Review article

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Garengeot's Hernia: two case reports with CT diagnosis and literature review

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Abstract: Garengeot's hernia (GH) is defined as the presence of the appendix inside a femoral hernia. It occurs in 0.9% of femoral hernias and is usually an incidental finding during surgery. Its treatment is controversial and the aim of this article is to review the diagnostic methods and surgical considerations.

We report two cases diagnosed preoperatively by contrast-enhanced computed tomography (CT) and discuss the treatment options based on a review of the literature published in PubMed updated on 1 December, 2015.

Fifty articles reporting 64 patients (50 women, mean age 70 years) with GH were included in the analysis. Diagnosis was performed by preoperative CT in only 24 cases, including our two. The treatment of GH is emergency surgery. Several options are available laparoscopic or open approach: insertion of a mesh or simple herniorrhaphy, with or without appendectomy.

Conslusion: The preoperative diagnosis with CT can guide the choice of treatment. Appendectomy and hernioplasty should be performed via inguinotomy, if there is no perforation or abscess formation.

Keywords: Hernia, inguinal; Hernia, femoral; Amyand's hernia; Garengeot's hernia; Appendix; Appendicitis

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The Authors have no conflict of interest

1 Introduction

Garengeot's hernia (GH) is defined as the presence of the appendix in a femoral hernia [1]. Less common is the finding of an acute appendicitis inside the femoral hernia sac. Although some cases have been diagnosed preoperatively, it is usually found incidentally during repair. We report two cases with preoperative diagnostic contrast-enhanced computed tomography (CT) and discuss the surgical considerations through a literature review.

2 Case Report 1

An 86-year-old female came to the emergency department with a 5-day history of a right-sided groin mass which had appeared after an episode of vomiting. The patient reported no change in bowel habits and no further episodes of vomiting. Her medical history included arthrosis treated with anti-inflammatory medication. On physical examination, her vital signs were stable and she was afebrile. The abdomen was soft and non-tender, with a painful right groin mass on palpation (approximately 5x3cm). There was no erythema or crepitus, nor any rebound or rigidity. Laboratory data on admission were as follows: white blood cells 6.28 x10³/mm³, with 76.4% neutrophils, and C-reactive protein (CRP) 20.7 mg/L. Abdominal X- ray did not reveal any significant abnormality. A CT was performed and revealed a right femoral hernia with the vermiform appendix in the hernia sac and mural thickening. There were no signs of perforation or abscess (Figure 1).

An open exploration was chosen. Prior to surgery, the patient received 2g/200mg of amoxicillin/clavulanic acid i.v. and analgesia. Under general anaesthesia, the hernia sac was opened through a Gregoire incision and a phlegmonous appendix was found inside (Figure 2). An appendectomy was performed with a purse-string suture. The hernia sac was closed and the defect was repaired using a polypropylene mesh plug (Lichtenstein technique).

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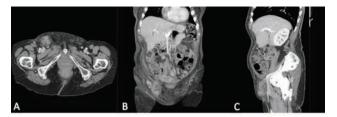


Figure 1: Case Report 1

Contrast-enhanced CT of the abdomen. (A) Axial view: tubular structure containing gas within the femoral hernia. (B) Coronal and (C) sagittal views: tubular structure extending from the caecal base.



Figure 2: Case Report 1 Inflamed appendix inside the femoral hernia sac.

Post-operative recovery was satisfactory, with three doses more of 1g/200mg amoxicillin/clavulanic acid i.v, and the patient was discharged on the third day. Histological examination of the appendix revealed acute appendicitis, as defined by infiltration of the appendicular muscularis propia by polymorphonuclear neutrophils. During the follow-up period of 20 months, the patient has not experienced complications or recurrence.

3 Case Report 2

A 77-year-old female was referred to the emergency department with diarrhea, vomiting and diffuse abdominal pain for two days. Her past medical history included hypertension, type 2 diabetes mellitus, dyslipemia, bronchial asthma, irritable bowel syndrome and hypothyroidism. On admission, the patient was haemodynamically stable and afebrile. Physical findings revealed a soft, non-distended

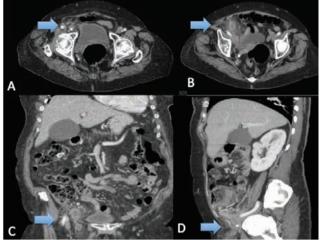


Figure 3: A,B,C,D Case Report 2. CT images Arrow: Appendix

abdomen with a tender mass in the right inguinal region measuring approximately 4x6cm. Laboratory tests showed a white blood cell count 12.30 x103/mm3, with 90.9% neutrophils, and CRP 2 mg/L. X- ray did not reveal bowel obstruction. A CT showed edematous appendix with an appendicolith inside it, in a right femoral hernia sac. The decision was taken to perform laparoscopic surgery. The patient received 2g/200mg of amoxicillin/clavulanic acid i.v. preoperatively. With general anaesthesia, an infraumbilical incision was made, and a pneumoperitoneum was obtained using Hasson's open technique. A 5-mm abdominal port was placed suprapubically and a 12-mm port in the left iliac fossa. A 10-mm, 30-degree telescope was used to examine the abdominal cavity. A large portion of the appendix, which had undergone necrosis and perforated, was seen to pass through the femoral defect from where purulent fluid was evacuated. The mesoappendix was stapled and transected with a tissue sealing generator, and the base of the appendix with a linear cutter stapler. The femoral defect was repaired primarily with an absorbable suture. Post-operative recovery was uneventful and the patient was discharged on the sixth postoperative day. Histology showed acute ischaemic and necrotic appendicitis with perforation. During eight months of follow-up, the patient has not experienced complications.

4 Discussion

Garengeot's hernia was first described in 1731 by Rene Jacques Croissant de Garengeot [1] and it accounts for 0.9% of femoral hernia repairs [2]. Moreover, acute appendicitis inside the sac is found in 0.08-0.13% of femoral hernias [3]. The first appendectomy in a GH was performed by Hevin

in 1785 [4]. The difference with regard to Amyand's hernia is that in the latter case the acute appendicitis is found in an inguinal hernia; it is named after the English surgeon Claudius Amyand, who performed the first appendectomy in an inguinal hernia sac in 1735 [4].

An unlimited literature search of the PubMed data base was performed on 1 December, 2015 with the following search criteria: ((Garengeot's hernia) OR (Amyand's hernia) OR (Hernia)) AND (Appendix). The search yielded 546 results. We initially identified 136 studies relevant to the subject, of which 81 were eligible for our analysis. The references of the selected studies were checked. We found 64 cases, 22 of them diagnosed on the basis of CT findings. The results of the review are listed in Table 1 [2,4,5-52].

Garengeot's hernia is a rare occurrence. Two possible aetiologies have been proposed. According to the first

Table 1: Cumulative presentation of cases included.

Author	Age	Sex	СТ	Surgery	Mesh	Appendix	
Shands AR et al (1903) [5].	29	F	No	Open	No	NA	
Gerami S et al (1970) [6].	71	М	No	Open	No	perforated AA	
Voitk AJ et al. (1974) [7].	59	F	No	Open	No	Enterocutaneous fistula	
Voitk AJ et al. (1974) [7].	76	F	No	Open	No	perforated AA	
Cutolo L et al (1978) [8].	78	F	No	Open	No	gangrenous and perforated AA	
Guirguis EM et al (1989) [9].	80	F	No	NA	NA	gangrenous AA	
Zissin L et al (2000)[10].	87	F	Yes	Open	Yes	perforated AA	
Isaacs LE et al (2002) [11].	76	М	No	Open	No	NA	
Mansari 0 et al (2002) [12].	60	F	No	Open	No	catharral AA	
Nguyen ET et al (2004) [13].	88	М	No	Open	No	perforated AA	
Barbaros U et al (2004) [14].	85	F	No	Open	Yes	gangrenous AA	
Fukukura Y et al (2005) [15].	54	F	Yes	Open	No	phlegmonous AA	
Priego P et al (2005) [16].	78	F	No	Open	Yes	AA	
Priego P et al (2005) [16].	83	F	No	Open	No	AA	
Priego P et al (2005) [16].	75	F	No	Open	Yes	AA	
Priego P et al (2005) [16].	75	F	No	Open	No	AA	
Priego P et al (2005) [16].	82	F	No	Open	No	Normal	
Priego P et al (2005) [16].	80	F	No	Open	Yes	Normal	
Akopian G et al (2005) [4]	81	F	No	Open	Yes	AA	
D´Ambrosio N et al (2006) [17].	71	F	Yes	NA	NA	perforated AA. Right hemicolectomy	
Comman A et al (2007) [18].	38	F	No	Laparoscopic	Yes	AA	
Gillion JF et al (2007) [19].	NA	NA	Yes	NA	NA	NA	
Sharma H et al (2007) [20].	NA	F	No	Open			
Sharma H et al (2007) [20].	NA	F	No	Open	4 Yes	1 perforated AA	
Sharma H et al (2007) [20].	NA	F	No	Open	3 No		
Sharma H et al (2007) [20].	NA	F	No	Open			
Sharma H et al (2007) [20].	NA	М	No	Open			
Sharma H et al (2007) [20].	NA	М	No	Open			
Sharma H et al (2007) [20].	NA	М	No	Open			
Maizlin ZV et al (2007) [21].	83	F	Yes	NA	NA	NA	

continued Table 1: Cumulative presentation of cases included.

Kibilw et al (2009) [22].NANANANANANANAChung A et al (2009) [23].83FNoOpenYesAAEbisawa K et al (2009) [26].77FYesBothNoAARing A et al (2009) [26].67NaNANANAShah A et al (2001) [27].46FYesBothNoAACaygill P et al (2011) [28].78FNoOpenNoAACaygill P et al (2012) [29].83FNoOpenNoAAAllen B C et al (2012) [31].76FYesOpenNoAASalkade Pt et al (2012) [31].73FYesOpenNoAASalkade Pt et al (2012) [31].73FYesOpenNoAACoskun K et al (2013) [33].65FYesNANAAASalkade Pt et al (2013) [34].66FYesNaNaAAHamcan Set al (2013) [35].66FNoOpenNoAAArdeleanu V et al (2013) [38].76NoOpenNoAABeysens M et al (2013) [38].76NoOpenNoAAAdeleanu V et al (2013) [38].76NoOpenNoAAAdeleanu V et al (2013) [38].76NoOpenNoAAAdaleanu V et al (2013) [38].76NoOpenNoAAAdaleanu V et al (2013) [38].76N							
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	García-Amador C et al. (2015)	77	F	Yes	Laparoscopic	No	perforated AA

NA= no available. AA=acute appendicitis.

theory, the appendix may be in an abnormal anatomical position owing to different degrees of intestinal rotation during embryological development, or to variations in caecal attachments. The second theory suggests that an anatomically large caecum forces the appendix into the pelvis, which therefore has a high risk of entering a hernial sac from the pelvic peritoneum [28]. The inflammatory process of the appendix is usually caused by the extraluminal obstruction of the appendix at the hernial neck, rather than by the more usual cause of intraluminal obstruction [10]. The obstruction of the neck leads to a vascular compromise which allows bacterial overgrowth [53]. Our case reports reflect the two presentations; in the first one, no obvious intraluminal obstruction was noted, and in the second an appendicolith was identified.

The female-to-male incidence ratio of GH is 5:1 with an age range from 29 to 91 years old (mean age 70). GH has been attributed to body changes during pregnancy. Other risk factors include increased intra-abdominal pressure, smoking, age and collagen disease. Most GHs occur on the right side [54]. Weakening of the transversalis fascia is also thought to play a role.

The clinical presentation usually takes the form of an incarcerated or strangulated femoral hernia and a painful groin mass. Abdominal pain, nausea, vomiting and diarrhoea are not usually reported. The narrowness and rigidity of the femoral canal usually prevents intraperitoneal spread of infection, and so there are no symptoms of peritonitis. Laboratory tests often show leucocytosis and elevated CRP [20]. X ray findings are usually nonspecific, but assist in recognizing small bowel obstruction if present.

In addition to inguinal hernia the differential diagnosis should include adnexitis, ectasia of the vena saphena magna, lymphomas, lipomas or other soft tissue tumors or a varix node [18].

Because of the rarity of the entity and the absence of the typical symptoms associated with acute appendicitis, achieving preoperative diagnosis is very difficult. Most patients are rushed to the surgical room with the inconclusive diagnosis of an incarcerated hernia. Most GH are diagnosed intraoperatively. Preoperative diagnosis by CT has been reported in only 24 cases, including the two case reports we present here. In our review, one more case was diagnosed using ultrasound [14].

In CT findings, GH should be considered when intramural air is present in an incarcerated femoral hernia sac without signs of bowel obstruction [24]. Therefore, a low-positioned caecum with a blind-ended tubular structure within the hernia sac and stranding of nearby fat on CT has been reported to have 98% sensitivity and specificity for the diagnosis of appendicitis inside a hernia sac [46].

The treatment of this disease is emergency surgery. Due to the rarity of the condition there is no standard procedure. The options available include laparoscopic or open approaches either with a mesh or simple herniorrhaphy, with or without appendectomy. In our review, most cases were performed via an open approach. Appendectomy via the hernial sac is considered appropriate; in case of perforation and abscess formation, a transabdominal access is preferred [24]. A combined approach was used in only three cases, all of which had been diagnosed preoperatively using CT. In two of them, the appendix was removed laparoscopically and the femoral hernia was repaired via a laparotomic approach [25,40]. In the other, laparoscopic drainage of the abscess and adhesiolysis were performed followed by a laparotomy for the appendectomy and herniorrhaphy [27].

Laparoscopy may be a valid technique for determining the condition of the hernia, but due to the difficulty of preoperative diagnosis it is unlikely to be the first choice for the surgical approach. In fact, in our review only three cases underwent laparoscopy [18,39,48], and only one of them had a preoperative diagnosis of GH [39].

In our two cases, the first one was treated via laparotomy and the second by laparoscopy. Both cases had been diagnosed preoperatively on the basis of the CT findings.

The appendectomy procedure is also controversial. It has been suggested that in the presence of a normal appendix appendectomy is not required. However, the surgery is not excessively complicated, and even in the absence of macroscopic inflammation the presence of microscopic inflammation from compression and ischemia within the hernia neck cannot be ruled out; for this reason, appendectomy should be performed [16]. In our review, the appendix was left in place in only one case [30].

Another controversial point is the use of a mesh, which was reported in only 55 cases in our review; in 35 a herniorrhaphy was performed. In the absence of abscess formation or perforation, the implantation of a mesh has been described as the hernia repair of choice [24]. In our first case, the surgery was performed immediately and there was no abscess formation in the hernia sac; a simultaneous appendectomy and primary hernioplasty using synthetic mesh was chosen. In the second case, as purulent fluid was evacuated, a herniorrhaphy was preferred.

The most important contributing factor to the increase in wound infection is delayed diagnosis [43]. The reported infection rates reached 29%, while severe complications such as necrotizing fasciitis and death were only rarely described [20].

5 Conclusion

GH is a rare condition which requires prompt treatment in order to avoid complications. Preoperative diagnosis with CT can indicate the correct management, that is, appendectomy via inguinotomy and hernioplasty if there is no perforation or abscess formation. More studies of procedures using the laparoscopic approach are required, especially in relation to the incidence of wound infection.

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