was reported in three patients (Table S2).

Among the seven patients with anorectal complications, two of them had neurogenic bladder.

The QoL of patients who underwent surgery for SCT was assessed using the PinQ and we found that most patients (92%) experienced mild impacts on their daily life from the functional outcomes described above. The patients had very good outcomes regarding mental, social, and physical demands. Only one patient reported to have severe mental and social impact.

DISCUSSION

In this study, 46% of patients who underwent surgery for SCT experienced long-term uro-anorectal dysfunction with minor impact on the QoL. Our findings suggest that the ITH may be a predictive factor for uro-anorectal impairment in this context.

Anorectal impairments were the most common type of dysfunction and manifested as constipation (67%) and/or fecal incontinence (23%). On the other hand, detrusor sphincter dyssynergia (60%) followed by neurogenic bladder (40%) were the most common urologic issues in patients with urinary dysfunction. Previous studies have shown a substantial heterogeneity in the frequency of urinary and anorectal problems following surgical excision of SCT. Urological and anorectal functional outcomes following SCT resection were reportedly observed in 7–37% and 8–47% of patients respectively, without description of combined or isolated dysfunction. (Table 6). Whether functional disorders are caused by a mass effect of the tumor size or surgical damage to intrapelvic nerves and muscles remains controversial in the literature [5,6].

A high incidence of functional impairment in patients operated for SCT resection found to be related to the age at the time of surgery, tumor size, or high-grade Altman classification [5,6,12,13].

In a study similar to ours, Partridge et al. reviewed the outcomes of SCT resection in 45 patients and demonstrated that 58% of the patients did not have impairment of bladder or bowel function, while 42% had dysfunctional sequelae [12]. Likewise, Malone et al. found that 41% of the patients in his study had functional sequelae following resection of SCT [5]. Both Partridge et al. and Malone et al. showed that tumors with Altman type III or higher grades had a higher incidence of fecal and/or urinary incontinence [5, 12]. However, some studies showed no correlation between the Altman type and functional impairment disorders in patients who underwent surgery for SCT [14-16]. Masahata et al. and Hambraeus et al. reported that the maximum tumor size was significantly associated with the high incidence of dysfunctional outcomes [6, 15], while Güler et al. found no significant difference [7]. We were unable to find significant correlations between a high Altman classification or histopathological characteristics of tumors and dysfunctional outcomes ($p>0.05$) (Table 1). Similar observations were reported by Shalaby et al. [9] However, we observed a significant clinical correlation between ITH and functional impairment ($p <0.05$) (Table 4), which may be explained by anorectal and urinary bladder innervation. The parasympathetic fibers in the inferior hypogastric plexus originate in between the second and fourth sacral segments of the spinal cord. The sympathetic fibers that innervate the bladder originate between spinal cord segments T11 and L2, and fibers from L5 supply the rectum and anal internal sphincter [17,18]. This supports our observation that functional impairments in patients who underwent surgery for SCT were significantly correlated to ITH and explains why all patients with urinary dysfunction also experienced anorectal dysfunction. Therefore, this suggests that ITH may be a better predictive factor than Altman classification for uro-anorectal dysfunction in patients who underwent surgery for SCT.

The QoL of our patients was assessed using the PinQ, which showed that most of the patients (92%) reported that uro-anorectal functional outcomes only mildly impacted their daily life activities. It has been suggested that the QoL of patients treated for SCT may be impaired due to urinary and anorectal dysfunctions [8,14], however, no correlation was found in our study. Hambraeus et al. were also not able to establish a statistical correlation between physical function and QoL [19].

This study had some limitations, which include its retrospective and single center design and small sample size. The number of patients was insufficient to establish significant correlations for various outcomes. Moreover, only a few patients reported both urological and anorectal dysfunction, but some experienced isolated anorectal dysfunction. Thus, a larger sample size is required to explore this finding. Nonetheless, this has been one of the few studies to use both medical records and questionnaire analysis to evaluate the long-term functional outcomes and predictive factors of uro-anorectal dysfunction in patients treated for SCT. This study emphasizes the need for multi-institutional collaborative research to highlight the main predictive factors of functional impairment.

In conclusion, urinary and anorectal dysfunctions are non-negligible long-term outcomes in patients treated for SCT. These functional impairments were found to be significantly related to the ITH. Thus, preoperative radiological exploration by MRI or CT should be considered for all patients with SCT as a predictive assessment. Moreover, postoperative follow-up allows for the early detection of bladder and anorectal impairments and treatment by conservative measures, which can improve a patient's QoL. Further multi-center studies are needed to confirm these findings.

Abbreviations

CT: computed tomography; **IQR:** interquartile range; **ITH:** intrapelvic tumor height; **MRI:** magnetic resonance imaging; **QoL:** quality of life; **SCT:** sacrococcygeal teratoma; **UTI:** urinary tract infection; **VUR:** vesicoureteral reflux.

Declarations

Author's contribution:

- Study conception and design: Yousuf Al-shaqsi , Georges Audry , Sabine Irtan
- Data acquisition: Yousuf Al-shaqsi, Eleonore Blondiaux , Aurore Coulomb-Lhermine, Pauline Lallemant
- Analysis and data interpretation: Yousuf Al-shaqsi, Pauline Lallemant
- Drafting of the manuscript: Yousuf Al-shaqsi
- Critical revision: Sabine Irtan, Georges Audry

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No conflict of interest to disclose

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Not required

Ethics approval

This study is a retrospective study, conducted in accordance with the principles of Declaration of Helsinki. The study protocol was exempted by the Institutional Board of Armand Trousseau Hospital because it is a non-experimental medical study but cohort and quality-improvement study, the personal identities cannot be identified and anonymous questionnaires were used.

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Tables

Table 1. Demographic and clinical characteristics of patients in the entire cohort (n=26)

Characteristics	Entire cohort (n=26)	Functional impairment		
		Yes (n=12) 46%	No (n=14) 54%	p-value
Sex				
Male	5 (19%)	2 (17%)	3 (21%)	1.0000
Female	21 (81%)	10 (83%)	11 (79%)	1.0000
Age at diagnosis			10 (71%)	
Prenatal (WGA)	16 (62%)	6 (50%)	4 (29%)	0.7501
Postnatal (day)	10 (38%)	6 (50%)		0.7112
Weight of birth (g) median	3200 (1420–4500)	2980 (1700–4500)	3300 (1420–3730)	1.0000
Altman Classification				
I	7 (27%)	3 (25%)	4 (29%)	1.0000
II	10 (38%)	3 (25%)	7 (50%)	0.4682
III	6 (23%)	4 (33%)	2 (14%)	0.6880
IV	3 (12%)	2 (17%)	1 (7%)	0.5977
Radiology characteristic				
Tissue	2 (8%)	2 (17%)	0	0.4815
Cystic	3 (11%)	1 (8%)	2 (14%)	1.0000
Mixed	21 (81%)	9 (75%)	12 (86%)	0.7761
AFP (median/IQR)	15420/25492	18200/38359	13709/39298	0.3223
Chemotherapy	5 (19%)	4 (33%)	1 (7%)	0.1824
Age at surgery (days)				
Median	2.5 (0–4320)	8 (0–4320)	2.5 (1–3600)	0.1326
IQR	68	53	101	
Surgical approach				
Perineal	20 (77%)	7 (58%)	13 (93%)	0.5510
Abdominoperineal	6 (23%)	5 (42%)	1 (7%)	0.1779
Resection				
Complete	24 (92%)	10 (83%)	14 (100%)	0.7827
Incomplete	2 (8%)	2 (17%)	0	0.4815
Histology				
Mature	21 (81%)	7 (58%)	14 (100%)	0.5506
Immature	4 (15%)	4 (33%)	0	0.1029
Malignant	1 (4%)	1 (8%)	0	0.4815
Recurrence	4 (15%)	3 (25%)	1 (7%)	0.5977
Reoperated	3 (11%)	2 (17%)	1 (7%)	0.5977

IQR: interquartile range; WGA: weeks of gestational age. AFP: alpha fetoprotein

Table 2. Demographic and clinical characteristics of patients with functional impairments (n=12)

Characteristics	Uro-anorectal dysfunction (n=5) 19 %	Isolated anorectal dysfunction (n=7) 27%
Sex		
Male	0	2 (29%)
Female	5 (100%)	5 (71%)
Age at diagnosis		
Prenatal (WGA)	4 (80%)	2 (29%)
Postnatal (day)	1 (20%)	5 (71 %)
Weight of birth (g) median	3100 (1700–3420)	2650 (2200–4500)
Altman classification		
I	0	3 (43%)
II	2 (40%)	1 (14%)
III	2 (40%)	2 (29%)
IV	1 (20%)	1 (14%)
Radiology characteristics		
Tissue	0	2 (29%)
Cystic	0	1 (14%)
Mixed	5 (100%)	4 (57%)
AFP (median/IQR)	19515/43489	15420/41652
Chemotherapy	2 (40%)	2 (29%)
Age at surgery (day)		
Median	1 (0–30)	36 (0–4320)
IQR	22	329
Surgical approach		
Perineal	2 (40%)	5 (71%)
Abdominoperineal	3 (60%)	2 (29%)
Resection		
Complete	3 (60%)	7 (100%)
Incomplete	2 (40%)	0 (0%)
Histology		
Mature	3 (60%)	4 (57%)
Immature	2 (40%)	2 (29%)
Malignant	0	1 (14%)
Recurrence	1 (20%)	2 (29%)
Reoperated	1 (20%)	1 (14%)

IQR: interquartile range; WGA: weeks of gestational age; AFP: alpha fetoprotein.

Table 3. Urodynamic study findings in patients with urinary dysfunction outcomes (n=5)

Patient's order	Hypersensitive bladder (n=1)	Hypocompliant bladder (n=1)	Hyperactive bladder (n=1)	Detrusor sphincter dyssynergia (n=3)	VUR (n=3)
2	X	X			X
6				X	
9			X	X	
15					X
17				X	X

VUR: vesicoureteral reflux.

Table 4. Patients with preoperative radiological exploration by MRI or CT (n=18) demonstrating a relationship between functional impairment and intrapelvic tumor height

Patients' order (n=18)	Functional impairment (n=8)		No functional impairment (n=10)	Intrapelvic tumor height		p-value
	Uro-anorectal (n=2)	Anorectal (n=6)		Height (mm)	Vertebral level	
6	X			35	S1/S2	
15 (MRI-ANT at 34 WGA)	X			30	L5	
3		X		25	S2/S3	
5		X		27	S1	
11 (MRI-ANT at 33 WGA)		X		27	L5/S1	
16		X		27	S2/S3	
18		X		0	C	0.0283
21		X		24	S2/S3	
7			X	15	S4/S5	
8			X	20	S4	
10			X	25	S2	
12			X	20	S3	
13			X	0	C	
14			X	0	C	
19			X	6	S5	
20			X	15	S4	
24			X	22	S3/S4	
26			X	0	C	

CT: computed tomography; MRI: magnetic resonance imaging; C: coccyx; MRI-ANT: magnetic resonance imaging -antenatal; WGA: weeks of gestational age.

Table 5. Demographic and clinical characateristics of the questionnaire respondents (n=12)

Characteristics	Respondents (n=12)	Uro-anorectal dysfunction (n =2)	Isolated anorectal dysfunction (n=5)	No functional impairment (n=5)
Sex				
Male	2 (17%)	0 (0%)	1 (20%)	1 (20%)
Female	10 (83)	2 (100%)	4 (80%)	4 (80%)
Age of diagnosis				
Prenatal (WGA)	7 (58%)	1 (50%)	2 (40%)	4 (80%)
Postnatal (day)	5 (42%)	1 (50%)	3 (60%)	1 (20%)
Weight of birth (g) median	3258 (1420–4500)	3260 (3100–3420)	2650 (2200–4500)	3150 (1420–3635)
Altman classification				
I	2 (17%)	0 (0%)	2 (40%)	0 (0%)
II	7 (58%)	1 (50%)	1 (20%)	5 (100%)
III	2 (17%)	0 (0%)	2 (40%)	0 (0%)
IV	1 (8%)	1 (50%)	0 (0%)	0 (0%)
Radiology characteristic				
Tissue	1 (8%)	0 (0%)	1 (20%)	0 (0%)
Cystic	1 (8%)	0 (0%)	1 (20%)	0 (0%)
Mixed	10 (83%)	2 (100%)	3 (60%)	5 (100%)
AFP (median/IQR)	18638/50043	16430/30240	15420/30923	33516/54940
Chemotherapy	2 (17%)	1(50%)	1 (20%)	0 (0%)
Age of intervention (day)				
Median	1.5 (1–330)	16 (1–30)	1 (1–330)	2 (2–7)
IQR	23	29	59	4.5
Surgical approach				
Perineal	10 (83%)	1 (50%)	4 (80%)	5 (100%)
Abdominoperineal	2 (17%)	1 (50%)	1 (20%)	0 (0%)
Resection				
Complete	11 (92%)	1 (50%)	5 (100%)	5 (100%)
Incomplete	1 (8%)	1 (50%)	0 (0%)	0 (0%)
Histology				
Mature	10 (83%)	1(50%)	4 (80%)	5 (100%)
Immature	2 (17%)	1 (50%)	1 (20%)	0 (0%)
Malignant	0 (0%)	0 (0%)	0 (0%)	0 (0%)
Recurrence	2 (17%)	0 (0%)	2 (40%)	0 (0%)
Reoperated	1 (8%)	0 (0%)	1 (20 %)	0 (0%)

IQR: interquartile range; WGA: weeks of gestational age. AFP: alpha fetoprotein

Table 6. Literature review of functional outcomes in patients treated for SCT

Study	Patients (n)	Follow-up	Altman classification	Histology	Constipation	Fecal incontinence	Urinary incontinence	Predictive factors
Malone et al. [5] A monocentric, retrospective study	33	5 years (2–12)	I-II 56% (19) III-IV 44% (14)	Benign 79 % Malignant 21%	24% (8)	24% (8)	7% (2)	-Altman III / IV - Abdominoperineal approach
Hambraeus et al. [6] A monocentric, controlled study	17	Median age 7.3 years	I=41% (23) II=18% (11) III=29% (11) IV=12% (8)	Mature 82% Immature 18%	47%	29%	29%	-Tumor size
Güler et al. [7] A monocentric, retrospective study	27 out of 40	Mean f/u 78.5 months (26–206)	I=22.5% II=25% III=37.5% IV=15%	Mature 65% Immature 25% Malignant 10%	25.9% (7)	3.7% (1)	7.4% (2)	-Neither Altman nor tumor size is statistically significant
Rintala et al. [8] A monocentric, retrospective study	26	Mean age 30 years (19–45)	I 46% (12) II 30% (8) III-IV 23% (6)		34.6% (9)	11.5% (3)	19% (5)	-Altman has no statistical significance
Shalaby et al. [9] Multicentric, retrospective study	31	Median age 12 years (5–35)	I-II 59% III-IV 41%		39% (9)	19% (6)	29% (9)	-No correlation to sex, age, histology, or Altman
Partridge et al. [12] A monocentric, retrospective study	45	Median 41.5 months (12–124)	I=20% II=53% III=17.8% IV=8.9%		29.9 %	8.9%	15.6%	-Altman classification
Masahata et al. [15] A monocentric, retrospective study	29	≥ 3 years	III-IV 41.4%	Mature 79.3% Immature 20.7 %	17.2%	10.3%	13.8%	-Tumor size -Age at surgery
Hambraeus et al. [19] A multicentric, retrospective study	49	Median age 8.9 years (3.6–28.8)	I = 43% II= 21% III= 21% IV=15%	Mature 72% Immature 26% Malignant 2%	20%	23%	28%	-Gestational age
F. COZZI et al. (2007) [20] A monocentric,	13	Mean age 25 years (17–38)	I= 56% II= 11% III=11%	Not evaluated	38% (5)	8% (1)	15%stress incontinence 23% enuresis	-Not evaluated

retrospective study			IV=22%					
M. Berger et al. (2010) [21] A monocentric, retrospective study	24	Mean age at f/u 79 months	I = 21% (5) II= 21% (5) III= 21% (5) IV = 37% (9)	Mature 71% (17) Immature29% (7)	8% (2)	4% (1)	8% (2)	-Altman has no statistical significance
B.A. Khalil et al. (2009) [22] A monocentric, retrospective study	12	Average f/u 10.6 years (1–17)		Malignant only	23%		8%	-Not evaluated
Van Heurn et al. [23] A multicentric, retrospective study	47	Mean age 26.2 years	I-II =73 % III = 16 % IV= 11%	Mature 64 % Immature 11% Malignant 25%	21.7% (10)	23.4% (11)	19.6 % (9)	-Altman has no statistical significance
Villamil et al. [24] A monocentric, retrospective study	8	Mean age 23 years (4–37)	I = 37.5% II = 25% III = 25% IV = 12 %	Mature 37.5% Immature 62.5%	25%	12 %	37%	-Not evaluated

f/u: follow-up

Supplementary Files

This is a list of supplementary files associated with this preprint. Click to download.

- SupplementaryMaterials.pdf