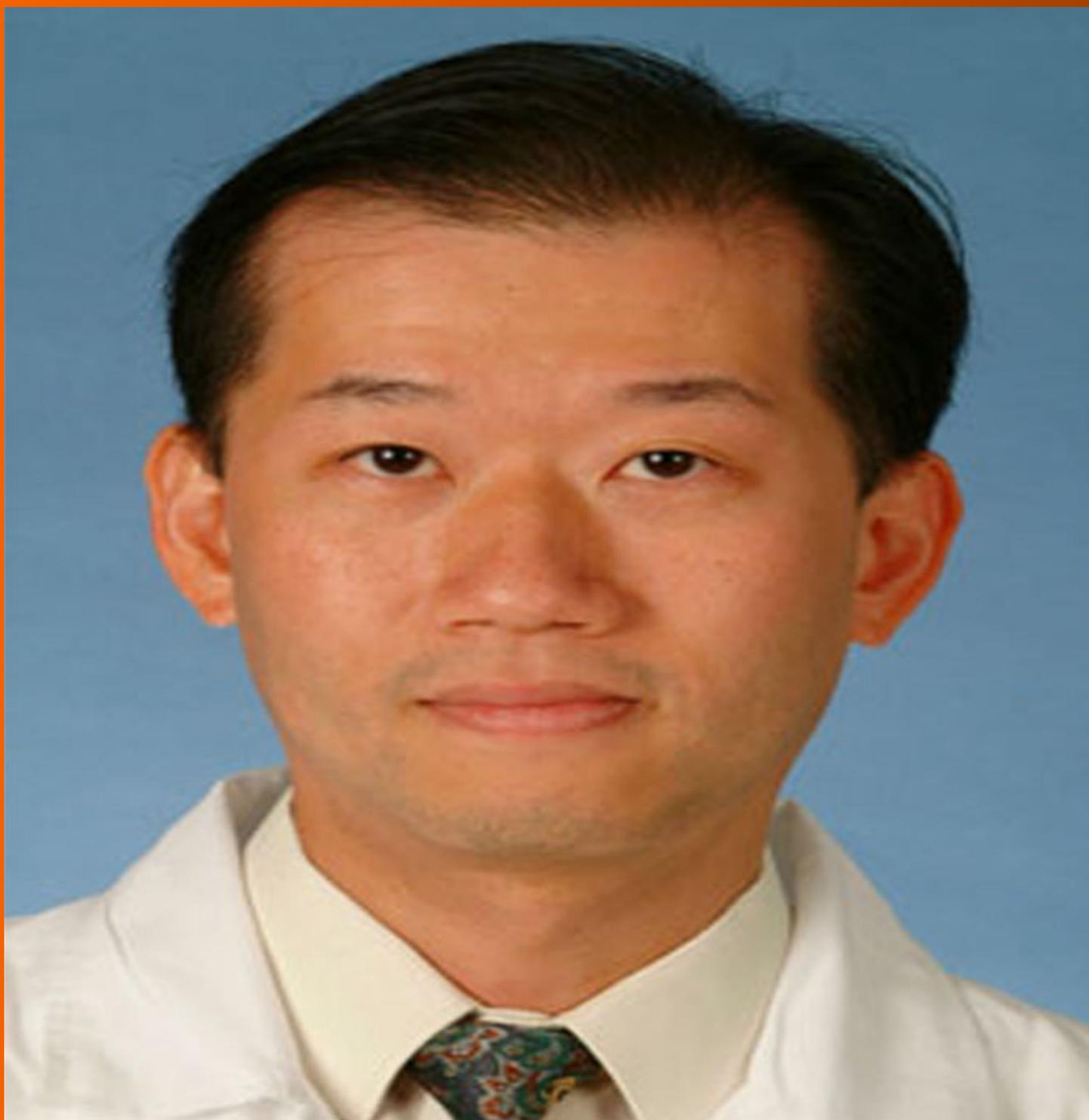


World Journal of *Clinical Cases*

World J Clin Cases 2019 November 6; 7(21): 3384-3682



EDITORIAL

- 3384 Current controversies in treating remnant gastric cancer: Are minimally invasive approaches feasible?
Ma FH, Liu H, Ma S, Li Y, Tian YT

ORIGINAL ARTICLE**Retrospective Study**

- 3394 Efficient management of secondary haemophagocytic lymphohistiocytosis with intravenous steroids and γ -immunoglobulin infusions
Georgiadou S, Gatselis NK, Stefos A, Zachou K, Makaritsis K, Rigopoulou EI, Dalekos GN
- 3407 Impress of intergenerational emotional support on the depression in non-cohabiting parents
Jia YH, Ye ZH
- 3419 Nomograms for pre- and postoperative prediction of long-term survival among proximal gastric cancer patients: A large-scale, single-center retrospective study
Chen QY, Hong ZL, Zhong Q, Liu ZY, Huang XB, Que SJ, Li P, Xie JW, Wang JB, Lin JX, Lu J, Cao LL, Lin M, Tu RH, Zheng CH, Huang CM

Observational Study

- 3436 Modified Cortex Mori Capsules improving the successful rate of functional filtering blebs after reclinical glaucoma filtering surgery
Yu J, Qiu LX, Qing GP, Zhao BW, Wang H
- 3446 Effect of cognitive behavior therapy combined with exercise intervention on the cognitive bias and coping styles of diarrhea-predominant irritable bowel syndrome patients
Zhao SR, Ni XM, Zhang XA, Tian H

Prospective Study

- 3463 Normal values of shear wave velocity in liver tissue of healthy children measured using the latest acoustic radiation force impulse technology
Sun PX, Tong YY, Shi J, Zhang H, Liu SJ, Du J

SYSTEMATIC REVIEW

- 3474 Characteristics of clear cell renal cell carcinoma metastases to the thyroid gland: A systematic review
Khaddour K, Marernych N, Ward WL, Liu J, Pappa T
- 3486 Irritable bowel syndrome and functional constipation management with integrative medicine: A systematic review
Dai L, Zhong LL, Ji G

- 3505 How about the reporting quality of case reports in nursing field?

Yang KL, Lu CC, Sun Y, Cai YT, Wang B, Shang Y, Tian JH

CASE REPORT

- 3517 Gastro-gastric intussusception in the setting of a neuroendocrine tumor: A case report

Zhornitskiy A, Le L, Tareen S, Abdullahi G, Karunasiri D, Tabibian JH

- 3524 Retroperitoneal perivascular epithelioid cell tumours: A case report and review of literature

Touloumis Z, Giannakou N, Sioros C, Trigka A, Cheilakea M, Dimitriou N, Griniatsos J

- 3535 First Italian outbreak of VIM-producing *Serratia marcescens* in an adult polyvalent intensive care unit, August-October 2018: A case report and literature review

Iovene MR, Pota V, Galdiero M, Corvino G, Di Lella FM, Stelitano D, Passavanti MB, Pace MC, Alfieri A, Di Franco S, Aurilio C, Sansone P, Niyas VKM, Fiore M

- 3549 Transfemoral aortic valve implantation in the case of pre-existing mitral prosthesis and pure aortic regurgitation: A case report

Erdem A, Esen Zencirci A, Ozden K, Terzi S

- 3553 Methotrexate-related lymphoproliferative disorders in the liver: Case presentation and mini-review

Mizusawa T, Kamimura K, Sato H, Suda T, Fukunari H, Hasegawa G, Shibata O, Morita S, Sakamaki A, Yokoyama J, Saito Y, Hori Y, Maruyama Y, Yoshimine F, Hoshi T, Morita S, Kanefuji T, Kobayashi M, Terai S

- 3562 Re-revision surgery for re-recurrent valgus deformity after revision total knee arthroplasty in a patient with a severe valgus deformity: A case report

Du YQ, Sun JY, Ni M, Zhou YG

- 3569 Liver transplantation for severe portopulmonary hypertension: A case report and literature review

Chen XJ, Zhu ZJ, Sun LY, Wei L, Zeng ZG, Liu Y, Qu W, Zhang L

- 3575 Leiomyosarcoma of the stomach: A case report

Kang WZ, Xue LY, Tian YT

- 3583 Out-of-hospital cardiac arrest in a young adult survivor with sequelae of childhood Kawasaki disease: A case report

Zhu KF, Tang LJ, Wu SZ, Tang YM

- 3590 Squamous cell carcinoma of the nail bed: A case report

Li PF, Zhu N, Lu H

- 3595 Multidisciplinary treatment of a patient with necrotizing fasciitis caused by *Staphylococcus aureus*: A case report

Xu LQ, Zhao XX, Wang PX, Yang J, Yang YM

- 3603** Myocardial ischemic changes of electrocardiogram in intracerebral hemorrhage: A case report and review of literature
Lin XQ, Zheng LR
- 3615** Adenomyoma of the distal common bile duct demonstrated by endoscopic ultrasound: A case report and review of the literature
Xu LM, Hu DM, Tang W, Wei SH, Chen W, Chen GQ
- 3622** Child with Wiskott–Aldrich syndrome underwent atypical immune reconstruction after umbilical cord blood transplantation: A case report
Li BH, Hu SY
- 3632** Epiphyseal distraction and hybrid reconstruction using polymethyl methacrylate construct combined with free non-vascularized fibular graft in pediatric patients with osteosarcoma around knee: A case report
Liang YH, He HB, Zhang C, Liu YP, Wan J
- 3639** Bilateral common carotid artery common trunk with aberrant right subclavian artery combined with right subclavian steal syndrome: A case report
Sun YY, Zhang GM, Zhang YB, Du X, Su ML
- 3649** Giant gastroduodenal trichobezoar: A case report
Dong ZH, Yin F, Du SL, Mo ZH
- 3655** Compound heterozygous mutation of *MUSK* causing fetal akinesia deformation sequence syndrome: A case report
Li N, Qiao C, Lv Y, Yang T, Liu H, Yu WQ, Liu CX
- 3662** Hypoparathyroidism with Fahr’s syndrome: A case report and review of the literature
Zhou YY, Yang Y, Qiu HM
- 3671** Primitive neuroectodermal tumors of the abdominal wall and vulva in children: Report of two cases and review of the literature
Xu QQ, Xing WW, Chen G, Dang YW, Luo YG, Chen P, Liang SW, Chen JB

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Gastro-gastric intussusception in the setting of a neuroendocrine tumor: A case report

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Abstract

BACKGROUND

While in children intussusception is often idiopathic, in adults it is commonly caused by a pathologic condition functioning as a lead point. It is important to note that a variety of pathologic conditions may trigger intussusception, with malignancy being a relatively frequent culprit in adults; this should be considered high on the differential diagnosis during evaluation.

CASE SUMMARY

This is a case of a 40-year-old female presenting to the emergency department (ED) with three days of acute on chronic, peri-umbilical abdominal pain described as waxing and waning, and pressure-like in nature. Initial computed tomography (CT) of the abdomen and pelvis with contrast in the ED (after her pain had resolved) re-demonstrated a previously noted 13 mm lesion in the gastric antrum but no clear cause of the pain. Endoscopic ultrasound was pursued, and the mass lesion was sampled *via* fine needle biopsy. Post-procedure the patient experienced another episode of severe pain which prompted a repeat stat CT abdomen and pelvis with contrast; this re-demonstrated the 13 mm antral lesion and in addition was remarkable for a gastro-gastric intussusception. An upper gastrointestinal gastrograffin series was ordered (completed only after the pain had subsided) and showed resolution of the intussusception. Histopathology was consistent with a diagnosis of low-grade neuroendocrine tumor (NET). Surgery was initially deferred during the hospitalization given the low grade pathology of the lesion; however further multidisciplinary discussion between Surgery, Oncology, and Gastroenterology recommended resection given

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the patient's recurrent abdominal pain with the NET functioning as a lead point for further intussusception, and the patient thus underwent robotically-assisted wedge resection.

CONCLUSION

We present a unique case of severe, intermittent, peri-umbilical pain related to gastro-gastric intussusception caused by an antral NET lead point. The case highlights the importance of considering neoplasms as the cause of intussusception in adults and the greater diagnostic yield when imaging is obtained while symptoms (in this case severe, episodic abdominal pain) are most apparent.

Key words: Abdominal pain; Gastro-gastric intussusception; Neuroendocrine tumor; Case report

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Core tip: Intussusception in adults is an important etiology to consider in patients with acute abdominal pain; the cause may be a neoplasm, which can function as a lead point, and timely imaging is a vital part of the diagnostic evaluation. This case report describes a 40 years-old female with multiple emergency department visits for acute abdominal pain with and what was initially thought to be an incidental 13 mm gastric antral mass. The patient was ultimately diagnosed with gastric-gastric intussusception and a gastric neuroendocrine tumor as a lead point.

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INTRODUCTION

The clinical presentation of intussusception includes symptoms of abdominal pain and/or bowel obstruction due to telescoping of a portion of a bowel segment into the lumen of another. It is most frequently seen in children, where it is commonly idiopathic; in adults, intussusception is predominantly secondary to a pathologic condition acting as a lead point (*e.g.*, a stricture due to inflammatory bowel disease, large polyp, or hamartoma), with more than half of cases being caused by an underlying malignancy.

CASE PRESENTATION

Chief complaints

A 40-year-old female presenting to the emergency department (ED) with three days of severe, pressure-like, peri-umbilical abdominal pain.

History of present illness

The patient's abdominal pain was waxing and waning in nature, exacerbated by food and associated with nausea and occasional diarrhea. The patient denied melena, hematochezia, change in stool caliber, weight loss, early satiety, flushing, fevers or chills. She had experienced multiple episodes of pain over the past six months resulting in two ED visits. On the most recent episode one month prior, she presented with similar abdominal pain, at which time computed tomography (CT) of the abdomen and pelvis with contrast showed a 13 mm mass in the gastric antrum. The antral lesion, however, appeared too small to be causing the patient's pain, and given spontaneous resolution of her pain, she was discharged.

Past medical and surgical history

Notable for well controlled rheumatoid arthritis and cholecystectomy five years ago.

History of family illness

Family history includes gastric cancer in her father that was diagnosed at 61 years old.

Physical examination

Vital signs on presentation were notable for tachycardia to 101 beats per minute and hypertension of 159/105 mm Hg. Physical examination revealed a non-distended, soft, abdomen with diffuse tenderness to minimal palpation, without rebound tenderness.

Laboratory examinations

Laboratory studies, including complete blood count, serum electrolytes, lactate, and gastrin levels, were within normal limits.

Imaging examinations

In the ED, initial CT abdomen and pelvis with contrast was ordered but not completed until after resolution of the patient's abdominal pain. It re-demonstrated a 13 mm mass in the gastric antrum seen on CT during her last presentation to the ED one month prior but was otherwise unremarkable. An endoscopic ultrasound was recommended to further investigate the antral lesion; a 13 mm subepithelial, hypoechoic lesion was seen in the antrum, and fine needle biopsy (FNB) was performed (Figure 1). Several hours after the procedure, the patient again had an episode of the same abdominal pain she had been experiencing; this prompted a stat CT of the abdomen with contrast that showed a gastro-gastric intussusception (Figures 2 and 3) without any evidence of bleeding or other FNB-related adverse events. The pain again subsided with supportive measures.

Pathologic evaluation

Histopathology was notable for a low mitotic rate, with Ki67 index < 1%, and staining positive for synaptophysin and chromogranin, suggestive of a grade 1 neuroendocrine tumor (NET) (Figures 4 and 5).

FINAL DIAGNOSIS

Gastro-gastric intussusception secondary to a gastric NET functioning as a lead point.

TREATMENT

General surgery was consulted for management of the intussusception and recommended an upper gastrointestinal gastrograffin series for further visualization. The study was completed after resolution of the patient's abdominal pain and demonstrated resolution of the intussusception. Given the location of the tumor, its low-grade nature based on Ki67 and mitotic count, as well as the resolution of symptoms, a multidisciplinary decision was made to defer any surgical interventions during the hospitalization and instead continue further workup as an outpatient with close Oncology, Gastroenterology, and Surgery follow-up.

OUTCOME AND FOLLOW-UP

A follow-up nuclear medicine octreotide whole body scan was performed showing an intense focus of uptake at the antrum corresponding to the known mass with no other areas of uptake that would be suggestive of signs of metastasis. The patient continued to note episodes of severe abdominal pain during outpatient clinic visits and repeat multidisciplinary discussion between Surgical Oncology and Gastroenterology resulted in the decision to proceed with surgical resection and she is now status post robot-assisted partial gastrectomy and doing clinically well. While it was acknowledged that the risk of metastasis is low given the histopathology, the tumor's continued presence appeared to be the underlying cause of her recurrent abdominal pain as it remained a lead point for repeat intussusception, hence warranting surgical resection.

DISCUSSION

This case highlights what is most frequently seen in adults as the cause of



Figure 1 Endoscopic ultrasound demonstrating a well-circumscribed, hypoechoic, subepithelial antral mass.

intussusception: a pathologic condition functioning as lead point. However, there are multiple interesting aspects of this case that have not previously been described in published literature, including the tumor location and underlying NET as a lead point.

The nomenclature for neuroendocrine masses is confusing with the acronyms neuroendocrine neoplasms (NEN), NET, and neuroendocrine carcinomas (NEC) often used interchangeably. According to the World Health Organization (WHO), the overarching term is NEN with NETs referring to masses that show more benign characteristics while NECs are more aggressive and have malignant features^[1]. Simply put, NENs are a heterogeneous group of tumors that can be found throughout the body and are characterized by their endocrine functions, which are monoamines synthesis and secretion. These neoplasms are quite rare and account for only 0.5% of all newly diagnosed malignancies. The most frequent primary sites are the gastrointestinal tract (62%) and the lung (22%)^[2]. Histologically, NETs are well-differentiated and identified by intense immuno-expression of neuroendocrine markers such as synaptophysin and chromogranin A, whereas similarly named NECs are poorly differentiated, have decreased expression of neuroendocrine markers, and higher mitotic rates. Per 2010 WHO guidelines, NENs found in the gastrointestinal tract are classified based on their mitotic count and Ki67 proliferation^[1].

The most common locations for intussusception are small bowel only (entero-enteric), large bowel only (colo-colonic), or small bowel into large bowel (termed ileocolic or ileo-cecal if the ileocecal valve is determined to be the lead point)^[2]. The frequency of the intussusception at certain sites was evaluated in a study of 745 adults and showed that 52% were found in the small intestine and 38% in the large intestine (17% ileocecal, 17% colocolic, 4% appendiceal)^[3]. However, intussusception involving the proximal gastrointestinal tract such as the stomach is exceedingly rare. A recent literature review identified only 10 instances of gastro-gastric intussusception since 1950, all of which were associated with a mass, either benign or malignant, functioning as a lead point^[4-13] (Table 1).

The underlying etiology of intussusception is essential to consider, as it determines management. As alluded to before, in children the most common etiology of intussusception is overwhelmingly idiopathic, whereas in adults, more than half of the cases are due to an underlying malignancy, with adenocarcinoma being the most likely culprit^[14]. Therefore, if initial biopsies are negative for malignancy, clinicians should maintain a high index of suspicion and have a low threshold for repeat biopsy such as in this case. A NET leading to intussusception is exceedingly rare, with only a few case reports described in the available literature^[15]. Furthermore, none of these aforementioned cases involved the stomach or resulted in gastro-gastric intussusception as observed in our case.

In the absence of an underlying systemic disorder (*e.g.*, Celiac disease, lymphoma), the mainstay of management of intussusception is typically definitive treatment with surgical resection^[16,17]. This case would similarly require surgical intervention for definitive treatment since the NET continues to put the patient at risk for recurrent episodes of intussusception.

CONCLUSION

This case describes a previously unreported presentation of severe, episodic, peri-umbilical pain due to gastro-gastric intussusception caused by a gastric NET.

Table 1 Summary of 10 known cases of gastro-gastric intussusception

Ref.	Age/sex	Presentation	Histological findings	Intussusception diagnostic modality
Grundy <i>et al</i> ^[4]	78 F and 76 F	Dysphagia, weight loss, epigastric pain, vomiting	Leiomyoma	Fluoroscopy
Thomson <i>et al</i> ^[5]	72 M	Epigastric pain, nausea, vomiting	Pedunculated intragastric tumour	Laparotomy
Javors <i>et al</i> ^[6]	81 F	Anemia	Leiomyoma with leiomyoblastomatous elements	Single contrast UGI series
Raw ^[7]	66 F	Epigastric discomfort, vomiting	Malignant gastric papilloma	Laparotomy
Shanbhogue <i>et al</i> ^[8]	83 F	Melena, anemia, weight loss	Gastrointestinal stromal tumour	CT abdomen
Vikram <i>et al</i> ^[9]	65 F	Epigastric pain, nausea, vomiting, epigastric mass	Hypertrophic gastric polyps	Double contrast barium meal, CT abdomen
Davila <i>et al</i> ^[10]	77 F	Fever, abdominal discomfort, left lateral abdominal mass	Tubulo-villous adenoma	MR abdomen
Eom <i>et al</i> ^[11]	73 F	Vomiting, general weakness	Gastric adenocarcinoma	CT abdomen
Jo <i>et al</i> ^[12]	82 F	Chest pain and vomiting	Primary gastric lymphoma	CT abdomen

CT: computed tomography; UGI: upper gastrointestinal; MR: magnetic resonance.



Figure 2 CT of the abdomen and pelvis, sagittal view, revealing gastro-gastric intussusception.



Figure 3 CT of the abdomen and pelvis, axial view, demonstrating gastro-gastric intussusception.

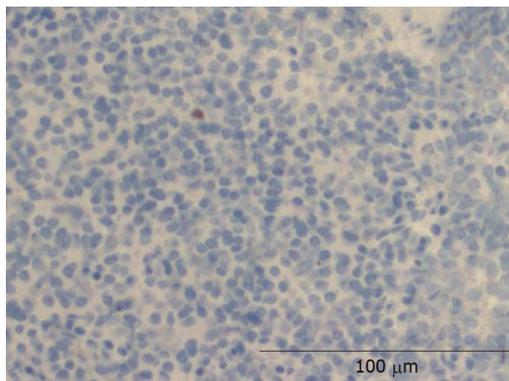


Figure 4 Histopathology showing low Ki67 staining (100x magnification).

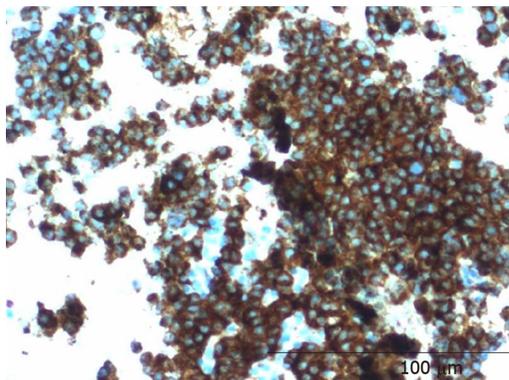


Figure 5 Histopathology showing positive synaptophysin staining (100x magnification).

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