International Surgery Journal
Alexander S et al. Int Surg J. 2025 Apr;12(4):611-613
http://www.ijsurgery.com
pISSN 2349-3305 | eISSN 2349-2902

DOI: https://dx.doi.org/10.18203/2349-2902.isj20250744

Case Report

Incidental low-grade appendiceal mucinous neoplasm discovered during emergency caesarean section: a case report

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Received: 01 March 2025
Revised: 13 March 2025
Accepted: 15 March 2025

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ABSTRACT

Low-grade appendiceal mucinous neoplasm (LAMN) is a rare epithelial tumour, often discovered incidentally. The condition is of particular concern due to its potential to cause pseudomyxoma peritonei (PMP). LAMN in pregnancy is exceedingly rare, with only a limited number of cases reported. Its diagnosis is often challenging due to the physiological changes of pregnancy, which can obscure symptoms and delay detection. We present a case of a 22-year-old female who underwent an emergency lower-segment caesarean section (LSCS) at 39 weeks gestation due to failure to progress and foetal distress. Intraoperatively, the peritoneal cavity contained copious mucinous material, and an enlarged, erythematous appendix was noted. The general surgical team were consulted and performed an appendicectomy, and histopathological examination confirmed LAMN with acellular mucin deposition on the serosal surface. Cytological analysis of the peritoneal fluid was negative for epithelial cells. Postoperative surveillance included tumour marker assessment, colonoscopy, and staging imaging, all of which showed no evidence of peritoneal dissemination. A follow-up diagnostic laparoscopy at six months confirmed the absence of disease progression. This case highlights the challenges of diagnosing LAMN in pregnancy and underscores the importance of intraoperative vigilance during caesarean sections. Given the potential for peritoneal spread, structured long-term surveillance is critical. Early recognition and appropriate management can help prevent complications such as PMP and improve patient outcomes.

Keywords: Low-grade appendiceal mucinous neoplasm, Pregnancy, Caesarean section, Pseudomyxoma peritonei, Appendectomy

INTRODUCTION

Low-grade appendiceal mucinous neoplasm (LAMN) is a rare and often incidental finding, representing 0.2-0.7% of all appendix specimens.[1] It is characterized by mucinous epithelial proliferation with varying degrees of mucosal atrophy, fibrosis, and acellular mucin deposition.[2] Although LAMN is considered a neoplasm with low malignant potential, its primary clinical concern lies in the potential for peritoneal dissemination and the development of pseudomyxoma peritonei (PMP), a condition associated with significant morbidity.[3] LAMN often remains asymptomatic until it presents with complications, such as acute appendicitis, rupture, or an incidental discovery during unrelated abdominal surgeries.[4]

LAMN in pregnancy is particularly rare, with only a handful of cases reported in the literature.[5] Pregnancy itself can obscure symptoms due to the physiological changes in the abdomen, leading to delayed diagnosis and challenges in surgical decision-making.[6] Most reported cases in these circumstances were identified incidentally during caesarean section or surgical interventions performed for presumed appendicitis.[7] The presence of mucinous material in the peritoneal cavity during

caesarean poses unique challenges and concerns, highlighting the importance of a multidisciplinary approach to management.[8]

Here, we present a case of an incidental LAMN discovered during an emergency caesarean section. The case highlights the diagnostic and management challenges associated with LAMN in pregnancy and emphasizes the importance of appropriate postoperative surveillance to mitigate the risk of disease progression.[1]

CASE REPORT

A 22-year-old female, gravida 1 para 0, was admitted at 39 weeks gestation in spontaneous labour. Despite spontaneous onset, labour failed to progress beyond 9 cm dilation, necessitating an emergency LSCS (EmLSCS) due to foetal distress. No imaging had been performed prior to the caesarean section. A healthy neonate was delivered. Upon entry into the peritoneal cavity, copious yellowish mucinous fluid was encountered. Further exploration revealed an enlarged and erythematous appendix without perforation or abscess formation (Figure 1). The on-call general surgical team was consulted intraoperatively, and following discussion with the consultant, an appendicectomy was performed. The appendix was ligated using an endoloop and divided at the base, and the peritoneal cavity was irrigated with two litres of normal saline. It was noted that the appendiceal base did not appear dilated and looked healthy in comparison to the rest of the appendix.

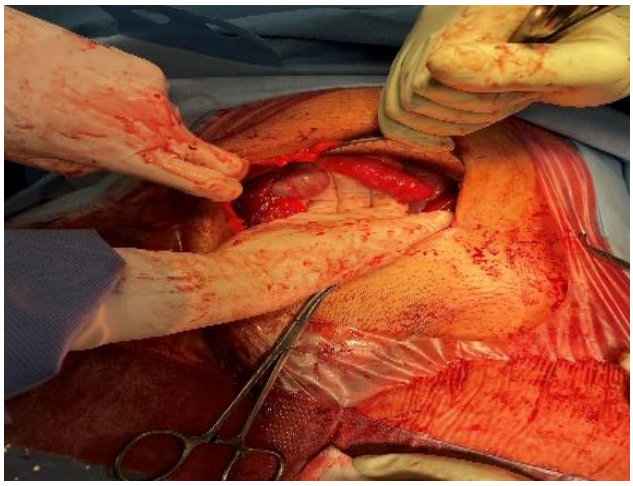

Figure 1: Intraoperative findings during the emergency caesarean section.

An enlarged, erythematous appendix with adjacent mucinous material is demonstrated within the peritoneal cavity, consistent with a LAMN. The appendix was subsequently resected, and peritoneal lavage was performed.

The patient's preoperative blood investigations revealed a leukocytosis of 22.4×10^9/l with neutrophilia at 18.65×10^9/l. Hemoglobin was mildly elevated at 143 g/l. Platelet count was 313×10^9/l. Biochemical markers, including renal and liver function tests, were within normal limits. Her past medical history included a multinodular goitre.

The patient's postoperative recovery was uneventful. Histopathological examination of the appendix confirmed a LAMN with acellular mucin on the serosal surface. The neoplasm did not demonstrate any infiltrative features, and the proximal appendiceal margin was clear. Cytological analysis of the peritoneal fluid confirmed the presence of acellular mucin without epithelial cells, indicating localized disease without evidence of dissemination.

Following multidisciplinary discussion, the patient was referred to a colorectal surgical team for ongoing surveillance. Baseline tumour markers, including CEA, CA-125, and CA 19.9, were measured, showing values of 0.5 µg/l, 32 kU/l, and 10 kU/l, respectively. A colonoscopy was performed, revealing no abnormalities or evidence of residual disease. A staging CT of the chest, abdomen, and pelvis showed no signs of metastasis. At six months postoperatively, a diagnostic laparoscopy was performed, which confirmed the absence of peritoneal disease with a peritoneal cancer index (PCI) score of 0. Biopsies taken during the laparoscopy showed only vascular congestion, with no mucin or malignancy present.

At one year postoperatively, the patient remains disease-free with no clinical or radiological evidence of peritoneal dissemination. She continues to undergo regular follow-up with oncology and colorectal specialists to monitor for any recurrence or progression of disease.

DISCUSSION

LAMN is a distinct pathological entity within the spectrum of appendiceal neoplasms, primarily distinguished by its low-grade cytological features and potential for peritoneal spread despite lacking invasive characteristics.[2] The risk of peritoneal dissemination is primarily dictated by tumour rupture, with up to 23% of LAMN patients developing peritoneal metastases despite negative initial imaging.[3] Consequently, long-term surveillance strategies, including imaging and tumour marker assessments, are critical to detecting occult disease progression.[3]

In pregnancy, LAMN presents unique diagnostic and management dilemmas. Physiological changes, including abdominal distension and altered pain perception, can obscure symptoms, leading to missed or delayed diagnoses.[6] In many cases, imaging fails to detect LAMN preoperatively, as seen in this case where no imaging was performed prior to caesarean section.[5] Incidental findings during caesarean section, such as the presence of mucinous material, should prompt intraoperative assessment by a surgical team, as a delay in intervention may increase the risk of future presentation with PMP.[8]

The management of LAMN identified intraoperatively is debated, but consensus suggests appendicectomy with peritoneal lavage is appropriate for localized disease with no evidence of extra-appendiceal spread.[4] Histopathological confirmation and cytology are essential in guiding postoperative surveillance, as even acellular mucin in the peritoneal cavity warrants close follow-up due to the risk of delayed disease manifestation.[2] Current literature supports the use of colonoscopy, cross-sectional imaging, and, in select cases, diagnostic laparoscopy for disease staging and surveillance.[3]

Despite the indolent nature of LAMN, progression to PMP remains a serious concern. Current recommendations suggest surveillance for at least five years, with periodic imaging and tumour marker assessments, to monitor for delayed peritoneal dissemination.[3] The role of cytoreductive surgery with hyperthermic intraperitoneal chemotherapy (CRS/HIPEC) is reserved for cases with confirmed peritoneal dissemination.[9]

CONCLUSION

In conclusion, LAMN is rare, and its incidental discovery in pregnancy presents unique clinical challenges. Given the potential for delayed complications, a structured follow-up plan is crucial to ensuring optimal patient outcomes.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Alexander S, Cross T. Incidental low-grade appendiceal mucinous neoplasm discovered during emergency caesarean section: a case report. Int Surg J 2025;12:611-3.