Sacrococcygeal Teratoma: 25 Year Experience

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Abstract

We retrospectively studied all thirty-five children (M 6, F 29) with sacrococcygeal teratomas admitted to Siriraj Hospital between 1974 and 1999. Although an abdominal delivery is recommended for lesions greater than 5 cm to avoid dystocia, the average diameter of masses which required interventions from dystocia (n = 3) was not different from vaginal delivery (n = 27). All except two first presented with sacral masses recognized at birth. One patient presented with an abdominal mass and the last one was diagnosed after suffering from difficulty in urination. Ninety-seven per cent of cases were completely excised initially (32 sacral, 2 abdomino-sacral approaches), however, six patients required other treatment for recurrent diseases. One mature teratoma recurrence was resected. Two patients who had malignant recurrences following complete benign excisions, died from advanced malignancy. Four presented with malignancy initially. Wound infection, bladder atony and UTI were the most common complications postoperatively. Advanced malignancy was the major cause of death. No patient died directly from the procedure.

Key word : Sacrococcygeal Teratoma, Teratoma, Immature Teratoma, Yolk Sac Tumor, Endodermal Sinus Tumor Germ Cell Tumor, Sacral Tumor, Histology

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Although sacrococcygeal teratoma and their malignant counterparts are the most common solid tumours in neonates, these tumours are really uncommon. Each paediatric surgical unit in Thailand has its own very limited experience. The infrequency of sacrococcygeal teratoma precludes

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an accumulation of sufficient data to study its background, clinical presentation, treatment and the outcome of treatment. Many unique behaviors including an increasing incidence of malignancy in sacrococcygeal teratomas which correlates proportionately with age and a malignant recurrence encountered in a patient whose primary tumour was reported as benign, were fascinating enough for us to conduct this study.

Purpose of the study

The aim of this study was to reveal the natural history of these tumours, clinical manifestations, differentiation between benign and malignancy, the malignant transformation, methods of treatment and results of treatment. This study also examined what the best route of delivery for newborns with this disease is.

MATERIAL AND METHOD

We retrospectively studied all children with sacrococcygeal germ cell tumours admitted to Siriraj Hospital between January 1974 and January 1999. All details of each patient recorded in the medical records have been revealed. All information, i.e, routes of delivery, clinical presentations, physical examination findings, methods of treatment, operative treatments, histological findings, results of treatment and recurrent diseases were analyzed.

Size of each mass was estimated by an average diameter which was a mean of three diameters. Volume of each mass was calculated from an average diameter based on an assumption that all masses were spherical. Analysis of the difference between each comparable group was performed by Student's *t* test. The statistical significance was p value < 0.05.

RESULTS

Thirty-five sacrococcygeal germ cell tumours (M 6, F 29) were diagnosed and treated in Siriraj Hospital between January 1974 and January 1999. All but three were full term newborns. No hereditary history was found. The routes of delivery were 27 vaginal delivery, 5 caesarian section (C/S) (due to 2 cephalopelvic disproportion, 1 previous C/S, 1 eclampsia and 1 unknown), one forcep delivery and two unknown delivery methods. Although most patients were born with big sacral masses (average diameter of 7.94 cm, S.D. = 2.94 cm, a range of 2.3- 14.0 cm), dystocia occurred only on three occasions requiring two caesarian sections and one forcep delivery. With regard to the relationship between sizes of the mass at birth and methods of delivery, we found that the average diameter +, - standard deviation of masses of patients requiring intervention from dystocia (n = 3) were not different from those delivered vaginally (n =27) (7.72 +, - 2.76 cm *vs* 9.88 +, - 1.84 cm, respectively and p value = 0.0752).

All except two first presented with sacral masses recognized at birth. Two of these also had fluid leakage from the masses. One patient with a sacral mass presented with bleeding from a rupture of the mass. Although every sacral mass was noticed at birth, the average age at which patients were transferred and operated on was 349.15 days. One patient was detected by a palpation of an abdominal mass at the age of 5 months and received a laparotomy 7 days later. The last patient developed difficulty in urination at the age of 9 months but she was diagnosed and operated on 3 months later.

Most sacral masses were located centrally. Seven of them extended from the midline to the left and three masses deviated from the midline to the right. At the time of operation, the average diameter +, - standard deviation and average volume +, - standard deviation of masses were 8.68 +, - 3.34 cm and 475.44 +, - 473.12 cc res-pectively. Only one patient also had an associated anomaly which was a branchial sinus.

During the period of 25 years, there were 35 admissions for primary operations. From these, six patients required other admissions for treatment of recurrent diseases. Among 35 primary operations, 31 patients had mature sacrococcygeal teratomas removed. However, there was one mature teratoma recurrence. Interestingly, two patients who had sacral masses completely removed and the first histological sections confirmed mature teratomas grade 0, developed malignant teratomas intrapelvicly following the first operations at 10 and 24 months. These two patients died eventually from advanced diseases. The last four patients presented with malignant diseases initially. In order to demonstrate the natural history and treatment for all patients with sacrococcygeal germ cell tumours clearly, we categorized two patients who had malignant recurrences following

Table 1. History and clinical presentation of each group of patients before operation.

	Mature teratoma (n = 29)	Mature teratoma with malignant recurrence (n = 2)	Malignant teratoma (n = 4)	Total (n = 35)
	F 23, M 6	F 2	F 4	F 29, M6
Average age +, - std deviation of the first operation (day)	295.59 +, - 703.12 range 3-2932	123 range 90-156	564.50 +, - 447.05 range 8-1065	340.77 +, - 661.78 range 3-2932
Statistical significance			p = 0.4657 *vs* mature teratoma	
Clinical presentation				
1. Sacral mass	93.1% (27)	100.0% (2)	100.0% (4)	94.3% (33)
2. Abdominal mass	3.4% (1)	0	0	2.9% (1)
3. Difficulty in urination	3.4% (1)	0	0	2.9% (1)
Size of the mass at operation				
No. of recorded cases	n = 29	n = 1	n = 1	n = 31
1. Average diameter +, - std deviation (cm)	8.68 +, - 3.54 range 2.34-15	7.00	10.00	8.68 +, - 3.34 range 2.34-15
2. Average volume +, - std deviation (cc)	505.25 +, - 494.12 range 6.65- 1767.86	179.67	523.81	475.44 +, - 473.12 range 6.65- 1767.86
Consistency of the mass				
No. of recorded cases	n = 29	n = 2	n = 3	n = 34
1. Cystic	51.7% (15)	0	33.3% (1)	47.1% (16)
2. Mixed cystic and solid	41.4% (12)	0	0	35.3% (12)
3. Solid	6.9% (2)	100.0% (2)	66.7% (2)	17.6% (6)
Position of the mass				
No. of recorded cases	n = 29	n = 2	n = 3	n = 34
1. Extrapelvis	89.7% (26)	100.0% (2)	66.7% (2)	88.2% (30)
2. Intrapelvis	6.9% (2)	0	0	5.9% (2)
3. Intra/extra pelvis	3.4% (1)	0	33.3% (1)	5.9% (2)

mature teratoma excision and the last four malignant cases as separate entities. Primary history and clinical presentations of each group of patients are described in Table 1.

The average age of primary operations for mature teratomas was younger than for malignant teratoma (295.59 days *vs* 564.50 days, respectively). This difference was not statistically significant because the sample size of the malignant group was too small. The most common clinical presentation of either benign or malignant teratomas is a sacral mass. One patient, who presented with an abdominal mass, actually had a huge pelvic mature teratoma which extended beyond the hollow of the pelvis into the extraperitoneal cavity. This extensive tumour was the same scenario as one patient who presented with difficulty in urination. Malignant tumours tended to be solid (66.7%), whereas, benign teratomas were either cystic or mixed cystic and solid in consistency. Anecdotal evidence strongly suggested that a mass with combined cystic and solid components was probably malignant, which was not so in our case. All mixed cystic and solid masses were benign and had no malignant recurrence. One third of malignant tumours had an extension from the sacral region into the pelvic cavity, whereas, only 10.3 per cent of mature teratomas did so.

Primary surgery for each group of patients is shown in Table 2. Ninety-seven per cent of cases were completely excised. Only one of malignancy received 90 per cent incomplete excision of the cystic sacral mass. This patient was lost to follow-up. Twenty-seven mature teratomas either solely extrapelvis (n = 26) or combined intra/extra pelvic extension (n = 1), were resected by a sacral approach. One of this group had a benign recurrence at the sacrum 7.8 months later and was re-excised and eventually cured. The other two benign abdominal masses were removed completely by combined exploratory laparotomy and sacral incisions. There was no recurrence.

Fifty-seven per cent of primary operations had some complications. All complications

Table 2. Primary surgery for each group of patients.

	Mature teratoma (n = 29)	Mature teratoma with malignant recurrence (n = 2)	Malignant teratoma (n = 4)	Total (n = 35)
1. Complete excision	100.0% (29/29)	100.0% (2/2)	75.0% (3/4)	97.1% (34/35)
1.1. Sacral approach	93.1% (27/29)	100.0% (2/2)	100.0% (3/3)	94.1% (32/34)
1.1.1 Chevron incision	14	0	1	15
1.1.2. Elliptical incision	7	0	0	7
1.1.3. Transverse incision	5	0	0	5
1.1.4. Curve linear incision	1	1	0	2
1.1.5.Vertical chevron incision	0	1	0	1
1.1.6. Unknown incision	0	0	2	2
1.2. Abdomino-sacral approach	6.9% (2/29)	0% (0/2)	0% (0/3)	5.9% (2/34)
1.2.1. Midline incision	1	0	0	1
1.2.2. Pfannelstiel incision	1	0	0	1
2. 90% Excision (Sacral)	0% (0/29)	0% (0/2)	25% (1/4)	2.9% (1/35)

Table 3. Complications of the primary operation.

	Mature teratoma (n = 29)	Mature teratoma with malignant recurrence (n = 2)	Malignant teratoma (n = 4)	Total (n = 35)
Complications*	48.3% (14)	100.0% (2)	100.0% (4)	57.1% (20)
1. Recurrent disease				
1.1. Benign recurrence	3.4% (1)	0	0	2.9% (1)
1.2. Malignant recurrence	0	100.0% (2)	100.0% (4)	17.1% (6)
2. Wound infection, disruption	31.0% (9)	50.0% (1)	50.0% (2)	34.3% (12)
3. Bladder atony	6.9% (2)	0	25.0% (1)	8.6% (3)
4. UTI	6.9% (2)	0	25.0% (1)	8.6% (3)
5. Diarrhea	3.4% (1)	0	0	2.9% (1)
6. Pneumonia	3.4% (1)	0	0	2.9% (1)
Coccyx excision				
No. of recorded cases	26	1	0	27
Coccyx excised	73.1% (19)	100.0% (1)	0	74.1% (20)

* includes recurrent diseases

are shown in Table 3. The most common complication was wound infection leading to partial wound dehiscence (34.3%). Eventually, all wounds healed gradually after treatment with warm sitz bath and antibiotics. This type of infection did not lead to sepsis in our series. Bladder atony (8.6%) and urinary tract infection (8.6%) occurred following extensive operations in the pelvic either by sacral or abdominal approaches. Delayed dwelling of an urethral catheter for 2-3 weeks solved this problem in most cases. Failure to adequate drainage of urine postoperatively will precipitate sepsis from urinary tract infection. However, in our series, we had no occurrence of sepsis.

The details of all recurrences (n = 7) either benign or malignancy are described in Table 4. Patients who had benign tumours removed and later developed malignant recurrences were categorized separately from the benign tumour group. (Table 1, 2). Two girls had sacral mature teratomas removed at the age of 156 and 90 days. Ten and 24 months later, they had recurrent solid intrapelvic masses. At the stage of recurrent detection, one already had lung and peritoneal metastasis and died shortly before any adjuvant treatment was added. The other had attempted radical re-excision of the pelvic tumour. At this stage, her vagina and uterus were also removed. The operation was

futile and she suffered from both locally advanced malignancy and a complication of the operation, urethral stricture. Although chemotherapy (Carboplatin, VP 16, Bleomycin) was instilled, she died eventually. All details of malignant teratomas are been demonstrated in Table 4 and 5.

Although many complications occurred, no one died directly from the operative procedure. Six malignant cases (2 recurrences from benign, 4 malignancy treated initially) had grim survivals. Three died from advanced diseases. Two cases who were lost to follow-up, were expected to expire but we have no record of this.

DISCUSSION

Although sacrococcygeal teratoma is the most common germ cell tumour in neonates, it is relatively rare. The incidence is approximately 1 in 40,000 live births[1]. Each paediatric surgical centre has its own limited number of cases. These tumours are much more common in girls, with the female to male ratio being at least 3 to 1. In our series, the ratio was 4.83: 1. Familial sacrococcygeal teratomas are reported sporadically, but in this series, no familial history was identified.

Logic dictates that a large sacrococcygeal teratoma that produces a probable risk of dystocia during labor may benefit from a caesarian section. Although an abdominal delivery is recommended for lesions greater than 5 cm[2,3] to avoid dystocia, tumour rupture or haemorrhage into the tumour which might occur with vaginal delivery, this recommendation is debated because the incidence of dystocia and other complications derived from a vaginal delivery are rare. The infrequency of sacrococcygeal teratoma typically precludes an accumulation of sufficient experience in any one center to complete a randomized trial to evaluate the true benefit from caesarian section[4]. In our series, sizes of tumours did not correlate with the probability of dystocia requiring obstetric intervention. The average diameters of teratomas requiring intervention for dystocia (n = 3) and those delivered vaginally (n = 27) were 7.72 cm and 9.88 cm respectively.

Diagnosis of a protuberant sacrococcygeal teratoma can be obtained easily from its characteristic appearance. The tumour protrudes from the space between the anus and coccyx and is usually covered with normal intact skin. If the tumour extends cephaladly through the bony pelvis, a

Table 4. Recurrent diseases.

Case	Recurrence	Year	Sex	First operation	First histological section	Age of 1st op (day)	Recurrent interval (month)	Recurrent presentation	Second operation
1	Benign	2524	F	Excision	Mature teratoma	9	7.8	Sacral mass, fluid leakage	Re-excision
2	Malignancy	2519	F	Excision	Mature teratoma	156	10	Solid intrapelvic mass	Nil
3	Malignancy	2536	F	Excision	Mature teratoma	90	24	Solid intrapelvic mass	Re-excision**
4	Malignancy	2513	F	Excision	Unknown	1,065	1	Solid sacral mass	Nil
5	Malignancy	2517	F	Excision*	Embryonal carcinoma	8	23	Solid sacral mass	Re-excision
6	Malignancy	2525	F	90% Excision	Endodermal sinus tumour	730	0	Cystic sacral mass	Nil
7	Malignancy	2542	F	Excision	Endodermal sinus tumour	455	2	Solid sacral mass	Re-excision

* includes colostomy
** includes hysterectomy and vaginectomy

Table 5. More details of recurrent cases.

Case	Age of 2nd op (day)	Second histological section	Third operation	Age of 3rd op (day)	Metastasis	Adjuvant therapy	Complication	Result
1	243	Mature teratoma	Nil		Nil	Nil	Wound infection	Cure
2		Nil	Nil		Lung, peritoneum	Nil		Death
3	1,096	Malignant teratoma	Cystostomy***	1,131	Nil	Chemotherapy****	Advance disease	Death
4		Unknown	Nil		Lung, lymph node	Nil	UTI	Loss F/U
5	698	Embryonal Carcinoma	Biopsy	743	Lung, liver	Radiation	Haemoptysis	Death
6		Nil	Nil		Nil	Radiation		Loss F/U
7	515	Endodermal Sinus Tumour	Biopsy	568	nil	Chemotherapy****	Wound infection	Survive

*** due to stricture urethra
**** Carboplatin, VP 16, Bleomycin

suprapubic mass may be palpable. Symptoms of urinary tract obstruction, which one patient in our series presented with, indicate an extension of the tumour into the presacral space. Most malignant teratomas have substantial solid components, whereas, mature teratomas are predominantly cystic or mixed cystic and solid. Mature teratomas are not often purely solid (6.9%).

As the frequency of malignant change in sacrococcygeal teratomas has been documented to increase proportionately with age, early diagnosis and excision of these lesions are essential. The frequency of malignant transformation increases from 10-20 per cent in neonates to 67 per cent in patients over 2 months of age. In our series, all extrapelvic sacral masses (n = 33) were recognized at birth. However, the patients were referred and operated on at the mean age of 349.2 days. Basically, the age when patients have malignant tumours removed should be older than the age of benign tumour removals. In this study, the average age of operation +, - standard deviation for malignant tumour removals (n = 4) was higher than for benign removal (n = 29) (564.5 +, - 447.1 days *vs* 259.6 +, - 703 days, respectively). However, the difference shows no statistical significance because the sample size of malignant cases is too small (n = 4).

Tumorigenesis of sacrococcygeal teratomas remains poorly understood. Recently, ras family of oncogenes, fos and jun oncogenes and nm23 and p53 tumor suppressor genes were identified in congenital sacrococcygeal teratomas indicating a possible role of genesis alteration leading to tumour development[5].

Complete surgical excision, which can be accomplished mostly by sacral incision (94.1%), is the initial advisable therapy for all patients with sacrococcygeal teratomas. If the tumour extends cephaladly through the hollow of the bony pelvis into the abdomen, an abdominal incision (5.9%) may be necessary to mobilize the upper portion of the teratoma and to interrupt the main blood supply to the tumour from the median sacral artery. For a massive sacrococcygeal tumour, ligation of the median sacral artery prior to tumour removal has been advocated to reduce the risk of fatal haemorrhage occurring during the operation. Moreover, cardiac failure from the profound shunt passing through the giant tumour can be controlled before the excisional operation. Many methods

have been proposed including an aortic snare[6], laparoscopic clipping of the median sacral artery[7] and hypothermic perfusion with extracorporeal membrane oxygenation[8,9]. In our centre, with formal meticulous dissection, there was no morbidity from massive haemorrhage during excision of these tumours. Resection of a coccyx concomitant with tumour excision reduces the possibility of recurrence.

If the tumour is composed of benign mature tissue, surgical excision is adequate therapy and long- term cure is anticipated. However, sacrococcygeal teratoma, although histologically benign, has an alarming potential to recur either as a benign or malignant tumour during the first 3 years of life[10-12]. Close follow-up for at least 3 years is recommended for all patients who have undergone excisions of sacrococcygeal teratomas[10-12]. All patients should undergo regular perineal and rectal examinations. Examination at 2 months, 4 months, and then at 6- month intervals for at least 3 years, is advisable. Although postoperative increase in serum alpha-fetoprotein (AFP) levels is an indicator of malignant recurrence, its concentrations may be difficult to interpret since it may normally be high in the newborn period due to fetal production[13]. Plain abdominal films can detect calcification foci indicating recurrent disease but magnetic resonance imaging appears to be a better modality for assessing a recurrent tumour, its invasion and metastasis[14]. When any recurrence is found, malignancy should be suspected [12] and the mass has to be promptly re-excised.

Rarely, a malignant recurrence is encountered in a patient whose primary tumour was reported as benign. Most reported cases were endodermal sinus tumours. This scenario occurred in 2 of 31 of our benign mature teratomas. Two girls, who initially had benign sacral masses excised completely, had malignant solid tumours recurring intrapelvicly following the first operation at 10 and 24 months. One developed lung and peritoneal metastasis and died shortly from advanced disease. The other died from advanced malignancy although all modalities of treatment were attempted. The details of these cases are shown in Table 4 (case 2, 3). Whether benign tumours undergo malignant degeneration from genetic alterations[15] or small residual malignant foci not identified at the time of initial examination[16] is still problematic.

In the past, the survival of malignant sacrococcygeal germ cell tumours was dismal especially a recurrent one. Prior to 1993, despite every adjuvant method provided, the eventual prognosis of patients with malignant sacrococcygeal teratomas was less than 50 per cent[17]. All malignant cases admitted to Siriraj Hospital before 1993 died or were expected deaths after losing follow-up. Now aggressive multiagent chemotherapy has made it possible to increase the survival. Previously, the BEP chemotherapy protocol (cisplatinum, bleomycin and etoposide (VP 16)) was employed. Recently the treatment of choice for malignant sacrococcygeal germ cell tumours is the JEB regime (carboplatin, bleomycin and etoposide (VP 16)) which is given for 4 courses or continued for 2 courses beyond a documented Complete Response. Excision of the primary tumour and coccyx should be done in all cases even if Complete Responses have been documented. Combined therapy by chemotherapy and surgery has improved the survival rate[11,18].

Wound infection leading to wound disruption is the most common complication (34.3%) encountered after sacral excisions. However, most cases can be treated successfully by warm sitz bath and local wound care. Surprisingly, it did not lead to fecal incontinence.

The incidence of voiding dysfunction post sacrococcygeal teratoma removal is high and usually neurogenic in origin. One series[19] reported a 12 per cent incidence of neurogenic bladder, comparable to 8.6 per cent in our series. The major cause of neurogenic lower urinary tract dysfunction seems to be related to surgical trauma of the pelvic plexus and sacral nerves[20]. The other associated urological complications, most of which are reported in children with malignancy, include vesicoureteral reflux, ureteral and urethral obstruction[19] which occurred in one of our malignant cases. Although preoperative hydronephrosis, which appears to be related to poor bladder emptying and high intravesical pressure, is common, it resolves spontaneously after tumor resection[21].

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ประสบการณ์ 25 ปีในการรักษาเนื้องอกซาโครคอคซิเจี้ยล เทอราโตมา

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ผู้ศึกษาได้ทำการศึกษาย้อนหลังในผู้ป่วยเนื้องอกซาโครคอคซิเจี้ยล เทอราโตมาทั้งสิ้น 35 ราย (เป็นชาย 6 ราย และเป็นหญิง 29 ราย) ซึ่งได้รับการรักษาในโรงพยาบาลศิริราชระหว่างปี พ.ศ. 2517 ถึงปี พ.ศ. 2542 ถึงแม้จะเป็นที่แนะนำกันว่าก้อนเนื้องอกที่มีขนาดมากกว่า 5 เซนติเมตรควรจะได้รับการผ่าตัดคลอดโดยทางหน้าท้อง ผู้ศึกษาพบว่าขนาดของก้อนไม่ได้มีความสำคัญในการกำหนดวิธีการคลอด ก้อนที่คลอดติดทั้ง 3 รายนั้นไม่ได้มีขนาดใหญ่กว่าก้อนที่สามารถคลอดทางช่องคลอดปกติซึ่งมี 27 รายเลย ผู้ป่วย 33 รายให้ประวัติว่ามีก้อนบริเวณก้นกบมาตั้งแต่แรกคลอด ผู้ป่วยรายหนึ่งมาด้วยเรื่องก้อนในท้องและอีกรายหนึ่งมาด้วยปัญหาการถ่ายปัสสาวะลำบาก การผ่าตัดเอาก้อนเนื้องอกออกโดยสมบูรณ์สามารถกระทำได้เก้าสิบเจ็ดเปอร์เซนต์ของผู้ป่วย โดยการผ่าตัดทางก้นกบ 32 รายและการผ่าตัดร่วม ทั้งทางหน้าท้องและทางก้นกบอีก 2 ราย ผู้ป่วย 6 รายได้เกิดมีก้อนเนื้องอกขึ้นใหม่ ในจำนวนนี้ 1 รายเป็นเนื้องอกไม่ร้ายซึ่งสามารถผ่าตัดเอาออกไปได้โดยง่าย ผู้ป่วย 2 รายได้เกิดเนื้องอกมะเร็งขึ้นใหม่ แม้ว่าผลการตรวจทางพยาธิวิทยาของชิ้นเนื้อแรก ได้ยืนยันว่าก้อนเนื้องอกซึ่งได้รับการผ่าตัดตอนแรกนั้นเป็นเนื้องอกไม่ร้าย ผู้ป่วยทั้ง 2 รายนี้ถึงแก่กรรมจากมะเร็งแพร่กระจาย ผู้ป่วย 4 รายสุดท้ายมาด้วยก้อนมะเร็งตั้งแต่แรกวินิจฉัย ผลข้างเคียงของการผ่าตัดที่พบได้บ่อยได้แก่ การติดเชื้อของแผลผ่าตัด การไม่สามารถบีบตัวขับปัสสาวะของกระเพาะปัสสาวะ และการติดเชื้อในระบบทางเดินปัสสาวะ สาเหตุการตายที่สำคัญก็คือการแพร่กระจายของเนื้องอกมะเร็ง ในการศึกษานี้ไม่มีผู้ป่วยรายใดเสียชีวิตจากตัวหัตถการโดยตรง

คำสำคัญ : ซาโครคอคซิเจี้ยล เทอราโตมา, เทอราโตมา, อิมมาทัวร์เทอราโตมา, เนื้องอกถุงไข่แดง, เนื้องอกเอนโดเดอมัลไซนัส, เนื้องอกเยิมเซลล์, เนื้องอกที่ก้นกบ, พยาธิวิทยา

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