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Unusual cause of defecation disturbance: a presacral tailgut cyst

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Abstract. – BACKGROUND: The aim of this study was to provide an overview of the literature on Tailgut cysts (TGCs) arising in the presacral space.

MATERIALS AND METHODS: We present a new case of presacral TGC and a literature review of English-language studies of presacral TGCs, accessed through the PubMed and Google Scholar databases. Keywords used were presacral or retrorectal tailgut cyst, presacral mucus-secreting cyst, retrorectal cystic hamartoma, retrorectal tumor, vestigial retrorectal or presacral cyst, and presacral cystic tumor.

RESULTS: A 29-year-old woman presented to our Clinic with defecation disturbance caused by a presacral TGC. Our literature review resulted in the inclusion of 94/111 articles and 155/332 described cases (129 women, 26 men; age, 0-80 years) of presacral TGC in this study. Although most patients presented with complaints such as rectal bleeding, rectal fullness, perianal pain, constipation, and pain reflected to the back, some asymptomatic cases were identified incidentally and others were detected during the investigation of atypical complaints such as pilonidal abscess, sinus, vaginal obstruction, and perianal abscess. Malignant transformation was found in 47/332 cases, including adenocarcinoma (n = 26); carcinoid tumor (n = 16); endometrioid, adenosquamous, and squamous carcinomas; sarcoma; and paraganglioma.

CONCLUSIONS: The high rate of malignant disease development from TGCs, which comprise a significant proportion of presacral masses; the development of significant postoperative recurrence, causing atypical conditions such as perianal fistula; and the high rates of infection due to partial resections make it essential to perform complete tumor resection with adequate margins.

Key Words:
Presacral space, Retrorectal space, Tailgut cyst, Retrorectal cystic hamartoma, Developmental cyst.

Introduction

The presacral (retrorectal) space is bound posteriorly by the sacrum, anteriorly by the rectum, superiorly by the peritoneal reflection, inferiorly by the levator ani and coccygeus muscles, and laterally by the ureters[1-6]. This region contains structures derived from the embryonic neuroectoderm, notochord, and hindgut. A variety of congenital, neurogenic, osseous, inflammatory, and miscellaneous disorders may occur within this space[1,5,6]. Each of these disorder categories can be further subdivided by more specific pathological characteristics.

Congenital lesions are by far the most common tumor type in this region, accounting for 55-70% of all presacral tumors. These tumors are present from birth because of the growth of embryological remnants, and they include cystic (developmental cysts, rectal duplication cysts, and anterior meningoceles) and solid (chordomas, teratomas, and adrenal rest tumors) lesions[5].

Developmental cysts, which account for 60% of all congenital presacral tumors, can arise from any embryonic layer. Based on morphology, developmental cysts are classified into epidermoid cysts, dermoid cysts, cystic teratomas, and tailgut cysts (TGCs). Epidermoid cysts result from a closure defect of the ectodermal tube and thus have a squamous epithelial lining. Dermoid cysts are similar defects, but they are made up of more mature elements and contain dermal appendages, such as hair follicles and sweat glands[5,7]. Teratomas are true neoplasms and contain tissue from each germ cell layer. They can be cystic or solid and often contain both components. Many teratomas possess germ-cell elements that are capable of malignant degeneration, and up to 10% of retrorectal teratomas progress to cancer if left untreated[5,6].

TGCs, also known retrorectal cystic hamartomas or mucus-secreting cysts, are rare benign cystic lesions of the presacral space[1-113]. Most TGCs occur in the presacral area, but cases have been reported in the perirenal, perianal, anterior rectal and posterior sacral regions; the anorectal junction; and the buttock and thigh[2,85]. Although

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the origin of TGCs is unclear, they are generally believed to arise from remnants of the embryonic hindgut. In human development from 35 to 56 days of gestational age, the embryo possesses a true tail and the primitive hindgut extends into this tail, which is caudal to the site of subsequent formation of the anus. This caudal extension is called the tailgut or postanal gut, and usually regresses completely by 56 days of gestational age. The persistence of this embryological remnant is very rare and results in the development of TGCs[23,32,38,41,70,72,76,90]. TGCs were first described by Middledorpf in 1885[5,48,76]. In 1928, Peyron investigated the embryological tailgut and concluded that a cystic tumor of tailgut origin must have an intestinal type of epithelial lining and lack a definite muscular or serosal coating. In 1938, Gius et al introduced the term "tailgut vestige," and in 1961, Edwards suggested the term "retrorectal cyst hamartoma." In 1987, Hjermstad et al suggested "tailgut cyst" because it is descriptive, unambiguous, and readily comprehensible[23,41,77]. TGCs are usually multicystic or multiloculated. The cysts are lined by a wide variety of epithelia, which varies among cysts or even within the same cyst. According to Hjermsad et al, the following histological criteria have been established for the diagnosis of TGC: (1) the epithelial lining of the luminal surfaces of the cysts must contain transitional and/or glandular-type (columnar) epithelium, with or without stratified squamous components; and (2) the underlying stroma must be composed of fibrous connective tissue containing scattered, discontinuous bundles of smooth muscle fibers, without a well-defined muscular layer containing a myenteric plexus and serosa[7,23,48,72,77,90]. Although radiological tools such as transrectal ultrasonography (TRUS), computed tomography (CT), and magnetic resonance imaging (MRI) provide sufficient information for the differential diagnosis of most presacral TGCs, pathological examination is always required to attain a definite and accurate diagnosis. The histopathological identification made by Hjermsad et al in 1988 has facilitated the differential diagnosis of TGC among many presacral lesions. Our primary aim in this study was to summarize the recent literature related to TGCs in the presacral region and to present the clinicopathological properties of these cysts. Our second aim was to examine the extent to which TGCs have exhibited malignant transformation.

Materials and Methods

In this paper, we present and discuss a new case of defecation disturbance caused by a TGC. We also conducted a review of the English-language medical literature in the PubMed and Google Scholar databases to identify every case report, series, letter to the editor, original article, and review related to presacral TGCs. Keywords used were presacral or retrorectal tailgut cyst, presacral mucus-secreting cyst, retrorectal cystic hamartoma, retrorectal tumor, vestigial retrorectal or presacral cyst, and presacral cystic tumor. The search included all articles published between 1932 and August 2011. Articles containing adequate information about patients' age, sex, and initial symptoms; cyst size and histopathological features; surgical approach; and recurrence were included in this study.

Results

Case Report

A 29-year-old woman was admitted to our Clinic with the complaints of defecation difficulty and a sense of rectal fullness. The patient stated that she often felt the need to go to the toilet, but could not fully defecate. She had undergone surgery due to a left ovarian cyst at another center 2 months previously, during which a mass was detected in the posterior rectum. The gynecologist at that Center performed no further operation after the ovarian cyst excision because he could not find a general surgeon to provide intraoperative consultation. Although physical examination yielded no pathological finding other than the Pfannenstiel incision, a mass that felt like the application of external pressure was palpated by digital rectal examination in the posterior rectum, 6-7 cm proximal to the anal verge. The laboratory findings showed a hemoglobin level of 15.1 g/dL, leukocyte count of 8,800/μL, platelet count of 248,000/μL, blood urea nitrogen level of 25 mg/dL, and creatinine level of 0.8 mg/dL, all of which were within the normal ranges. The serum levels of tumor markers (CA 15-3, CEA, CA 125) were also within normal ranges. Contrast-enhanced abdominal CT showed a 5 × 5-cm hypodense cystic lesion pressing externally on the rectum in the presacral region. MRI revealed that the cystic lesion was 5.5 × 5 cm in size and pushed the rectum anteriorly by external compression. The lesion

was isointense on the T1-weighted sequence and hyperintense on T2-weighted images of the presacral region with peripheral contrast enhancement (Figure 1). Colonoscopy provided a clear image of the mass compressing the external wall of the rectum. Biopsies of rectal mucosa collected during the colonoscopy yielded the finding of nonspecific colitis. Laparotomy was performed to completely excise the presacral mass. The patient developed no recurrence during the 2-year follow-up period.

Literature Review

The search of the PubMed and Google Scholar databases using the above-described criteria identified 111 article titles related to the subject of TGCs. Examination of the the abstracts and/or full text of these articles revealed that a total of 332 cases of presacral TGC was published since 1932. Seventeen articles presenting a total of 177 cases were excluded; 13 articles did not provide sufficient patient information and the full text of two articles was inaccessible[14,18,21,23,26,49,58,60,71,82,93,101,104,108].

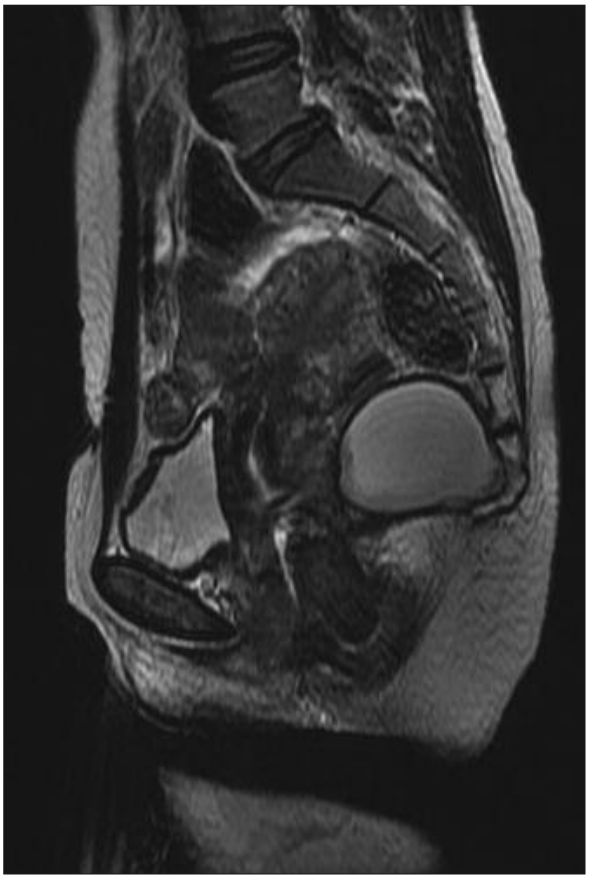

Figure 1. Sagittal pelvic magnetic resonance image showing the cystic tumor in the presacral space.

The clinical and pathological features of the 155 cases presented in the remaining 94 articles are summarized in Table I.

Discussion

The presacral space, also known as the retrorectal space, is defined anteriorly by the fascia propria of the rectum and posteriorly by the presacral fascia. It is laterally bordered by the lateral stalks of the rectum, the ureters, and the iliac vessels. The pelvic peritoneal reflection forms the superior border, and the levator ani and coccygeus muscles form the inferior border[7,77,80,108]. This space normally contains loose connective areolar tissue; the middle sacral, iliolumbar, and hemorrhoidal vessels; assorted lymphatics; branches of the sacral plexus; and, occasionally, isolated embryological cell rests.

Tumors occurring in the presacral space are uncommon and diverse. Although the majority of the literature on presacral tumors is limited to individual case reports, such tumors are responsible for an estimated 1/40,000 hospital admissions[5,6]. Presacral lesions may be classified as inflammatory (granuloma, fistula, perineal abscess, pelvirectal abscess), neurogenic (neurofibroma, ependymoma, ganglioneuroma, neurofibrosarcoma), osseous (osteoma, osteogenic sarcoma, sacral bone cyst, Ewing's sarcoma, giant cell tumor), miscellaneous (metastatic disease, liposarcoma, lipoma, fibroma, desmoid tumor, leiomyomas, fibrosarcoma), or congenital [chordoma, teratoma, anterior meningocele, developmental cyst (dermoid, epidermoid, rectal duplication, TGC)] lesions[5,6,77,108]. Congenital lesions generate more than half of all presacral tumors, and developmental masses generate more than half of all congenital lesions. Similarly, TGCs (= hamartoma/mucinous cysts) generate about half of all developmental cysts, and the rest are formed by dermoid/epidermoid cysts, teratomas, and rectal duplication cysts, in order of frequency[5].

Presacral TGCs are rare congenital developmental lesions that are thought to be derived from remnants of the embryonic postanal gut due to incomplete regression during development[4]. TGCs occur almost exclusively within the retrorectal or presacral space, and rarely in the perirenal area, the subcutaneous tissue in the anorectal region, the ischioanal fossa, and anterior to the rectum[77,93]. They can occur at any age and are three to four times more common in

Table I. Summary of the clinicopathological characteristics of 155 tailgut cysts reported in the English-language literature between 1932 and 2011.

References	Year	Age	Sex	Symptoms	Size cm-	Surgical approach	Tumor develop.	Recur.
Ballantyne	1932	38	F	Pain	1 Pint	NA	Adenoca	Yes
Gerwig	1954	27	F	Rectal discomfort	1	Posterior	No	No
		20	F	Perianal sinus	5*4	Excision	No	Yes
		31	F	Rectal discomfort	10*5	Posterior	No	No
		26	F	Rectal discomfort	4*4	Posterior	No	No
		37	F	Rectal discomfort	4	Posterior	No	No
Crowley	1960	37	F	Pain	5	NA	Adenoca	NA
Edward	1961	38	F	Rectal bleeding	UN	Transanal	No	No
		27	F	Constipation	1	Posterior	No	No
		35	M	Back pain	UN	Posterior	No	No
		34	F	Perianal fistula	2,5	Coccygectomy	No	No
Kraft	1962	34	F	Diarrhea	10	Posterior	No	No
Campbell	1973	30	F	Abscess	3	NA	No	No
Caropreso	1975	30	F	Asymptomatic	2*1.5	Transrectal	No	UN
Marco	1982	49	F	Perianal fistula	9*6	Posterior	No	No
		62	F	Painless mass	15*8.5	Parasacrococcygeal	Adenoca	No
Millis	1984	29	F	Asymptomatic	1	P.Transsacral	No	No
		57	F	Asymptomatic	3*2	P.Transsacral	No	No
		18	M	Acute abdomen	12*10	Abdominal	No	No
Abel	1985	25	F	Pilonidal cyst	4	Parasacrococcygeal	No	UN
		25	F	Asymptomatic	5	Parasacrococcygeal	No	UN
Ottery	1986	20	F	Menst disfonction	6*4	P.Transsacral	No	UN
		27	F	Asymptomatic	4	P.Transsacral	No	No
		UN	F	Asymptomatic	6.8*5	P.Transsacral	No	UN
Burchell	1987	19	F	Rectal pressure	UN	Transanal	No	No
Hood	1988	23-50	F:4 M:1	Rectal bleeding	UN	UN	No	UN
				Painful defecation	UN	UN	No	UN
				Constipation	UN	UN	No	UN
				Mass	UN	UN	Carcinoid	UN
				Mass	UN	UN	Carcinoid	UN
De Lange	1988	29	F	Asymptomatic	1*1	Excision	No	UN
		57	F	Asymptomatic	3*2	Excision	No	UN
		67	M	Urine retention	19*16	Excision	No	UN
Heij	1990	4	F	Constipation	UN	P.Transsacral	No	UN
Pyo	1990	38	F	Abdominal pain	10*8	LAR	No	UN
McDerm.	1991	52	F	Pilonidal abscess	7	Postanal	No	UN
Hutton	1992	40	F	Anal fissur	5*2	Postanal	No	UN
Lin	1992	18	F	Rectal pain	10*6	P.Transsacral	Carcinoid	UN

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Table I (Continued). Summary of the clinicopathological characteristics of 155 tailgut cysts reported in the English-language literature between 1932 and 2011.

References	Year	Age	Sex	Symptoms	Size cm-	Surgical approach	Tumor develop.	Recur.
Hannon	1994	19	F	UN	UN	P.Transsacral	No	No
		24	F	UN	UN	Transanal	No	No
		39	F	UN	UN	Abd.sacral	No	No
		59	F	UN	UN	Abd.sacral	No	No
Schnee	1994	62	M	Asymptomatic	18	Combine approach	Carcinoid	No
Gips	1994	76	F	Rectal fullness	10*8	Parasacrococcygeal	No	No
		60	M	Abd discomfort	15*10	Abdominal	No	No
Fujitaka	1995	72	M	Rectal bleeding	2.5*2	LAR	No	UN
Liessi	1995	75	F	Pain	UN	P.Transsacral	No	UN
		50	M	Asymptomatic	UN	UN	Adenoca	Yes
Wagner	1995	42	F	Rectal fullness	UN	Kraske op.	No	UN
Levert	1996	63	F	Pain while sitting	9	Parasacrococcygeal	Adenoca	No
		20	F	Abd pain, abscess	3	Parasacrococcygeal	No	UN
Roche	1997	23	F	Asymptomatic	17	Perineal	No	No
Kim	1997	49	F	Abd discomfort	4.5*3	P.Transsacral	No	UN
Jain	1997	39	F	Groin pain	24*12	Excision	No	No
Horenstein	1998	19	F	Pain,metrorragia	8	UN	Carcinoid	No
Lim	1998	40	F	Constipation	25*10	LAR	Adenoca	No
Maruyama	1998	66	F	Tenesmus	9*7	P.Transsacral	Adenoca	No
Rafindadi	1998	Neo.	M	Cystic swelling	7.9*7.6	UN	No	No
Williams	1998	4 mo	M	Poor feeding	2.5*2	P.Transsacral	No	No
Graad	1999	43	F	Imminent abort	13	Radial	Adenoca	Yes
Moulop.	1999	36	F	Recurrent TGC	19*12	APR	Sarcoma	UN
		70	F	Asymptomatic	UN	X		No
		80	F	Rectal pain	UN	Posterior	No	UN
Costello	2000	37	F	Mass	3	Abdominal	No	Yes
Thomas	2000	48	F	Anal Pain	UN	P.Transsacral	No	No
Schwarz	2000	47	M	Flank pain, const.	16	Abd.sacral	Adenoca	No
Umar	2000	74	M	Rectal bleed,fistula	1,5	Biopsy3	Squamousca	No
Prasad	2000	50	F	Rectal discomfort	UN	Excision	No	No
		36	F	Asymptomatic	9.5*9.2	Excision	Adenoca	No
		43	F	Painful bowel mov.	UN	Excision	No	No
		54	F	Rectal bleeding	2	Excision	No	No
		69	F	Rectal bleeding	4	Excision	Carcinoid	No
Oh	2000	Neo.	F	Mass	9	P.Transsacral	No	No
Oyama	2000	52	M	Diarrhea	22	Excision	Carcinoid	No
Verbeke	2000	25	F	Rectal pressure	7*3	Parasacrococcygeal	No	UN
		67	F	Mass	5*3	Parasacrococcygeal	No	UN

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Table I (Continued). Summary of the clinicopathological characteristics of 155 tailgut cysts reported in the English-language literature between 1932 and 2011.

References	Year	Age	Sex	Symptoms	Size cm-	Surgical approach	Tumor develop.	Recur.
Moreira	2001	64	F	Const,pelv press	12*10	Excision	Adenoca	No
		68	F	Rectal fullness	18*4	Excision	Adenoca	Yes
Yamaguc.	2001	32	M	Perianal fistula	UN	Pelvic evisceraiton	No5	UN
Satyadas	2002	34	M	P.Sinus	UN	Excision	No	No
Rao	2002	14	F	Asymptomatic	8*8	Excision	No	No
Mourra	2003	68	M	Perianal pain	1,7	P.Transsacral	NEC	No
Singer	2003	33	F	Rectal pain	UN	Excision	UN	UN
		45	F	Const, pain	UN	Excision	UN	UN
		49	M	Anal Pain	UN	Excision	UN	UN
		43	F	Draining wound	UN	Coccygectomy	UN	UN
Lev	2003	21-62	F:12	Asymptomat:3 Pain:7 Abscess:1		Anterior:3 Posterior:9	No	No
Ansari	2003	33	F	Urin retention	15*10	Abdominal	No	No
Song	2004	41	F	Perianal pain	2	APR	Carcinoid	No
Kanthan	2004	76	F	Pain,Tenesmus	6.5*4.5	P.Transsacral	Endom ca	No
Jacob	2004	42	F	Mass,Const.	12x11	Anterior	Carcinoid	UN
Antao	2004	Neo.	M	Rectal stenosis	1.3*0.7	Abdominal	No	No
Cho	2005	40	F	Perianal pain	10x8	Abd.sacral	Adenoca	Yes
Krivokapic	2005	47	F	P.Sinus	8,5	Abdominal	A.squamous	UN
Karcaaltin.	2005	38	F	UN	23*15	Abdominal	No	No
Piura	2005	45	F	Abd. discomfort	15*15	Abdominal	No	No
Skiadas	2005	48	F	Abd. pain, dysuria	11*10	APR	No	UN
Killings.	2005	25	F	Rectal pain,absc	9*7	P.Transsacral	No	No
Podber.	2005	2	F	Sacral dimple	2.6*1.2	Post.Sagital	No	UN
Menassa	2005	28	F	Hirsutism	10*8.5	Abdominal	No	No
Mathieu	2005	49	F	Rectal fullness	2,3	Excision	Carcinoid	No
Andea	2005	47	F	Painful mass	4*4	Excision	Adenoca	No
Sriganes.	2006	37	F	Vaginal obst.	6*5	Abdominal	No	UN
Doyle	2006	19	M	Back pain	17*16	Abdominal	No	UN
Gonul	2007	37	F	Back pain	3.5*2.5	Excision	No	UN
Tampi	2007	57	F	Back pain	12*10	Abdominal	Adenoca	No
Hall	2007	49	F	Rectal mass	2.5*2.3	Biopsy	No	UN
Buchs	2007	37	F	Rectal discomfort	6*5	Intersphincteric	No	No
		26	F	Mass	6*5.5	Intersphincteric	No	No
		57	F	Asymptomatic	7*5	Parasacrococcygeal	No	No
		21	F	Asymptomatic	7.5*6.5	Parasacrococcygeal	No	No
		37	F	Rectal discomfort	5*5	Intersphincteric	No	No
		33	F	Asymptomatic	6*4	Intersphincteric	No	No
		51	F	Pain	6*5	Intersphincteric	No	No
		39	F	Recurrent fistula	7*6	Parasacrococcygeal	No	No

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Table I (Continued). Summary of the clinicopathological characteristics of 155 tailgut cysts reported in the English-language literature between 1932 and 2011.

References	Year	Age	Sex	Symptoms	Size cm-	Surgical approach	Tumor develop.	Recur.
Zoller	2007	49	F	Rectal pain,absces	UN	Transanal EM	No	No
		55	F	Fistula	UN	Transanal EM	No	No
		59	F	Rectal pain,Const.	UN	Transanal EM	No	No
Galluzzo	2007	12	F	Back pain	12*11	Combine approach	No	UN
Rogers	2007	54	F	Abdominal pain	25*15	Abdominal	Adenoca	Yes
Garcia.	2007	38	F	Asymptomatic	10*8.5	P.Transsacral	No	No
Jarboui	2008	49	F	Perineal pain	15	Abdominal	Adenoca	No
Mayo	2008	43	M	Perineal swell.	25*14	Abdominal	Adenoca	UN
Mentes	2008	50	F	Asymptomatic	7*6	Post.Sagital	No	UN
Liang	2008	51	F	Hip pain	4.9*4.2	Abdominal	Carcinoid	No
Ipekci	2008	55	F	Rectal pain	12*12	Transcoccygeal	No	No
Mathew	2008	24	F	Back pain+Const	11*10	Posterior	No	UN
Gunkova	2008	28	F	Rectal pain	10*5.5	Abdominal,Lap	No	UN
Au	2009	24	F	Rectal discomfort	11*11	LAR+ileostomy	No	No
		60	M	Asymptomatic	UN	AR+ileostomy	No	UN
Zappa	2009	37	F	Rectal fullness	6.5*3.6	Abdominal	Adenoca	Yes
Peter	2010	72	M	Urinary symptom	15*10	Abdominal	No	UN
		19mo	M	Mass	UN	Excision	No	UN
Lu	2010	24	F	Difficult stool	12*10	Abdominal,Lap	No	No
Fujisawa	2010	68	M	Mass	25.7*15.4	APR	Adenoca	No
La Rosa	2010	73	M	Back pain	3.9*3.2	Abdominal, lap	No	No
Chong	2010	56	M	Melena	3.9*3.3	Transcoccygeal	P.ganglioma	No
Mueller	2011	27	F	Abdominal pain	7*6	Abdominal,Lap	No	UN
Lim	2011	36	F	Asymptomatic	4*3.8	Abdominal,Lap	No	No
Spada	2011	63	F	Back pain	4*3	Abdominal	Carcinoid	No
		41	F	Rectal pain	UN	Parasacrococcygeal+ Intersphincteric	Carcinoid	No
Deltour	2011	60	F	Rectal discomfort	11,8.3	Perineal	No	No

women than in men[40,63,70,77,80,88,90,93,97]. Half of all patients with presacral TGCs are asymptomatic and the cysts are found incidentally during routine examination. Digital examination can identify a palpable mass in the majority of patients, and is, thus, the most important, effective, and least expensive means of identifying presacral tumors[70,88]. Symptomatic patients present with constipation, rectal fullness, rectal pain and bleeding, painful bowel movements, change in stool caliber, lower abdominal pain, back and waist pain, and symptoms associated with urinary obstruction[67,97,105]. Some patients may present with recurrent retrorectal abscesses or following multiple procedures for anal fistulae, pilonidal abscess, or sinus.

Macroscopically, TGCs manifest as multi- or unilocular cystic lesions, several centimeters (1-25.7 cm) in diameter, with thin walls and glistening or inflamed linings. Microscopically, TGCs are characterized by cysts lined with variable types of epithelium, but the presence of some columnar or transitional epithelium is required to exclude epidermoid and dermoid cysts[2,40,93,97].

The differential diagnosis of presacral cystic lesions includes simple epithelial cysts, dermoid and epidermoid cysts, teratomas, rectal duplication cysts, chordomas, mucinous adenocarcinoma, cystic lymphangioma, and pyogenic abscess[38,40,76,80]. The differential diagnosis can be narrowed using a combination of diagnostic tools (digital rectal examination, rectoscopy, barium enema, TRUS, CT, and MRI) to reach a preoperative diagnosis of TGC[70,77]. Due to their location, almost all presacral tumors are palpable on rectal examination, and TGCs usually manifest as extrinsic masses[77]. Rectosigmoidoscopy can be of some value to determine the involvement of rectal mucosa and define the proximal extent of the lesion[5,77]. Radiologically, a well-defined, uni- or multilocular, thin-walled cyst is the most common feature, which cannot be differentiated easily from other lesions[72,77,85,88]. Barium enema examination is useful for the initial differentiation of a TGC from an intrinsic polypoid mass of mucosal origin and may reveal a fistulous tract in infected cases. However, the differentiation of a TGC from other diverse retrorectal tumors cannot be made using barium studies[40,41,72]. TRUS has been used to characterize the lesion as cystic and occasionally shows internal echoes due to mucoid or inflammatory debris[41]. CT scans show a well-defined homogeneous retrorectal mass, with a density ranging from that of water to that of soft tissue[41,48]. Keratinous or inflammatory debris within a cyst may result in a more solid appearance. Because calcification is not a feature of TGCs, its presence favors the diagnosis of a teratoma or malignancy. If malignant degeneration is present, CT scans may show a loss of discrete margins, calcification inside the lesion, and the involvement of contiguous structures. CT scans are useful for the definition of the upper extent of the lesion, which helps to guide operative therapy[5]. MRI reveals a hypointense lesion on T1-weighted images and a homogenously hyperintense lesion on T2-weighted images[1,2,4,36,41,48,72,100]. MRI is particularly useful in delineating soft-tissue planes and evaluating the presence or absence of bony invasion and nerve involvement. Such information, particularly the extent of sacral involvement, is vital for preoperative surgical planning[5].

The expressions "'rare" and "extremely rare" are frequently used in the literature to describe the development of malignancy from TGCs[7,23,24,48,52,67,80]. In my opinion, these words should not be used without an appropriate review of the literature. The first case of mucinous adenocarcinoma development from a TGC was described by Ballantyne in 1932[8]. In our search of the literature published between 1932 and 2011, we found that malignancy developed from TGCs in 47/332 (14.1%) patients. Twenty-six of the detected tumors were mucinous adenocarcinomas, 16 were carcinoid tumors, and one each was adenosquamous carcinoma, squamous carcinoma, endometrioid carcinoma, sarcoma, and paraganglioma. Thirty-nine patients with determined malignancy were included in this study (Table I)[2,7,8,10,16,23,24,31,33,36,38,42-44,47,48,52,53,56,57,63,67-69,72,73,80,81,86,91,93–95,97,103,107,110]. These results suggest that malignancy developed from TGCs in at least 14% of cases, and show that the expression "rare" used in many articles is misleading. In my opinion, surgical planning should include the radiological identification of TGCs and the principles of classical cancer surgery should be observed because this rate of malignancy is much higher than a statistically negligible rate.

According to our literature review, 9/155 (5.8%) patients developed recurrence within 5 months to 3 years after surgery[8,9,36,47,50,57,72,91,103]. Adenocarcinoma developed from a TGC in seven of these patients, and the remaining two patients had no malignant disease. Recurrence was observed in seven of the 39 (17.9%) cases of malignancy, all of which were adenocarcinoma; this finding indicates that TGCs should be removed at the adequate margins of the lesion, in accordance with cancer surgery protocol.

For lesions in the presacral space, complete removal of the mass is the most appropriate approach for a definite diagnosis and appropriate treatment. The performance of preoperative biopsy for diagnostic purposes remains controversial. Biopsy can be performed only in unresectable cases or in local, advanced-stage tumors to arrange the adjuvant therapy. In such cases, the use of a CT- (rarely, MRI-) guided extrarectal and presacral approach is recommended. An abdominal or transrectal approach is not recommended because of the risk of malignant transformation. Abdominal biopsy collection risks the shedding of tumor cells into the peritoneal cavity and their spread throughout the tract[1,6,52,63,88]. If the patient's pathological results after biopsy suggest a malignant disease, the removal of the biopsy tract during surgery is required.

Due to the potential for infection, the recurrence of perianal fistulas, and variable rates (14-25.1%) of malignant transformation, the complete surgical excision of these presacral masses is extremely important[7,67,70,77]. Three different operative approaches for the resection of presacral masses are commonly used: an abdominal or anterior approach, a transsacral or posterior approach, and a combined abdominosacral approach[5,88]. The most important parameters used in the selection of surgical procedure are the mass size, location, and relationship to surrounding structures. In the anterior (abdominal) approach, an open or laparoscopic method can be used. This approach is suitable for large tumors that have invaded surrounding tissue and are located proximal to the sacral promontory. The advantages of the anterior approach include better access to the pelvic structure, ureters, and iliac vessels. The posterior approach is most appropriate for tumors that develop in areas that can be accessed by rectal examination. This approach is recommended for small, benign masses that have not invaded surrounding tissue and are below the S4 level. The most frequently used methods in the posterior approach are para- or transsacrococcygeal, inter- or transsphincteric, transsacral, transanorectal, and transvaginal. The combined approach is commonly used for large tumors and those that descend both proximally and below the S4 level. The benefits of the combined approach include improved visualization of structures, such as the ureters and the rectum, via the anterior incision, as well as enhanced exposure of the nerve roots provided by the posterior approach[5,77,88].

Conclusions

The high rate of malignant disease development from TGCs, which comprise a significant proportion of presacral masses; the development of significant postoperative recurrence, causing atypical conditions such as perianal fistula; and the high rates of infection due to partial resections make it essential to perform complete tumor resection with adequate margins.

Conflict of Interest

None to declare.

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