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# Acute Appendicitis in De Garengeot Hernia – A Systematic Review

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## Abstract

**Introduction:** Presence of appendix within a Femoral Hernia is a rare pathology named eponymously as De Garengeot's hernia that is mostly identified as an incidental finding during exploration for an incarcerated Femoral Hernia and even rarer is the incidence of acute appendicitis in these cases.

**Aim:** To undertake a systematic review of the published case reports, focussing on the incidence of acute appendicitis within a De Garengeot hernia, the relationship between pre-operative diagnosis and surgical technique, as well as the incidence and surgical outcome.

**Materials and Methods:** A PRISMA modelled literature search was carried out across PubMed, ProQuest, and BMJ Case Report databases with the search terms 'Femoral Hernia', 'Appendix', 'De Garengeot's' in various combinations, limited to English language, published between 1960 to 2019. Authors report another case report, not included in the systematic review.

**Results:** Systematic review identified 83 published data reporting 111 cases. 74 (66%) cases presented with painful groin swellings. Computerised Tomography performed in 43 (38%) accurately diagnosed De Garengeot's in 32 (74%) patients. The most common surgical approach was Lockwood's low approach 35 (31%) followed by the Lotheissen's approach 23 (21%). 81 (72%) underwent herniorrhaphy with non-absorbable sutures and 20 had mesh repairs (18%). Ten (9%) patients were reported to have postoperative morbidity with wound infection being the most common complication and one recorded death.

**Conclusion:** Rarity of De Garengeot's hernia is shown by the limited availability of data that restricted the author's ability to conclude a single diagnostic pathway or a specific surgical technique for this condition. CE CT is shown to be a relatively accurate form of imaging tool, as well as Ultrasound scans in the event that there was accessibility issue. Sequestered appendicectomy and hernia repair via laparoscopy could be an option; however, there is lack of data to warrant its effectiveness as the surgical option of choice over an open groin approach in the presence of an incarcerated groin swelling.

**Keywords:** Surgery; Colorectal; General Surgery

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## Introduction

Caecal appendix appearing inside a Femoral Hernial sac was first described by the French surgeon René Jacques Croissant De Garengeot [1] in 1731 and is eponymously named after him (in 2005 by Akopian G et al. [2]) and even rarer is the incidence of an appendicitis reported as low as 0.8% - 0.13% [3]. Femoral

Hernia occurs as a result of the protrusion of the peritoneal sac with/without abdominal contents through the femoral ring into the femoral canal, medial to the femoral vein and below the inguinal ring [4]. It is described as the third most common type of hernia, with a higher incidence in women (20%) than men (5%) [5]. The rigid and narrow femoral neck increases risk of strangulation (15% -20%) In rare cases, the appendix can travel

and be a content of the Femoral Hernia, reported in 0.5% - 5% of strangulated hernias.

De Garengeot Hernia remains an incidental finding and is often approached as a strangulated hernia prior to intervention. There is no prescribed diagnostic methodology either. A systematic review for this rare surgical condition was undertaken to identify the reported incidence of acute appendicitis in a De Garengeot hernia, tools used for pre-operative diagnosis, the surgical technique applied and their outcome.

## Literature Search

A literature search with terms 'De Garengeot', 'Femoral Hernia', and 'Appendicitis' was undertaken on PubMed database for any publication in the English Language, between 1960 and 2019. The search results returned studies published after 2005, given

that the term 'De Garengeot' was associated with the condition in 2005 [2]. Amending the search with only 'Femoral Hernia' and 'Appendicitis' identified earlier published case reports, with a total of 135 articles available with full text. Another search was run in ProQuest, with the terms 'Femoral Hernia', 'De Garengeot', 'Femoral Appendicitis', 'Appendix' and 'Appendicitis' with the search limited to English language peer-reviewed case reports published after 1970 that resulted in 68 published articles. A further manual search through the references of the published articles was also undertaken to find any further case reports. The results were screened for duplications and other factors as outlined in the PRISMA flow diagram (Figure 1).

A total of 83 studies that fulfilled the criteria were analysed for patient demographics, presentations, diagnostic evaluation tools, surgical approach, and post-operative outcome. Surgical approach

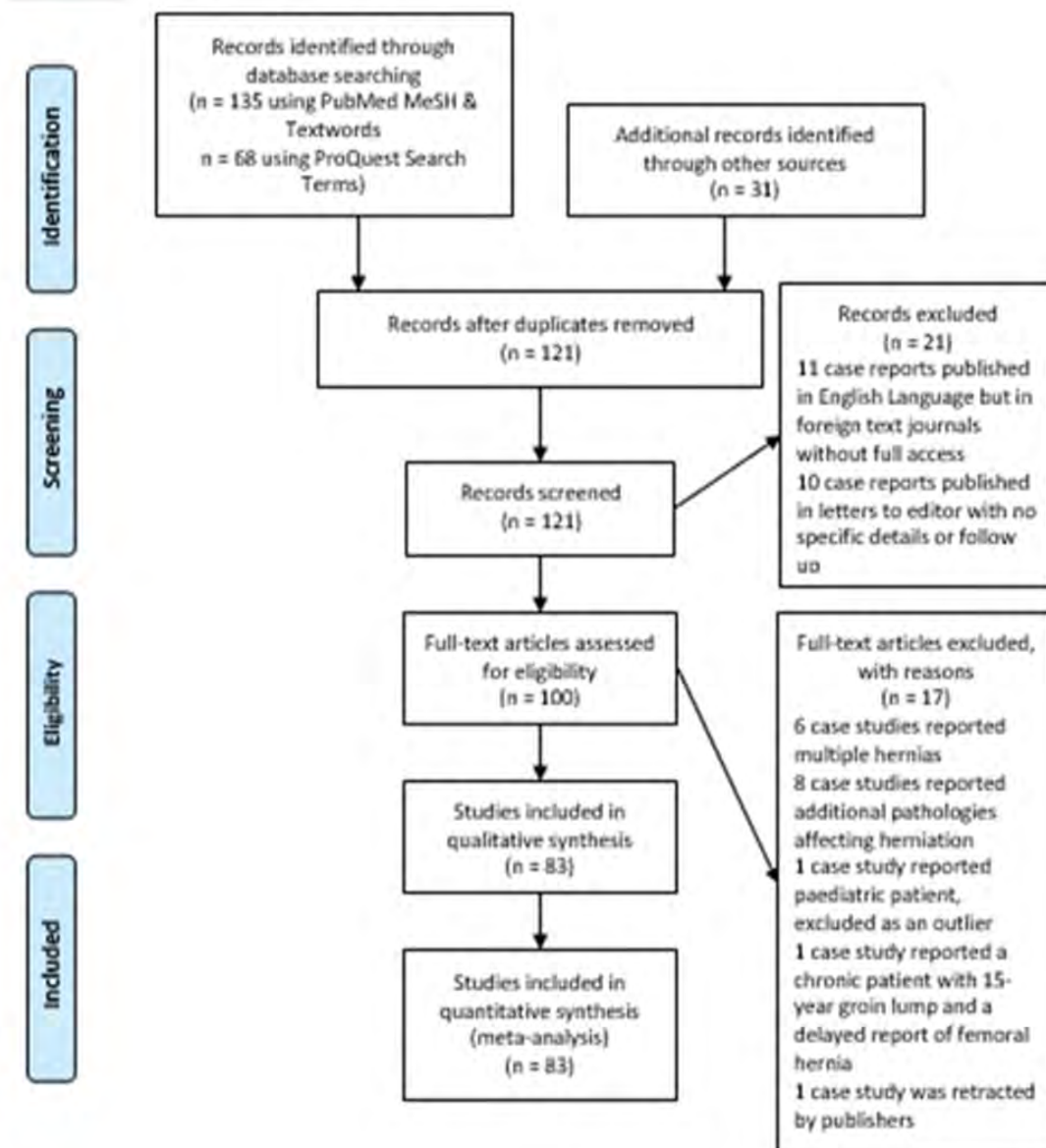


Figure 1 PRISMA flowchart.

was categorised in relation to the inguinal incision: Lockwood's infra-inguinal, Lotheissen's trans-inguinal and Mc Evedy's high approach [6], unless specific techniques were mentioned. Any incision away from the inguinal was categorised as a laparotomy.

Publication bias - Data taken from published case reports risks significant publication bias as there will be reluctance for publication for cases with unfavourable outcomes. To counter the publication bias, two of the authors ran independent searches and compared results. Manual searches were also done in 'Grey Literature' (other searches in PRISMA diagram), such as working papers and scholar publications in addition to the popular journals to increase available data. Only peer reviewed articles were included in the final search, excluding any articles that the authors could not get full access to.

## Results

Total 111 patients were reported across 83 published articles [7-87]. The author's case report is not included in the systematic review. Of the 83, 70 published articles were single case reports, while the remaining 13 involved multiple cases.

Voitk AJ et al. [7] reported one chronic inguinal sinus of over two years in the two case studies reported. The chronic case was excluded to avoid skewing of the data, with the other case in the study retained.

The results are compared by totals and means when possible. Data is collated in tabular forms to summarise the interventions.

The most common presentation among these case reports was sudden onset painful groin swelling in 74 (66%) patients within which 88% (n=65) had no associated nausea or vomiting; 15 (13%) patients presented with painless groin swelling as their primary presentation and only 2 (0.2%) presented with diarrhoea. Mean duration of the symptoms was 4 (0-7) days for patients with acute presentation. Eight patients reported symptoms of pain longer than 10 days, with one patient reporting pain for 42 days. Out of 111 patients, 103 patients were identified by gender and of these 103 cases, 81% were females.

Clinically, 79% of the patients (n=91) had presented with a groin swelling including 49 with associated erythematous skin changes. 46% (n=51) of the patients were febrile on admission and 4 had features of shock. Eight had undergone previous hernia repairs (**Table 1**).

Blood investigations were performed in 92 (82%) patients. In 47 (51%), there was reported leucocytosis and elevated CRP and the

remaining 45 (49%) showed normal blood picture. 71% (n=76) of the reported case studies had additional investigations, with some patients having undergone more than one investigation. 21 patients had X-ray taken, 43 patients Contrast Enhanced CT, 21 patients had an USS of the groin, 2 patients had MRI, and 1 patient had a Barium scan. In 18 (16%), decision to treat was based on clinical examination.

There is significant discussion around the establishment of preoperative diagnosis of De Garengeot's hernia and more so regarding the diagnosis of appendicitis within it. **Table 2** outlines the accuracy CE CT, when used, in establishing a pre-operative diagnosis in 74% cases. In the absence of discussion about CT imaging, it is assumed that CT was not used in the diagnostic work up.

74 studies have reported appendicitis in the Femoral Hernia. **Table 3** outlines the histological outcome of the appendix. Of the 87 cases that have reported the histopathology, 12 (14%) were normal appendixes within a De Garengeot's.

The surgical technique is most often determined by the diagnosis of Femoral Hernia. Of interest in this study were the subsequent appendicectomy and the implications of encountering De Garengeot's hernia, especially with an inflamed or infected appendicitis within. In the current collection of cases, most of the surgeries were determined by the surgeon's preference to access the hernia, with the Lockwood approach being the most employed (n=35) in this review. In 64% of the cases (n=72), the appendix was removed through the initial incision. **Table 4** outlines the initial surgical approach, the procedure for the subsequent appendicectomy performed and the surgical approach for hernia repair.

In retrospect, there is value in noting the impact of the pre-operative diagnosis on the approach decision. With a limited number of cases that have been diagnosed specifically as appendicitis within a De Garengeot's, **Table 5** compares the preoperative diagnosis to the impact of the surgical approach in whether the appendix could be accessed through the initial incision.

There was one reported post-operative death, and of the case reports with post-operative morbidity, 11 patients reported post-operative complications. Of these, one patient had mobility issues due to Parkinson's disease [20], unrelated to surgery and has been excluded. The post-operative death occurred in a patient (76/F) that presented with diabetic ketoacidosis. The mean post-operative stay was 5 (1-22) days.

**Table 1:** Initial presenting symptoms in reviewed publications.

Symptoms	Number of Cases*	%
Swelling in Right Groin	91	82%
Groin Pain at Presentation	81	73%
Fever	51	46%
Nausea at Presentation	26	23%
Abdominal Pain	14	13%
No Pain	16	14%

\*Some patients exhibited multiple symptoms. Each presenting symptom is counted per patient at initial observation.

**Table 2:** CE CT finding in reviewed data.

Diagnostic Imaging		Number of Cases
CE CT used for pre-operative diagnosis	Diagnostic	32
	Non-Diagnostic	11
	Total	43
CE CT not used for pre-operative diagnosis		69

**Table 3:** Histological outcomes in reviewed studies.

Histological Outcome	Number of Reported Cases
Normal Appendix [22,24,35,41,44,49,54,55,75]	12
Acute Appendicitis [1]	61
Not Reported	25
Perforated Appendicitis [2]	8
Necrotic Appendicitis [51,85]	2
Appendiceal Abscess [8]	1
Chronically relapsing Appendicitis [25]	1
Ischemic Gangrenous Appendicitis [16]	1
<b>Total No of Patients</b>	<b>111</b>

**Table 4:** Surgical approach and procedures in reviewed studies.

Hernia Repair/Appendectomy Approach/ Approach to Hernia		Number of Reported Cases
<b>Herniorrhaphy<sup>3</sup></b>		32
Appendectomy via initial incision	McEvedy Approach	3
	Laparotomy	4
	Lotheissen's approach	1
	King's College approach	1
	Laparoscopic Approach	1
Laparoscopic appendectomy	Approach Not Specified	4
	Lotheissen's approach	2
	Laparoscopic Approach	1
Laparotomy for Appendectomy	Approach Not Specified	1
	Laparoscopic Approach	1
No Appendectomy	Laparoscopic Approach	1
	Approach Not Specified	8
Hemicolectomy	Lotheissen's approach	1
	Approach Not Specified	1
Appendectomy Technique Not Specified	Approach Not Specified	3
<b>Hernia Repair by McVay Technique<sup>4</sup></b>		22
Appendectomy via initial incision	Lockwood approach	6
	Lotheissen's approach	5
	McEvedy Approach	2
	Approach Not Specified	1
Appendectomy via initial incision	McEvedy Approach	2
	Lotheissen's approach	1
Laparoscopic appendectomy	Laparoscopic Approach	1
	Laparotomy for Appendectomy	3
Lockwood approach	Lotheissen's approach	1
<b>Herniorrhaphy through low approach<sup>5</sup></b>		20
Appendectomy via Initial incision	Lockwood approach	12
	Lotheissen's approach	3

Hernia Repair/Appendectomy Approach/ Approach to Hernia		Number of Reported Cases
Appendectomy via Lanz incision	Lockwood approach	1
Laparotomy for Appendectomy	Lockwood approach	3
Incidental Appendectomy	Lockwood approach	1
<b>Mesh Hernia Repair<sup>6</sup></b>		20
Appendectomy via initial incision	Lockwood approach	8
	Lotheissen's approach	6
	Gregoire incision	1
	Approach Not Specified	2
Laparoscopic appendectomy	Laparoscopic Approach	1
	Lotheissen's approach	1
Laparotomy for Appendectomy	Approach Not Specified	1
<b>Herniorrhaphy through high approach<sup>7</sup></b>		4
Appendectomy via initial incision	Lotheissen's approach	1
	McEvedy Approach	2
Laparotomy for Appendectomy	McEvedy Approach	1
<b>TAP Femoral Hernia Repair<sup>8</sup></b>		2
Laparoscopic appendectomy	Laparoscopic Approach	1
	Laparoscopic TAP Approach	1
<b>Herniorrhaphy through Lichtenstein Technique<sup>9</sup></b>		1
Appendectomy via initial incision	Approach Not Specified	1
<b>TEP Hernia Repair<sup>10</sup></b>		1
Laparoscopic appendectomy	Laparoscopic TEP Approach	1
<b>Lockwood Repair of Femoral Hernia<sup>11</sup></b>		1
Appendectomy via Lanz incision	Lockwood approach	1
<b>De Oliveira Hernia Repair<sup>12</sup></b>		1
Appendectomy via initial incision	Lotheissen's approach	1
<b>Hernia Repair Not Specified<sup>13</sup></b>		7
Appendectomy Technique Not Specified	Approach Not Specified	7
<b>Grand Total</b>		<b>10913</b>
3,4 [12,14,24,25,30,52-50,63]		
5 [6,14,19,20,21,26,28,29,31,34,36,43,64-73]		
6 [17,18,36,74-76]		
7 [11,33,77]		
8 [78,79]		
9 [22]		
10 [80]		
11 [81]		
12 [82]		
13-Two case reports did not provide details about surgery		

## A Case Sample

An 86-year-old man presented with 48-hour history of progressive nausea, vomiting and right lower abdominal and groin pain. He had also not opened his bowels without passage of flatus for 24-hours. Medical history included hypertension, Chronic

**Table 5:** Pre-operative diagnosis vs. surgical approach in reviewed studies.

Pre-Operative Diagnosis and Surgical Approach		Number of Patients
No Preoperative Diagnosis	Appendicectomy via Initial incision	22
	Separate Incision for Appendicectomy	6
	Laparoscopic appendicectomy	3
	Not Specified	1
CE CT- De Garengeot's <sup>14</sup>	Appendicectomy via initial incision	19
	Separate Incision for Appendicectomy	7
	Laparoscopic appendicectomy	1
	Not Specified	5
USS- Femoral Hernia with Bowel Obstruction <sup>15</sup>	Appendicectomy via initial incision	6
	Separate Incision for Appendicectomy	2
	Laparoscopic appendicectomy	1
CE CT- Strangulated/ Incarcerated Hernia <sup>16</sup>	Appendicectomy via initial incision	6
	Laparotomy for Appendicectomy	1
AXR- No Bowel Obstruction <sup>17</sup>	Laparoscopic appendicectomy	1
	Appendicectomy via initial incision	3
	Laparotomy for Appendicectomy	2
AXR- Bowel Obstruction <sup>18</sup>	Appendicectomy via initial incision	5
	Laparoscopic appendicectomy	1
USS- De Garengeot's <sup>19</sup>	Appendicectomy via initial incision	4
	Separate Incision for Appendicectomy	3
	Laparoscopic appendicectomy	1
CE CT- Femoral Hernia, No Obstruction <sup>20</sup>	Appendicectomy via Initial incision	4
MRI- De Garengeot's <sup>21</sup>	Appendicectomy via initial incision	2
USS- Femoral Hernia, No Bowel <sup>22</sup>	Laparoscopic appendicectomy	2
Barium Scan- Appendico-Cutaneous Fistula <sup>23</sup>	Appendicectomy via initial incision	1
Total Number of Patients		111
14 [20,22,24,26,27,32,34,37,38,40,43,44,50,54,56,58,59,72,73,75,77]		
15 [19, 28, 31, 43, 49,55,61]		
16 [17, 46, 49, 50, 61,65,81]		
17 [11, 25,36, 52,83]		
18 [23,65,71]		
19 [21,22, 41,55,67]		
20 [21,43,45,49,58]		
21 [47,75]		
22 [24,43]		
23 [8]		

Obstructive Pulmonary Disease, Congestive Cardiac Failure, multiple chest infections, and a left hip replacement. Although WHO performance status was 3, his walking was limited to 100 yards with exertional dyspnoea.

Physical examination showed a mildly distended abdomen and an 8 × 6 cm tender swelling at the right inguinal ligament with no associated skin changes. Laboratory investigations revealed WCC>12 and CRP>70. Plain Abdominal X-Ray exhibited a few loops of gas-fluid filled small bowel, consistent with small bowel obstruction. A clinical diagnosis of obstructed Femoral Hernia was made. This was a high-risk case that was undertaken under spinal anaesthesia following discussion with consultant anaesthetist.

A high McEvedy approach was undertaken and following reduction of the Femoral Hernia, the sac revealed an inflamed long, thick, coiled appendix. Conventional appendicectomy was performed followed by the closure of the peritoneum. Femoral ring was approximated with 1 Ethibond sutures. Patient made a slow but uneventful recovery and was discharged home after 7 days. Histology confirmed acute appendicitis.

## Discussion

### Incidence

Current literature quotes incidence between 0.8% to 0.13% that was first reported by Ryan WH in 1937, following observation and



data collation of 8,692 cases of appendicitis with the appendix identified only in 11 (0.13%) hernia sacs [3].

### Pathogenesis

The exact pathogenesis of De Garengot's is still debatable. Femoral Hernias are four times more common in females [82], and more so in post-menopausal women. There is a school of thought that during embryological development differing degrees of rotation of the alimentary tract could result in the abnormal attachment of the vermiform appendix to the pelvis, resulting in a 'pelvic appendix' risking herniation through the femoral ring.

### Pre-operative diagnosis

Appendicitis within a Femoral Hernia does not usually present as a classical acute appendicitis, thus, limiting the scope of diagnosis (appendicitis independently). The detection of the irreducible hernia is, in itself, an indicator of surgery and historically literature reports exploration of the hernial sac confirming its contents. Powell HD et al. [83], reports pre-operative diagnosis of appendicitis within a femoral sac by examination alone and Voitk AJ et al. [7] were one of the first authors preoperatively reporting appendico-cutaneous fistula in a Femoral Hernia [8] in a chronic case of a groin swelling of 15 years.

Preigo et al. [35] observed that presentations in these cases are atypical and even computerised tomography was not accurate in all cases. However, Abdul-Ghaffar S et al. [62] report 100% sensitivity and 98.9% specificity for computerised tomography as a pre-operative diagnostic modality for acute appendicitis in De Garengot's hernia. Salkade PR [58] and Ikram S et al. [79] also report similar results with computerised tomography. Filatov J et al. [14] and Park HR et al. [84] provide supportive arguments in using ultrasonography as a diagnostic modality of choice, given its accuracy, cost effectiveness, and accessibility compared to computerised tomography or MRI [87]. Ultrasonography risks operator bias as the operator may not be trained to identify the hernial sac contents once the diagnosis of herniation is established. Halpenny D et al. [46] have reported successful use of MRI for establishing a pre-operative diagnosis.

Following review of the published literature in 2013, Kalles V et al. [85] noted computerised tomography to be diagnostic in 44% of the patients evaluated across 31 case reports. In this systematic review, computerised tomography showed 74% accuracy in diagnosing appendicitis within De Garengot's hernia.

From the data in the current study, the use of CT seems to provide pre-operative diagnosis, allowing the surgeon to make a more informed decision around surgical approach.

### Surgical technique

There is no consensus regarding the surgical approach to De Garengot's hernia. A major aspect of the decision making was affected by the pre-operative diagnosis and the surgeon's priority to explore the abdomen to prevent worsening of any incarceration. In cases where a pre-operative diagnosis of De Garengot's was established (with or without appendicitis), the approach was based on the need that both the appendix and the femoral ring

are accessible through single incision. The use of laparoscopy in the event of no pre-operative diagnosis is supported by Comman A et al. [77] to reduce incidence of postoperative ileus. The suitability of TAPP over TEP due to the potential benefit of evaluating intraabdominal and hernial contents is also noted, with Al-Subaie S et al. [86] favouring the use of TAPP over TEP as a diagnostic tool for a patient clinically diagnosed with an irreducible right Femoral Hernia. This is countered by Beysens M et al. [79] and Shiihara M et al. [71], who argue that due to the containment of the site of appendicectomy, a mesh repair can be done without risk of infection. There is discussion around the use of interval hernia repair, where the appendicectomy was initially done followed by the hernioplasty. Use of mesh is extensively debated across the literature, primarily for the risk of infection especially in a De Garengot's hernia with inflamed or infected appendix.

In the current review, of the 109 cases that specified the surgical techniques, the most common technique for hernia repair was suturing with non-absorbable material (n=86), and another 20 had mesh repair. The only reported post-operative complication in patients with mesh hernia repair identified was wound infection (n=3). When the decision for hernia repair was done without a mesh, the most common technique used were interrupted sutures (n=58) and McVay's repair (n=26).

Three case reports describe unusual surgical techniques. Mizumoto R et al. [26] chose the King's College approach where a single skin incision allowed exploration of the groin hernia and its contents and the subsequent entry into peritoneum for appendicectomy. De Oliveira technique for hernia repair as described by Couto HS et al. [81], explain that the choice was made due to its effectiveness in hernia repair without the need of mesh and its cost effectiveness. Lacaille-Ranger A et al. [76] used modified Nyhus technique, where they describe posterior approach to hernia repair by dividing the layers of the abdominal wall to expose the femoral ring from the preperitoneal space.

The post-operative outcomes show a 9.8% of the cumulative patients reporting post-operative complications, with seven patients reporting wound infection. The current study is limited in the ability to evaluate the relationship between the pre-operative surgical decision and the post-operative outcome. There is a lower incidence of post-operative morbidity (1%) in patients that have had an accurate pre-operative diagnosis; however, this is not conclusive given the limited number of patients (32) and the lack of a control group to compare [80-87].

### Limitations

The study is limited in that there is not enough data to establish a causal relationship between the pre-operative surgical decision and the post-operative outcomes. Without a structured case-control study, any correlational conclusions remain speculative.

### Conclusion

Appendicitis within a De Garengot's hernia often lacks a classic presentation. The irreducible hernia becomes the point of focus and an indicator for surgery and the contents of the sac are only detected after direct exploration. There have been cases where a

pre-operative diagnosis was established with the use of imaging, however there can be limitation based on their availability, cost, and operator training. A clinical scenario of irreducible, strangulated hernia warrants an urgent surgical exploration. However, if a pre-operative diagnosis can be established: laparoscopic exploration, sequestered appendectomy and

hernia repair could potentially be the appropriate option for the management of De Garengeot's hernia. Current literature review confirms the rarity of De Garengeot's hernia leading to the lack of a standardised operative technique and the authors feel the discretion for approach would depend on the surgeon's preference and available expertise.

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