

## Case Series

# Mucinous neoplasia of appendix: imaging and its relevance to management

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## ABSTRACT

Appendiceal mucocele is a rare pathology of the appendix that can present with non-specific symptoms. Therefore, imaging evaluation is essential for accurate diagnosis and proper management. To document importance of pre-operative imaging, we reviewed the scans, reports, and clinical data of nine patients with appendiceal mucocele who were treated at our institute. We also studied the literature on CT findings, histopathology, surgical management, prognosis, and recent guidelines for appendiceal mucocele. We suggest that pre-operative imaging can aid surgeons in selecting the most appropriate treatment plan for each patient, resulting in better outcomes. Certain critical imaging features, such as mural nodularity, irregular wall thickening, lymphadenopathy, cecal involvement, presence of free mucus, or pseudomyxoma peritonei, can alert surgeons to prepare for frozen section and an appropriate surgical approach. In conclusion, we recommend pre-operative imaging as a crucial tool for the diagnosis and management of appendiceal mucocele. Its use in identifying critical imaging features can help surgeons decide on the best surgical approach for each patient and ultimately lead to better outcomes.

**Keywords:** Appendiceal mucocele, Rare pathology, Imaging features

## INTRODUCTION

Appendiceal mucocele is a rare pathology of appendix. Most cases are identified during surgery or in post-surgical specimen. However incidence of incidental finding of appendiceal mucocele in imaging is increasing over the years with improving availability of CT scans. Appendiceal mucocele is a macroscopic descriptive term defined as dilated appendix with intraluminal accumulation of the mucin.

A variety of non-neoplastic and neoplastic conditions cause it. Management and prognosis is heavily depends upon its histopathological and imaging findings. Role of radiologists is crucial for identifying key features of appendiceal mucocele and additional findings which may

help surgeon to choose best for patients for better prognosis.

## CASE SERIES

A search of the radiology database at our medical centre from January 2015 to March 2023 was performed. We have diagnosed 15 patients as an appendiceal mucocele or possibility of forming mucocele. Among this 14 scans were performed at our centre, while one was sent for review. We retrieved the history sheets of all patients. Majority of them were presented with right lower quadrant pain mimicking acute appendicitis, one with abdominal distention and one case was incidental finding. Among these nine patients were operated at our medical centre, we retrieved pathological data of these patients. Five of them

were diagnosed as LAMN, two patients with non-neoplastic cause, one with pseudomyxoma peritonei and one with mucinous cystadenocarcinoma. Lap cecectomy was performed in three patients. One patient operated with lap. Right hemicolectomy with ileocecal and periappendiceal lymphadenectomy, one patient was planned for extensive surgery with right hemicolectomy along with cytoreduction plus HIPEC. Lap appendectomy alone was performed in four patients.

**Imaging interpretation**

We reviewed plain and contrast enhanced imaging series of all 15 patients at our dedicated workshop. We evaluated for maximal cystic diameter, length of appendix, wall

irregularity, mural-nodularity, attenuation of intraluminal content, periappendiceal fat stranding and lymphadenopathy, presence of calcification, internal septation, presence of free mucus, ascites, discontinuity of the appendiceal wall and invasion to the adjacent structures. In case of the presence of free mucus, we looked for the scalloping of any solid organs, serosal and peritoneal deposits, lymphadenopathy, distant metastasis and bowel obstruction. We had also used positive oral contrast in these patients for better visualisation of bowel serosal disease and disease in the adjacent mesentery and peritoneum.

Clinical, surgical and histo-pathological data with imaging findings of 9 patients are summarized in Table 1.

**Table 1: Observations.**

Size of appendix at the time of diagnosis (mm)	Co-existing appendicitis	Wall calcification	Adjacent mesenteric changes	Caecal thickening/base of appendix involvement	Lymph nodes	Mural nodularity and/or irregular wall thickening	Free mucus	Histopathology	Surgery
26	-	-	-	+	Peri appendiceal sub-centimeter sized	-	-	LAMN	Cecectomy
25	-	+	-	+	-	+	-	Mucinous cyst adenocarcinoma	Right hemicolectomy with lymphadenectomy
20	-	-	-	-	-	-	-	LAMN	Appendectomy
75x34	-	-	+	-	+	+	+	Mucinous cyst adenocarcinoma	Right hemicolectomy with cytoreduction+ HIPEC
50x27	-	-	-	+	-	-	-	LAMN	Cecectomy
13	+	-	+	-	Sub-centimeter sized	-	-	Non neoplastic	Appendectomy
24	-	-	-	-	-	-	-	Non neoplastic	Appendectomy
26	-	+	-	-	-	-	-	LAMN	Appendectomy

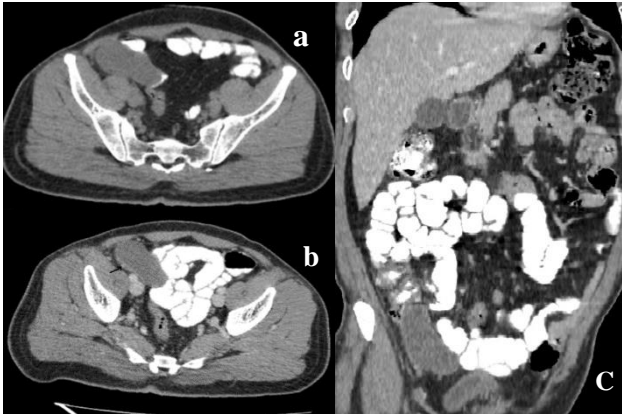
Note- LAMN- Low grade appendiceal mucinous neoplasia; HIPEC- hyperthermic intraperitoneal chemotherapy; +: present, -: absent.

**Case 1**

A 52 years old male patient presented with mild right sided lower abdominal pain. USG was done elsewhere, suggestive of well-defined tubular structure with anechoic content within. He was sent for further evaluation. CT

plain and contrast was performed after giving the oral contrast. It shows a well-defined right lower quadrant tubular cystic mass arising from caecum, measuring 26 mm in its diameter. Contrast enhanced scan shows rim enhancement. It has hypoattenuating content within. There is no evidence of peri-appendicular fat stranding and

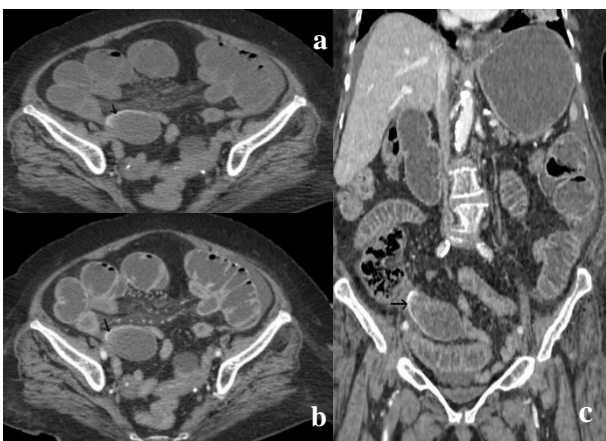
calcification of its wall. No evidence of wall irregularity or mural modularity. Features are suggestive of appendiceal mucocele. Patient was treated with laparoscopic appendectomy. It was diagnosed as low grade mucinous neoplasia on histopathology



**Figure 1: Case 1- axial unenhanced (a) contrast-enhanced; (b) CT scan images show tubular cystic right lower quadrant mass (black arrows). Coronal contrast-enhanced; (c) CT scan image shows right lower quadrant tubular cystic mass.**

**Case 2**

A 71 years old female with right lower quadrant pain with occasional nausea was evaluated with contrast enhance CT scan. Plain and post contrast CT images were obtained after giving the oral contrast. It shows tubular cystic mass in right lower quadrant with curvilinear calcification of its wall. No evidence of periappendicular fat stranding, wall irregularity, mural modularity or free mucin. Patient was treated with laparoscopic appendectomy alone. Post-operative histopathology confirmed it as low grade appendiceal mucinous neoplasia.

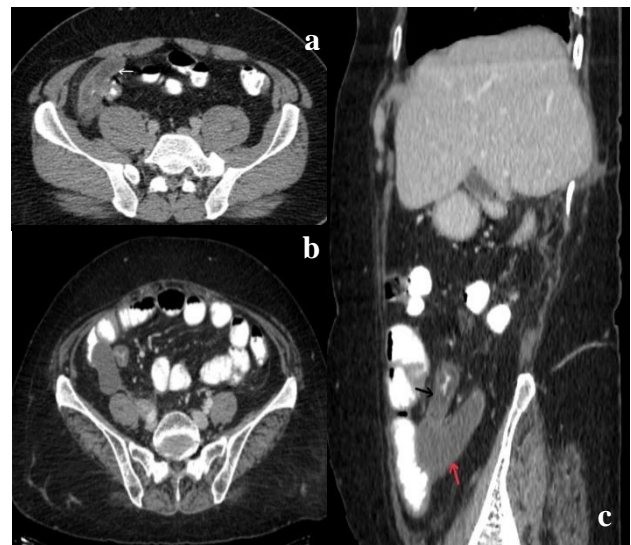


**Figure 2: Case 2- axial unenhanced CT scan image (a) shows tubular cystic mass in right lower quadrant with curvilinear calcification (black arrows); (b) axial; and (c) coronal contrast-enhanced CT scan images show thin enhancing wall (white arrows) of lesion.**

**Case 3**

A 52 years old female with lower abdominal pain with mild right lower quadrant tenderness and weight loss. Patient had past history of pulmonary Kochs. Plain and contrast enhanced CT scan was performed after giving oral contrast. CT images show low-attenuation tubular mass contiguous with base of cecum, along with thickened terminal ileum.

Patient was treated with laparoscopic appendectomy. Histopathology showed non neoplastic inflammatory etiology. Another case with the same finding was also treated with the laparoscopic appendectomy alone.



**Figure 3: Case 3- axial unenhanced (a) and contrast-enhanced; and (b) CT scan images show low-attenuation tubular mass (red arrows) contiguous with base of cecum, along with thickened terminal ileum. Sagittal contrast enhanced (c) CT scan image shows relation of base of cecum and lesion.**

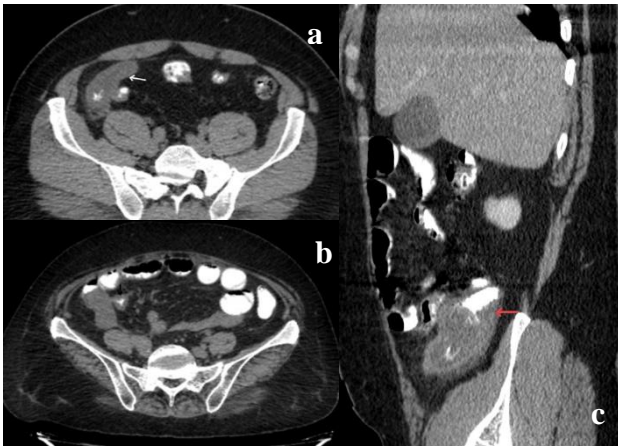
**Case 4**

A 42 years old male presented with mild right lower quadrant tenderness and occasional pain. CT abdomen pelvis was advised. Plain and contrast enhanced CT scan was performed after giving the oral contrast.

CT scan images show dilated appendix with thickened and enhancing appendicular wall. There is involvement of the base of caecum by irregular wall thickening of the base of the appendix. Surgeons were informed to prepare for frozen section. Patient was treated with cecectomy. Frozen section and post-operative histopathology has confirmed the diagnosis as low grade mucinous appendiceal neoplasia.

Two other cases with CT findings of cecum involvement in absence of lymphadenopathy and free mucin were

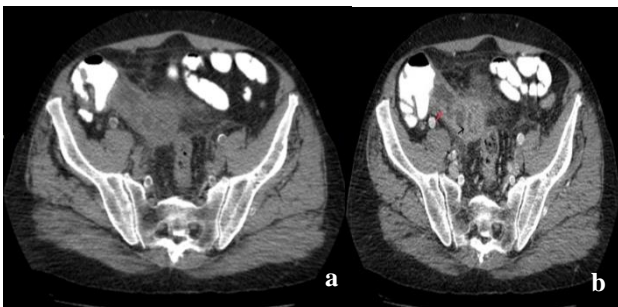
treated with laparoscopic cecectomy only. Post-operative histopathology showed negative margins in all cases.



**Figure 4: Case 4- axial unenhanced (a) CT scan shows dilated appendix (white arrow) with low attenuating intraluminal content. Axial contrast-enhanced CT image (b) shows thickened enhancing wall of appendix. Sagittal contrast-enhanced CT image (c) shows irregular thickening of base of appendix involving adjacent cecum (red arrow).**

**Case 5**

A 72 years old male patient presented with mild right lower quadrant pain and weight loss. Patient was advised CT abdomen pelvis. Plain and contrast enhanced CT images were obtained after giving the oral contrast. CT scan shows prominent appendix with heterogeneous attenuation. Periappendiceal fat stranding is seen. Contrast-enhanced CT scan shows enhancing mural nodularity with internal separation. Few sub centimeter sized non necrotic enhancing ileocolic lymphadenopathy were also noticed. No evidence of free mucin. Surgeons were informed for the need of the frozen section. Patient was treated with radical right hemicolectomy. Frozen section and post-operative histopathology confirmed diagnosis as mucinous cystadenocarcinoma.



**Figure 5: Case 5- axial unenhanced (a) CT scan shows prominent appendix with heterogeneous attenuation. Periappendiceal fat stranding is seen (white arrow). Axial contrast-enhanced (b) CT scan shows enhancing mural nodularity (red arrow) with internal separation (black arrow).**

**Case 6**

A 66 years old male patient presented with lower abdominal pain and weight loss. Post contrast CT scan shows enhancing peritoneal deposits with presence free mucin in the pelvis likely due to ruptured mucocele. Surgeons were informed for the need of frozen section. It showed changes of mucinous cystadenocarcinoma. Patient was treated with radical right hemicolectomy with cytoreduction and HIPEC. Post-surgery histopathology confirmed the diagnosis as mucinous cystadenocarcinoma.



**Figure 6: Case 6- axial contrast-enhanced CT scan image shows enhancing peritoneal deposits (red arrow) with presence free mucin in the pelvis (white arrow).**

**DISCUSSION**

Appendiceal mucocele was first described as pathological entity by Rokitansky in 1842. It is a rare occurrence, only reported in less than 1% specimen of the appendectomy.<sup>1</sup> However, incidental finding of appendiceal neoplasm in imaging is increasing over the years. Appendiceal mucocele described as mucin filled enlargement of the appendix. It is a macroscopic descriptive term. Aetiology include neoplastic and non-neoplastic causes. Non-neoplastic causes such as luminal obstruction, resulting in simple retention cyst e.g by appendicolith and mucosal hyperplasia, only account for 20% of mucocele.<sup>2</sup>

According to recent consensus issued by peritoneal surface oncology group international (PSOGI) mucinous neoplasm of appendix is classified in four groups- (a) LAMN (low-grade mucinous neoplasm); (b) HAMN (high-grade mucinous neoplasm); (c) mucinous adenocarcinoma; and (d) poorly differentiated, mucinous adenocarcinoma.<sup>3</sup>

Clinical diagnosis of the mucocele is difficult as its presentation is quite non-specific. It may present with right lower quadrant pain like acute appendicitis, abdominal distension, weight loss, per-rectal bleeding, unexplained anaemia, altered bowel habits, nausea and vomiting. It may complicate into intussusception or bowel obstruction. However 25% of cases are asymptomatic and incidentally detected during surgery or imaging for other purpose.<sup>4</sup> Imaging appearance of the appendix is well described in the literatures. In CT imaging, it usually appears as a well-

defined cystic, spherical or tubular mass of appendix with hypoattenuating intra luminal content (Figure 1).<sup>4,5</sup> Curvilinear wall or punctate calcification (Figure 2) is described as a characteristic feature of appendiceal mucocele but only present in less than 50% of cases.<sup>4,6</sup>

### **Critical imaging features**

Once the identification appendiceal mucocele is confirmed or suspected on imaging you should always look for the presence of extra appendiceal mucin. If the features suggestive of ruptured mucocele like wall discontinuity and/or the presence of free mucin in peritoneal cavity are observed, it will prompt the surgeon to prepare for frozen section and be ready for HIPEC and/or more extensive surgical approach. As it will negate the possibility of second and/or more extensive surgical approach, which is required if the pathology of resected specimen is suggestive of neoplasm.

In our study, we found one case with imaging features of invasive appendiceal mucocele with PMP (Pseudomyxoma peritonei) (Figure 6). Patient was treated with right hemicolectomy with lymphadenectomy with cytoreduction surgery followed by HIPEC. Findings were confirmed on frozen section and histopathology.

Some studies reported that non-specific findings like appendiceal wall thickening and mural nodularity (Figure 5) can be indicative of malignancy, secondary to inflammatory or both. While Wang et al study suggest that presence of wall irregularity and mural nodularity is strongly associated with appendiceal mucinous cyst adenocarcinoma.<sup>6</sup> They suggested that presence of these two imaging findings, right hemicolectomy should be primarily advocated. However this study has many limitation including small sample size. In our case series, we have found one patient with such findings in addition to ileocolic lymphadenopathy. Patient was treated with right hemicolectomy with ileocolic lymphadenopathy. However by revising literatures we recommend right hemicolectomy when lymphadenopathy is present along with mural nodularity and wall irregularity on imaging.<sup>6,7</sup> As LAMN usually do not cause lymph node metastasis as compared to higher risk of lymph node invasion and lower prevalence in appendiceal mucinous cyst adenocarcinoma.<sup>7,8</sup> Availability of frozen section provides surety. Imaging findings such as a irregular wall thickening involving the caecum and/or soft tissue thickening near or involving the base of appendix (Figure 4) prompt surgeons to look for the same during laparoscopy and if it is involved, cecectomy is advised in order to obtain negative margin in frozen section and histopathology. If margins are negative it's considered as definite treatment. Surgeons can perform preoperative colonoscopy for direct visualisation of cecal involvement based on imaging findings. This may clarify the need of cecectomy. In our case series, three patients were operated as cecectomy and all of them were diagnosed as LAMN on histopathology. In absence of all above features (Figure 1-

3), appendectomy is considered as completion of surgery, if histopathology of resected specimen shows negative margins.<sup>7,8</sup> Especially in cases of non-neoplastic etiology. In our case series, we have four such patients and managed by appendectomy alone. Two of them diagnosed as LAMN and rest two are of non-neoplastic etiology.

It is important to identify mucocele in background of appendicitis because presence of mucocele may modify the surgical approach or extra cautions should be taken to avoid rupture of mucocele. Study by Bennett et al suggest that cystic dilatation of appendix, diameter >13 mm, presence of calcification are suggestive of mucocele with co-existing appendicitis.<sup>9</sup> In our case study, we have found one case of mucocele in background of appendicitis and treated by lap appendectomy. In previous literature, open surgery was advocated when mucocele of appendix was diagnosed. Now laparoscopic approach with options of cecal cuff resection and right hemicolectomy are available based on the radiological and histopathological findings.<sup>10</sup>

### ***Pseudomyxoma peritonei***

Although comprehensive and uniform definition PMP is not available yet, it can be defined as heterogeneous disease which can be distinguish from the usual peritoneal metastasis by presence of the excessive amount of mucin in the peritoneal cavity. It is difficult to distinguish it from ascites in CT. However PMP may have internal septation, loculation and heterogeneous appearance, peritoneal deposits, serosal deposits causing scalloping effect on solid organs and/or curvilinear or amorphous calcification.

PSOGI expert panel has categorised and proposed grading system for PMP based on histology of peritoneal disease. Acellular mucin - characterised by absence of neoplastic epithelial cell within the peritoneal mucin, low grade mucinous carcinoma peritonei- G1, high grade mucinous carcinoma peritonei- G2, high grade mucinous carcinoma peritonei with signet ring cell- G3.<sup>11</sup>

Prognosis is heavily dependent on the histological grading of PMP.<sup>12-14</sup> Pre-operative evaluation and appropriate treatment of PMP is recommended because untreated disease has a 5 year survival rate of 55%-65%. Despite the increased in knowledge and insight of disease no strict guidelines is available for management of PMP. At present it is said that all kind of PMP should be considered for CRS+HIPEC.<sup>12-14</sup>

In literatures, various quantitative prognostic systems have been described which can be assessed by preoperative imaging such as peritoneal cancer index (PCI), peritoneal surface disease severity score (PSDSS), simplified pre-operative assessment for appendix tumour (SPAAT) score. These scoring systems are being used for selection of optimal candidates for CRS/HIPEC.<sup>15,16</sup> Aim of these scoring system is to predict likely hood of complete cytoreduction and good prognosis of this subset of

patients.<sup>16</sup> It is observed that radiological PCI underestimate the disease as compared to the surgical PCI. However accuracy can be improved by using positive oral contrast and intravenous contrast which increase sensitivity of bowel serosal disease, adjacent peritoneal and mesenteric disease.<sup>17</sup>

CT has its own limitations for PMP imaging to detect nodules smaller than 5 mm with reporting sensitivity of 11%-28%. This can cause trouble because miliary deposits is contraindication for HIPEC.<sup>17</sup> Sometimes it is difficult to interpret PCI in presence of tumour plaques, surgical scars and omental caking due to obscuration of boundaries and overlapping of pathology. Mucin can obscure and/or mimic as mets or can be mistaken for simple ascites.<sup>17</sup> Delayed contrast enhanced MRI with diffusion weighted imaging is shown to be superior with higher specificity to detect smaller implants and help to distinguish between acellular mucin and more cellular mucin.

Recent studies suggested that contrast enhanced CT is superior to PET alone to predict extent of peritoneal carcinomatosis. However combination of both provide best result and can be useful tool for selecting patient for HIPEC and cytoreduction.<sup>18</sup> Some authors suggest diagnostic laparoscopy because of given limitation of the CT imaging for selection of patients amenable to complete cytoreduction. However certain areas are not visualised well including intrahepatic and pancreatic parenchymal metastasis and intra luminal gastrointestinal implants. CT imaging will be helpful here to draw attention of surgeons to the presence of such disease.<sup>19</sup>

A standardised reporting format should be made by institute to mitigate CT limitations and in interest to reach uniform and consistent reporting. At our institute GI onco surgeons and radiologists collaborated to make a reporting format to maximise the reach to the disease present at possible locations. Recently a publication introduced the use of acronym 'pause' in order to standardise reporting of peritoneal disease. Which include- primary tumour and PCI, ascites and abdominal wall involvement, unfavourable sites of involvement, small bowel and mesenteric disease, extra peritoneal metastasis.<sup>20</sup>

Our reporting format is inclusive of all these criteria.

## CONCLUSION

Surgical approach for appendiceal mucocele has evolved from aggressive open surgery to laparoscopic surgery with cecal cuff resection and hemicolectomy. As appendiceal mucocele presents with non-specific symptoms, imaging plays important role in diagnosis. Surgical approach should be different on the basis of possible histopathology and extent of disease in interest to achieve best prognosis for each patients. Specific CT findings like mural nodularity, wall irregularity, lymphadenopathy, cecal involvement, presence of free mucin and features suggestive of PMP can predict appropriate surgical

approach and prompt surgeon to prepare for frozen section. Contrast enhanced CT imaging with use of positive oral contrast increase the sensitivity to localise peritoneal deposits and improve accuracy in predicting the tumour burden, which may help surgeons to select patients feasible to complete cytoreduction surgery. Imaging finding can predict possible etiology and extent of disease. As prognosis depends upon the histopathological subtype of mucocele and PMP and surgical approach, a Multispeciality cooperative teamwork is required for each patients to think up best suitable, customised treatment plan in interest of achieving and predicting best prognosis. Preoperative evaluation of PMP using standardised reporting formate may direct the surgeons during diagnostic and/or therapeutic laparoscopic surgery.

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