# **Tailgut cysts: MRI findings**

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Abstract Magnetic resonance imaging (MRI) features of 11 surgically resected pelvic tailgut cysts were analyzed with reference to histopathologic and clinical data. Homogeneity, size, location, signal intensity, appearance and presence of septa and/or nodules and/or peripheral rim and involvement of surrounding structures were studied. Histological examination demonstrated 11 tailgut cysts (TGC), including one infected TGC and one TGC with a component of adenocarcinoma. Lesions (3–8 cm in diameter) were exclusively or partly retrorectal in all cases but one, with an extension down the anal canal in five cases. Lesions were multicystic in all patients but one. On T1-weighted MR images, all cystic lesions contained at least one hyperintense cyst. The peripheral rim of the cystic lesion was regular and non or moderately enhancing in all cases but the two complicated TGC. Nodular peripheral rim and irregular septa were seen in the degenerated TGC. Marked enhancement of the peripheral structures was noted in the two complicated TGC. Pelvic MRI is a valuable tool in the preoperative evaluation of TGC.

**Keywords** Tailgut cyst · Retrorectal cyst · Hamartoma · MR study

#### Introduction

# Materials and methods

A tailgut cyst (TGC) is a rare developmental lesion thought to arise from the embryonic postanal gut, well described by Hjermstad and al. at the Armed Forces Institute of Pathology in a series of 53 cases [1]. Since this initial description, reports in the radiology literature have been limited [2–6], the largest series by Yang et al. consisted of five patients [7]. TGC are often fortuitously discovered. Early accurate diagnosis, localization and characterization of the cyst lesion will affect the surgical management. When a tailgut cyst is suspected, excision is important because of the unwanted, life-threatening complications in long-standing cases.

MR is a useful technique to evaluate pelvic disorders because of its multiplanar imaging capability and its good soft tissue contrast. We retrospectively evaluated the MRI features of tailgut cyst in 11 patients with correlation to pathological findings to further characterize the MR imaging findings encountered in pelvic tailgut cysts.

# Patients

Between June 2001 and June 2007, 22 consecutive patients with a preoperative suspicion of perirectal cystic mass were operated on in our hospital. A retrospective review of the pathology reports of these patients identified 16 with tailgut cysts. Among these, 13 had been imaged with MRI, but MRI was of suboptimal quality in two cases that were thus excluded. We retrospectively reviewed the clinical, radiological, and histological features of 11 patients with pelvic tailgut cyst, according to the French law on clinical studies. Eight patients were women, and three were men (range 25–67 years, mean age 40.8). The clinical findings at presentation included chronic pelvic or perirectal pain (n=7), constipation (n=2), symptoms due to recurrent perirectal abscess and/or fistula (n=3) and no apparent symptom (n=2). Digital examination of the rectum revealed a ruberry mass located posteriorly or laterally to the rectum in ten patients (91%). Only two patients had an abnormal anorectal region with a perianal draining sinus (n=1) or a posterior sacral dimple (n=1). Laboratory studies demonstrated elevated C-reactive protein level in one patient.

#### Imaging

All 11 patients underwent preoperative MRI; postoperative MRI was performed in one case. All MR examinations were achieved on a 1.5-T unit using a phased-array pelvic coil, thin-sections, either in our department (Siemens, MR Magnetom, Erlangen, Germany, n=8) or in other institutions (n=3). No rectal preparation nor rectal distension was performed. MR examination protocol included sagittal (n=9), axial (n=10) and coronal (n=3) T2-weighted images. Imaging parameters for T2-weighted sequences on our MR unit were: TR/TE, 5940/122; echo-train length 16, field of view 25 cm, 4 mm slice thickness, no interslice gap,  $240 \times$ 256 matrix, three excitations. Unenhanced gradient-echo T1weighted (TR/TE, 636/13; field of view 25 cm, 4 mm slice thickness, no interslice gap, 230×256 matrix, three excitations) were performed in the axial plane (n=11) and coronal plane (n=2) and followed by axial gradient-echo T1weighted images either with fat-suppression (n=9) or not (n=2), after intravenous administration of 0.1 mmol/kg of gadodiamide (Omniscan, Nycomed Imaging, Oslo, Norway).

### Image review

Radiological images were analyzed by consensus of two experienced abdominal radiologists. They were not aware of the pathological reports, but were aware of the diagnosis of tailgut cyst. The following features were analyzed: lesion location and extent, lesion size, presence of numerous cysts grouping together in the same location (multicystic lesion) or presence of one cyst, signal characteristics intensity of the main cyst with regard to muscle on both unenhanced T1-weighted images (T1WI) and T2-weighted images (T2WI), appearance of lesion surface and peripheral rim, homogeneous or heterogeneous appearance of the lesion, presence of internal nodules or vegetations or enhancing septa. The appearance of the surrounding fat, muscles and bones was also recorded. These features were selected from the previous case reports and our personal experience [2-8].

All tumors had been surgically resected, either through a posterior surgical approach (n=8) or through a combined posterior approach and anterior diverting colostomy (n=2), or during abdominoperineal resection in the case of the TGC with the component of adenocarcinoma. The MR imaging findings were compared to the histopathological features of the lesions. The lesions could not always be analyzed precisely for the characterization of the internal

fluid as the cysts had been at least partly emptied before their examination in the department of pathology. Microscopically, lesions were diagnosed as tailgut cysts accordingly to the literature [1] by experienced pathologists subspecialized in the GI field.

#### Results

The clinical, MRI and pathological findings in patients with tailgut cysts are summarized in Table 1.

Histopathological study demonstrated 11 tailgut cysts, including 1 infected cyst and 1 complicated by a welldifferentiated mucinous adenocarcinoma. The latter occured in a context of chronic fistula in ano. Cysts were filled with a content that varied from clear to yellowish pasty substance and at least some evidence of mucinous internal component was recorded in the main cyst in eight cases.

MRI finding study demonstrated that lesions were primarily located in the retrorectal space in ten cases with a downward and lateral extension to the ischioanal fossa in five cases (Fig. 3) and to the anal canal in five cases. TGCs were developped primarily in the ischio-anal fossa in one case. The rectum was displaced anteriorly or laterally in six patients, owing to a mass effect from the cystic lesion. The mean largest diameter of the lesions was of 5.7 cm (range 3-8 cm). Cysts were numerous (more than two) in all patients but one. Multicystic lesions consisted of a large main cyst, most often multiloculated (7/10) and associated with other smaller cysts grouping together in the same location, in an adjacent fibrous tissue, hypointense on both T1-weighted and T2-weighted images (Fig. 1). In eight patients (73%), some of the adjacent smaller cysts were adherent to the main one, resulting in a honeycomb pattern (Figs. 2 and 3).

As far, as non-complicated cysts are concerned, the cystic mass was either well-circumscribed (n=9/9) by a thin regular peripheral rim (7/9) or a thick (>1 cm) smoothbordered wall (2/9). The rim was hypointense on all sequences apart from three cases in which a moderate enhancement after gadolinium administration was seen (Fig. 4). This peripheral rim corresponded to a fibrous wall at pathology. The borders of the cystic lesions were irregular in the two complicated cysts, with a strong enhancement of the wall and of the peritumoral adipose and muscular tissues (Fig. 5). Septa of multiloculated noncomplicated cysts were always thin and regular, either non or mildly enhancing, whereas they were thick but regular in the infected cyst and irregularly thickened and markedly enhancing in the TGC complicated by an adenocarcinoma (Fig. 6). The signal of the tailgut cysts was mostly heterogeneous (8/11), either because of a difference in the signal intensity of the cysts content (2/11) or because of a heterogeneous content of the main cysts (6/11). Main cyst displayed high signal intensity on T1-weighted images in 8/11 patients (73%) and high signal intensity on T2weighted images in all cases. No evidence of fat was found

Patient/ age (year)sex	Symptoms	Localization	MR features	Pathology
1) 25/F	Lower abdominal pain, posterior sacral dimple	Retrorectal space extension to ischioanal fossa and anal canal	Multicystic and honeycomb pattern, well-defined hyperintense on T1W1, hyperintense on T2WI, non- enhancing wall	Tailgut cyst
2) 29/F	Lower abdominal pain	Retrorectal space, extension to anal canal	Multicystic and honeycomb pattern, well-defined, hyperintense on T1W1, hyperintense on T2WI, moderately enhancing wall	Tailgut cyst with abundant inflammatory reaction within the wall
3) 32/F	Lower abdominal pain, constipation	Retrorectal space	Multicystic and honeycomb pattern, well-defined, hyperintense on T1W1, hyperintense on T2WI, unenhanced after gadolinium	Tailgut cyst
4) 33/F	Chronic right sciatica, recurrent perirectal abscess (three previous drainages)	Retrorectal space, extension to ischio anal fossa	Multicystic and honeycomb pattern, irregular and thickened wall, isointense on T1W1, hyperintense on T2WI, enhancement of the wall and perilesional fat planes and adjacent structures	Infected tailgut cyst (Escherichia Coli)
5) 34/F	Lower abdominal pain, pain during defecation	Retrorectal space, extension to ischio anal fossa	Multicystic and honeycomb pattern, well-defined, isointense on T1W1, hyperintense on T2WI, moderately enhancing wall	Tailgut cyst
6) 36/F	Lower abdominal pain	Retrorectal space, extension to anal canal	Multicystic and honeycomb pattern, well-defined, hyperintense on T1W1, hyperintense on T2WI, moderately enhancing wall	Tailgut cyst with abundant inflammatory reaction within the wall
7) 37/F	Constipation	Retrorectal space, extension to anal canal	Multicystic and honeycomb pattern, well-defined, hyperintense on T1W1, hyperintense on T2WI, no enhancement	Tailgut cyst
8) 39/M	Anal pain, recurrent perirectal abscesses	Retrorectal space, extension to anal canal	Multicystic, well-defined, hyperintense on T1W1, hyperintense on T2WI, smaller para anal "distant cyst," no enhancement	Tailgut cyst
9) 56/F	Incidental detection	Retrorectal space, extension to ischioanal fossa	Multicystic, well-defined, hyperintense on T1W1, hyperintense on T2WI, no enhancement	Tailgut cyst
10) 61/M	Incidental detection	Right ischioanal fossa	Unilocular, well-defined, hyperintense on T1W1, hyperintense on T2WI, no enhancement	Tailgut cyst
11) 67/M	Perirectal abcess, anal fis- tula, anal pain	Retrorectal space, extension to ischioanal fossa	Multicystic and honeycomb pattern, intracystic vegetations, irregular and thick wall, hypointense on T1W1, hyperintense on T2WI, important pe- ripheral and internal enhancement, enhancement of the wall and perile- sional fat planes and adjacent structures	Tailgut cyst with com- ponent of mucinous adenocarcinoma

Table 1 Clinical MRI and pathologic findings of tailgut cysts





Fig. 1 Axial T2-weighted MR image in a 25-year-old female (patient 1). The multicystic retrorectal lesion consists of numerous cysts interspersed in a fibrous tissue (arrowheads). Note that the main cystic lesion displays heterogeneous signal

whenever fat-saturated images were available. Presence of intracystic enhancing vegetations was observed in the case of tailgut cyst with a component of adenocarcinoma (Fig. 6), with a good correlation with the pathological features. No evidence of calcified deposits was found, although in one case pathologic examination revealed small areas of calcification in the cyst wall.

## Discussion

Tailgut cysts are rare developmental lesions known to occur more frequently in middle-aged women (3/1), although cases at any age, including neonates, have been reported [1]. In our series, the average age of patients was 40.8 years and the majority of them were women (with a mean age of 35.2 years). The clinical symptoms in our patients were consistent with the current knowledge. TGCs are characterized as multicystic lesions lined by a variety of epithelial cell types. Hjermstad and Helwig in their review [1] required the presence of columnar or transitional epithelium to exclude dermoid or epidermoid cysts, and required the absence of myenteric plexus and serosa to exclude bowel duplication cysts. They are thought to be congenital lesions due to some developmental error during embryogenesis. TGCs are believed to be vestigial remnants of the tailgut [1, 9]. TGC are located predominantly in the retrorectal area, but cases in perirenal and subcutaneous areas have also been reported [10-14]. One tailgut cyst in our series was located in the ischioanal fossa. The only

TGCs involving the ischioanal fossa reported in the literature so far were large TGCs primarily developed in the retrorectal space with a downward and lateral extension to the ischioanal fossa as in four cases in our series [4, 5, 12, 15, 16]. However, the development of tailgut vestiges that may reside in the ischioanal fossa [13] can possibly lead to the formation of TGC in this space. A feature of clinical relevance was the rather frequent presence of small cysts extending down the anal canal, distant from the main retrorectal cyst. We indeed observed in one case for which postoperative MR imaging had been performed that the main cyst had been resected along with its neighbor smaller cysts, whereas the small cysts down the canal anal had been left in place. Even though the surgical removal of such associated cysts may be impossible, they have to be preoperatively identified with MRI because their persistence may explain recurrence or infection [17, 18].

The present series supports that MRI is suitable for preoperative evaluation of tailgut cyst. In agreement with a previous report [7], our lesions were rather large in size (average 5.7 cm). MRI improves tissue characterization because of its high contrast resolution between different tissue compartments. It allows detection of mucin and blood in a cystic lesion. However, signal intensity of



Fig. 2 Coronal T2-weighted MR image in a 37-year-old woman (patient 7) with a retrorectal TGC. Small cysts are grouped together in a honeycomb pattern (arrow) and are adjacent to a main larger cyst (arrowhead)



**Fig. 3** Coronal T2-weighted MR image in a 34-year-old woman (patient 5) demonstrates the lateral extension to the ischio-anal fossa of a retrorectal tailgut cyst (arrows). Note the honeycomb pattern (arrowhead)

mucinous fluid may vary depending on protein concentration. On T1-weighted images, the signal intensity may change from hypointense to hyperintense as protein concentration increases. On T2-weighted images, signal intensity of mucinous fluids can decrease from highly hyperintense to hypointense with increasing protein concentration and viscosity. This explains the variety of the signal intensity displayed by the cysts in our series, even in the same patient. However, a rather constant feature was the presence of high signal intensity relative to muscle in most of the main cysts (n=9). Mucinous or proteinaceous component of the cyst fluid demonstrated in most of the cases on histological examination was assumed to be the cause of this high signal intensity, and no blood product nor fat was found either radiologically or histologically within the lesions. An intermediate to high signal on T1-weighted images with or without hyperintense foci has already been reported in TGC [2, 3, 5, 6, 8, 18, 19]. Yang et al. [7] reported hypointense TGCs on T1-weighted images, but their standard of reference was the signal intensity of fat, whereas ours was that of muscle. The cystic lesion consisted of a unilocular cyst in only one case. Several reports [3, 19, 7, 6] have highlighted the common multicystic appearance of tailgut cysts. Less commonly, cases of unilocular cyst have been reported [1, 2]. A common feature in our series was the frequent presence of small cysts clustered together and adjacent to the main cyst,

thus resulting in a honeycomb pattern, an imaging finding confirmed at pathological examination. This finding of a large dominant cyst surrounded by a number of small irregular cysts with a diameter up to 15 mm has also been described at pathology in one case report by Kripocavic [20]. Kim et al. [3] describe in their report of one retrorectal TGC imaged with MRI a cystic lesion formed by small cysts grouped together in a honeycomb pattern. We presume that this feature may be suggestive of a tailgut cyst, although we did not compare TGCs to other developmental cystic lesions in this area.

Septas in multiloculated non-complicated TGCs were thin and presented a mild enhancement after IV administration of contrast material, in accordance with the literature [3, 5, 8]. The presence of a peripheral rim circumscribing the cystic lesions corresponded to a fibrotic



Fig. 4 Tailgut cyst in a 29-year-old woman (patient 2). Axial unenhanced (a) and fat-saturated gadolinium enhanced (b) T1-weighted MR images show a thin regular peripheral rim circumscribing a retrorectal cyst (arrows, a), displaying moderate enhancement (arrows, b)



Fig. 5 Infected TGC in a 33-year-old woman (patient 4). Axial fatsuppressed gadolinium enhanced T1-weighted MR image demonstrates a cystic lesion in the right ischioanal fossa with a thick enhancing border (arrowheads) as well as marked enhancement of the peripheral fat planes (black arrow) and muscular structures (white arrow)

rim with chronic inflammatory reaction at pathology. When the cyst wall was thick, cysts were interspersed in a dense fibrous tissue. Although peripheral inflammatory reaction was present in all cases, the peripheral rim did not enhance in six out of nine non-complicated TGCs. [8, 10, 21]. As far as non-complicated cysts are concerned, preservation of a fat plane between the adjacent tissue and/or the rectum was present in all cases but one. Although most TGCs are easy to resect because of a good cleavage plane, surgical resection may be difficult in cases of mutiloculated large lesions, infected cyst or rarely when the wall of the cyst is intimately related to the rectal wall muscle fibers, as in one non-complicated cyst in our series. Only four cases of such cysts involving the muscular layers of the rectal wall have been reported in the literature, and two of them were degenerated TGCs [2, 4, 12, 22]. Surgical approach depends on the location, size of the lesion and on the presence of complication. For most lesions, a posterior approach with removal of a portion of the coccyx will allow the best visualization and removal of the multiloculated cyst [1, 23, 24]. In the case of larger tumor, or when malignancy is suspected, a combined abdominalsacral approach may be used [23, 24, 25].

Differential diagnosis with other cystic lesions of the retrorectal space or of the ischioanal fossa is often difficult. Other developmental cysts (epidermal cyst, dermoid cyst, anal gland cysts and rectal cystic duplication) may have similar imaging characteristics although they are more often unilocular. The presence of fat content on fat-saturated images is suggestive of a dermoid cyst. Rectal duplication cysts are often communicating with the rectal lumen and anterior to the rectum [9]. Anal gland cysts have a lower location than tailgut cyst and are typically located in close approximation to the anal sphincter [8, 26]. Anterior meningocele is a well-defined unilocular thinwalled, fluid-filled lesion of the retrorectal space with a

stalk that may be seen communicating with the thecal sac. MRI is limited in detecting calcification, particularly when it would be helpful to exclude dermoid cyst or teratoma. Indeed, although peripheral calcifications have already been reported in tailgut cyst [3, 6, 8, 20], they are more frequently seen in teratoma, dermoid cyst and other tumors arising in the retrorectal space. Although our series is limited by the fact that we did not compare the MR findings of TGCs with those of other lesions that may occur in the same location, we believe that the diagnosis of TGC may be suggested if a multicystic lesion, with numerous small cysts adherent to the main lesion, displaying high signal intensity on T1-weighted images and with a fibrous wall, is present. However, this putative discrimination is of little practical importance because the treatment and diagnosis for developmental lesions is thorough complete surgical excision. Simple cyst excision or drainage would indeed lead to recurrence or infection.

The potential complications of TGC include infection with recurrent fistula, which can misdiagnose as a recurrent perineal abscess, and rarely, a malignant transformation [4, 25, 27].

Our series included one case of acutely infected cyst. Acute infection was suspected on the basis of the clinical, biological findings and of some MRI criteria, such as a markedly enhancing irregular peripheral wall and enhancing infiltration of the perilesional fat and muscles. Our description of an infected tailgut cyst imaged with MRI is the first detailed one in the literature. When the complicated lesion is discovered without any knowledge of an underlying tailgut cyst, differential diagnosis with an abscess is difficult. We assume it is possible mainly on the basis of the location of the lesion (either retrorectal or in the ischioanal fossa) and of its multicystic appearance particularly if small cysts are clustered around a main cystic lesion. A history of recurring pelvic abcesses, repeated operations for anal fistula with no underlying obvious disease should also raise suspicion for a retrorectal cyst [4, 19, 25, 27].

Differentiation between an infected tailgut cyst from an abscess is of importance because an abscess must be treated by a combination of drainage and antibiotherapy, whereas TGC needs to be surgically excised. Another possible complication of TGC is malignant degeneration. So far, fewer than 30 cases of degenerated tailgut cysts have been documented in the literature. Among them, few have been imaged and described with MRI [4, 5, 18, 28, 29]. Most of the degenerated TGCs are associated with mucinous adenocarcinoma, although some cases of carcinoid associated with tailgut cysts have also been reported [12, 21, 30, 31], as well as exceptional cases of sarcoma, endometrioid carcinoma and clear cell carcinoma. Our findings of a cystic lesion with markedly enhancing irregular wall, nodular thickening of the wall, enhancing cystic vegetations and markedly enhancing irregular septal walls are consistent with what has been described in the literature



**Fig. 6** TGC associated with mucinous adenocarcinoma (patient 11). Sagittal T2-weighted (**a**) and fat-supressed gadolinium-enhanced T1-weighted MR images (**b**) in a 67-year-old man. The cystic lesion displays heterogeneous high signal intensity on T2-weighted MR images (**a**), with irregular borders and septas (arrows), intracystic enhancing vegetations (arrowheads) and enhancement of the

peripheral planes (b). Photomicrographs of the cysts (c and d); original magnification  $\times 25$  (c) and  $\times 100$  (d), stain: hematoxylin and eosin, show a cyst containing some mucus (arrow) and lined with mucus secreting epithelium (arrowhead, c). The epithelium is partly dysplastic, and cancerous tubes are visible around the cyst. Papilla at higher magnification (d) is lined by mucus secreting cells (arrowhead)

[4, 5, 18, 29]. In addition, we observed marked enhancement of the surrounding fat planes and muscular structures. In our series, the TGC with a component of adenocarcinoma was associated with a fistula. Mucinous adenocarcinomas have been reported to arise from fistula in ano [32]. However, in our case, adenocarcinoma was clearly identified at pathological examination as arising from the TGC. Marked enhancement of the perilesional areas and of the lesion wall, nodular thickening of the wall as well as the presence of intracystic vegetations are not features of non-complicated TGC and are thus likely to suggest complication of a tailgut cyst. In such cases, biopsy is not advisable because it may not show the potential malignancy and because it carries a significant risk of spillage of tumor cells.

# Conclusion

Our series supports that MRI is a useful imaging investigation when a retro and/or peri anorectal cyst is suspected. If a multicystic mass with small cysts adherent to a large cyst and without any evidence of sacrococcygeal involvement is present, either in the retrorectal space or in the ischioanal fossa in a middle-aged woman, the diagnosis of tailgut cyst has to be considered. However, final diagnosis is based on pathological examination after surgical resection. MRI is a helpful technique to define the extent of the cystic mass, its relationship to the surrounding structures and also to demonstrate possible complications in order to choose the best surgical approach.

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