Da Vinci robot-assisted resection to treat abdominal ectopic thyroid: A case report and literature review

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Case report

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Abstract

Background : Although ectopic thyroid can be found in thoracic locations, the discovery of abdominal ectopic thyroid is often an accidental event. Moreover, abdominal ectopic thyroid is easily misdiagnosed due to the rarity of these cases and the difficulties in the preoperative diagnosis process. Thus, we aimed to assess the prevalence and features of abdominal ectopic thyroid and to highlight the current knowledge about the clinical characteristics and management of this condition by analyzing a case report of abdominal ectopic thyroid and reviewing the literature.

Case presentation : A 70-year-old woman with a 3-year history of gradually increasing abdominal distension in the right lower quadrant of the abdomen was admitted to the hospital. Contrast-enhanced computed tomography (CT) of the abdomen revealed a retroperitoneal mass. The patient underwent Da Vinci robotic surgery, and the retroperitoneal mass was completely resected. Subsequently, the pathologic diagnosis of the mass was ET. The patient had no discomfort or symptoms when she was discharged from the hospital and at the postoperative 1, 3, 6, 9 and 12 month follow-up. The summary of literature review suggested that abdominal ectopic thyroid is still rare although there are some reports. Nonetheless, the cause of abdominal ectopic thyroid is unclear, and abdominal ectopic thyroid has the following characteristics: more common in women than in men, asymptomatic in the majority of the cases, difficult to diagnose, and found by excluding metastasis. Most ectopic thyroid is treated with surgery, and minimally invasive techniques have been increasingly performed.

Conclusion : This is the first report on Da Vinci robotic resection for large rare retroperitoneal ET, and this case highlights that ET should be considered when patients present with similar imaging findings in the abdomen.

Background

In 1953, Horst et al reported the first case of hypothyroidism in which an ectopic thyroid (ET) was diagnosed by the absence of symptoms based on an ^{131}I scan[1]. ET is an infrequently observed condition and is more frequent in females than in males. During the fourth embryonic week, the thyroid tissue starts developing. The thyroid tissue is normally located anterior to the trachea by the seventh embryonic week. Although the cause of ET is still not fully known, the disrupted thyroid migration during the embryonic period may explain this disease[2]. Clinically, an occasional mass or unexplained obstruction related to ET location and size could be observed. Abdominal tumors arise from various reasons, and the diagnosis of abdominal tumors is very difficult. Most patients with abdominal ET are asymptomatic, and ruling out metastases from thyroid cancer is important. Here, we provided a rare case report, which may be helpful for the diagnosis and treatment of abdominal tumors. Furthermore, we also summarized the prevalence, features, diagnosis and surgical management of abdominal ET.

Case Presentation

A 70-year-old woman with a 3-year history of gradually increasing abdominal distension in the right lower quadrant of the abdomen was admitted to the hospital. Her family history was negative for thyroid diseases, and the patient had no unusual discomfort. The laboratory results were unremarkable, and no results were positive for cancer markers. Contrast-enhanced computed tomography (CT) of the abdomen revealed a retroperitoneal mass with a size of 100.9 mm × 74.2 mm × 75.9 mm (Fig. 1A). After completing the preoperative examinations, the patient underwent Da Vinci robotic surgery, and the retroperitoneal mass was completely resected (Fig. 1B). Subsequently, the pathologic diagnosis of the mass was ET, but the struma ovarii needed to be excluded (Fig. 1C). Thus, B-mode ultrasonography and contrast-enhanced CT of the patient's thyroid and bilateral ovaries were performed. Ultimately, the diagnosis of struma ovarii was excluded based on the lack of abnormal examination results. The follow-up approach was specifically designed for this patient. The patient had no discomfort or symptoms when she was discharged from the hospital and at the postoperative 1, 3, 6, 9 and 12 month follow-up, which were conducted by telephone and at outpatient clinics.

Discussion

Prevalence and features of abdominal ET. The specific prevalence of ET is approximately 1 per 100,000-300,000 persons[3]. Abdominal tumors arise because of various reasons, and the diagnosis of abdominal tumors is very difficult. ET is most commonly found in the neck, and abdominal ET is relatively rare. We reviewed the English literature in PubMed and summarized the prevalence and features of abdominal ET.

Adrenal gland ET

Guerra et al[4] reported that adrenal gland thyroid is very rare and is difficult to understand based on thyroid embryology. However, on the basis of the reported cases[5–14], the adrenal gland is not an uncommon site of abdominal ET. The cause of adrenal gland ET remains unknown. Although some diseases including adenocarcinoma are associated with adrenal masses with cystic lesions, dermoid cysts and cortical adenoma have been reported, ET should be considered when the adrenal gland shows both normal hormonal data and a cystic lesion.

Gallbladder ET

ET is occasionally found in the gallbladder. Campora et al[15] reported that only 3 cases of gallbladder ET have been described. However, to the best of our knowledge, at least 6 case reports on this disease existed in the literature[16–21]. Gallbladder ET generally has no specialized symptoms but is occasionally associated with recurrent right abdominal pain. ET could be observed in the wall of the gallbladder.

Pancreatic ET

Although ET could be found along the descending glands, ET in the pancreas is rare. To the best of our knowledge, 3 studies have been reported on pancreatic ET[22–24]. All the patients were middle-aged women, and all lesions had sizes of approximately 70*30 mm. One of the three patients had no

symptoms with a diagnosis of pancreatic cancer[22], one had recurrent pain in the right upper quadrant with a diagnosis of neuroendocrine neoplasm[24], and one had a complaint about dyspeptic symptoms with a diagnosis of a duodenal ulcer[23].

Liver ET

ET in the liver rarely occurs. Only three cases of ET in the liver[25, 26] and porta hepatis[3] have been reported. Additionally, a German report was also found in the references when we conducted out literature search[27]. All patients were women. Since the liver is an organ easily metastasized by cancers, ruling out thyroid cancer metastasis before the diagnosis of ET is important. Moreover, whether ET is combined in the liver metastases is key. Kondo et al[25] reported a mimicking ET in a 48-year-old woman with follicular carcinoma of the thyroid. However, the author also discussed that the liver tumor may not be an ET but rather an incidentally detected liver metastasis.

Gastric ET

ET is considered as normal thyroid tissue in the gastric mucosa and along the gastrointestinal tract[4]. However, distinguishing between metastatic thyroid cancer and normal thyroid tissue inside the stomach is important. ET was discovered in the stomach of a 35-year-old man without a thyroid tumor who suffered from gastric pain for six months. Biopsies were taken, and the microscopy sections showed thyroid tissue composed of colloid material and follicular cells[28].

Other sites

The other sites of abdominal ET included the duodenum[29], mesentery[30], appendix[31], gynecological organs (ovary, uterus and fallopian tube)[32–34] and others. Although few cases of other sites of abdominal ET have been reported, struma ovarii is a noteworthy disease because this condition may be easily misdiagnosed, and determining benign disease from malignant disease is difficult. Struma ovarii is diagnosed on the basis of thyroid tissue in the ovarian structures. Most patients with struma ovarii are asymptomatic, and the condition is incidentally found on ultrasonography or CT. The pathological diagnosis of our patient also suggested that struma ovarii should be excluded, based on further examinations and the combination of those results with clinical symptoms; the patient was ultimately diagnosed with ET.

Diagnosis. The mechanism of how ET migrates into the abdomen is not fully understood. Cassol et al[19] reported that ET in the gastrointestinal tract, liver and pancreas could be explained as a heteroplastic or metaplastic phenomenon because these locations and the thyroid share a common embryologic origin from the foregut endoderm. The thyroid gland tissue is composed of two cell types, the C cells and the thyroid follicular cells[28]. Romero-Rojas et al[2] showed that the lack of C cells in histology and immunohistochemical profiles is one of the important diagnostic criteria.

Iodine-131 or technetium-99m pertechnetate have been employed to discover ET and is based on the typical characteristics of thyroid tissues uptaking radioisotopes. Most patients were admitted to the hospital after the abdominal mass was accidentally found. Nonetheless, radioactive examinations are

rarely performed for patients without symptoms. CT scans, B-mode ultrasonography and magnetic resonance imaging (MRI) are the main imaging tools for abdominal ET. Fine needle aspiration cytology (FNAC) provides correct diagnoses at a rate higher than 95% and is considered the most accurate diagnostic method. FNAC is a very useful diagnostic tool when ET is not identified, especially before a surgery. Occasionally, intraoperative frozen pathology is also an effective method for some suitable patients.

Management

The best treatment strategy for ET is linked to the patient conditions, including age, sex, location of ET, local symptoms, tumor malignancy, anesthesiological risk assessment score and thyroid functional status. Most abdominal ET is treated with surgical resection. Rare cases are treated with palliative therapy after a diagnosis with puncture pathology. The surgical methods include open surgery, laparoscopy and Da Vinci robotics. Limited by technology, the previous surgery methods were mainly open. With the advancement of minimally invasive techniques, laparoscopy has been reported for abdominal ET[35]. No current studies of Da Vinci robotic resection for ET exist because of the complexity of the surgery and rarity of this condition. We provided the first case report of performing robotic surgery to treat abdominal ET, and our results also suggested that robotic surgery is safe and effective for treating suitable abdominal ET, which may be helpful for abdominal tumor therapy.

Conclusion

Abdominal ET is still rare, although there are some reports, as seen above. Nonetheless, the cause of abdominal ET is unclear, and abdominal ET has the following characteristics: more common in women than in men, asymptomatic in the majority of the cases, difficult to diagnose, and found by excluding metastasis. Most ET is treated with surgery, and minimally invasive techniques have been increasingly performed. In summary, this is the first report on Da Vinci robotic resection for large rare retroperitoneal ET, and this case highlights that ET should be considered when patients present with similar imaging findings in the abdomen.

Abbreviations

ET
Ectopic thyroid
CT
Computed tomography
MRI
Magnetic resonance imaging
FNAC
Fine needle aspiration cytology

Declarations

Ethics approval and consent to participate: The patient provided written informed consent before the operation, and this study was approved by the ethics committee of our hospital.

Consent for publication: Not applicable

Availability of data and materials: The datasets used and analysed during the current study are available from the corresponding author on reasonable request.

Competing interests: The authors declare that they have no competing interests.

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Authors' contributions Zhanwei Zhao wrote the main manuscript and participated in the study design. Yun Huang and Xinpu Yuan modified the article. Xiliang Zhang and Huibin Zhao participated in the design of the work and prepared the figures. Chaojun Zhang completed the design of the work and drafted the manuscript. All of the authors have reviewed the manuscript text.

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References

1. Kaplan M, Kauli R, Lubin E, Grunebaum M, Laron Z: **Ectopic thyroid gland. A clinical study of 30 children and review.** *J Pediatr* 1978, **92:**205-209.
2. Romero-Rojas A, Bella-Cueto MR, Meza-Cabrera IA, Cabezuelo-Hernandez A, Garcia-Rojo D, Vargas-Uricoechea H, Cameselle-Teijeiro J: **Ectopic thyroid tissue in the adrenal gland: a report of two cases with pathogenetic implications.** *Thyroid* 2013, **23:**1644-1650.

3. Ghanem N, Bley T, Altehoefer C, Hogerle S, Langer M: **Ectopic thyroid gland in the porta hepatis and lingua.** *Thyroid* 2003, **13:**503-507.
4. Guerra G, Cinelli M, Mesolella M, Tafuri D, Rocca A, Amato B, Rengo S, Testa D: **Morphological, diagnostic and surgical features of ectopic thyroid gland: a review of literature.** *Int J Surg* 2014, **12 Suppl 1:**S3-S11.
5. Tada A, Tanaka T, Takamoto A, Yanai H, Sato S, Kanazawa S: **Ectopic thyroid tissue in the adrenal gland: CT and MRI findings.** *Diagn Interv Imaging* 2016, **97:**373-375.
6. Gourmaud J, Bongiovanni M, Triponez F, Pusztaszeri M: **Ectopic thyroid tissue in the adrenal gland.** *Endocr Pathol* 2014, **25:**353-355.
7. Shuno Y, Kobayashi T, Morita K, Shimizu S, Nishio Y, Ito A, Kobayashi K, Kawahara M, Teruya M: **Ectopic thyroid in the adrenal gland presenting as cystic lesion.** *Surgery* 2006, **139:**580-582.
8. Casadei GP, Bertarelli C, Giorgini E, Cremonini N, de Biase D, Tallini G: **Ectopic thyroid tissue in the adrenal gland: report of a case.** *Int J Surg Pathol* 2015, **23:**170-175.
9. Tsujimura A, Takaha M, Takayama H, Sugao H, Takeda M, Kurata A: **Ectopic thyroid tissue in a cystic adrenal mass.** *Br J Urol* 1996, **77:**605-606.
10. Shiraishi T, Imai H, Fukutome K, Watanabe M, Yatani R: **Ectopic thyroid in the adrenal gland.** *Hum Pathol* 1999, **30:**105-108.
11. Takao H, Doi I, Watanabe T: **Ectopic thyroid in the adrenal gland: Computed tomography findings.** *J Comput Assist Tomogr* 2006, **30:**221-222.
12. Hagiuda J, Kuroda I, Tsukamoto T, Ueno M, Yokota C, Hirose T, Deguchi N: **Ectopic thyroid in an adrenal mass: a case report.** *Bmc Urol* 2006, **6:**18.
13. Bohinc BN, Parker JC, Hope WW, Kotwall C, Turner J, Cheng W, Lloyd RV: **Micropapillary thyroid carcinoma and concomitant ectopic thyroid tissue in the adrenal gland: metastasis or metaplasia?** *Thyroid* 2011, **21:**1033-1038.
14. Wang SC, Hung SW, Lee CH, Ho HC, Ou YC, Yang CK: **Ectopic Thyroid Tissue in the Adrenal Gland Mimicking a Pheochromocytoma.** *J Endourol Case Rep* 2017, **3:**149-151.
15. Campora M, Trambaiolo AC, Mastracci L, Pigozzi S, Grillo F: **A Never Ending Journey: Ectopic Thyroid.** *Int J Surg Pathol* 2017, **25:**241-242.
16. Curtis LE, Sheahan DG: **Heterotopic tissues in the gallbladder.** *Arch Pathol* 1969, **88:**677-683.
17. Harach HR: **Ectopic thyroid tissue adjacent to the gallbladder.** *Histopathology* 1998, **32:**90-91.
18. Ihtiyar E, Isiksoy S, Algin C, Sahin A, Erkasap S, Yasar B: **Ectopic thyroid in the gallbladder: report of a case.** *Surg Today* 2003, **33:**777-780.
19. Cassol CA, Noria D, Asa SL: **Ectopic thyroid tissue within the gall bladder: case report and brief review of the literature.** *Endocr Pathol* 2010, **21:**263-265.
20. Kachare MB, Khandelwal A, Kulkarni SB, Saboo SS: **Ectopic thyroid in duplicated gall bladder: a rare entity. Case report.** *Med Ultrason* 2013, **15:**73-75.

21. Liang K, Liu JF, Wang YH, Tang GC, Teng LH, Li F: **Ectopic thyroid presenting as a gallbladder mass.** *Ann R Coll Surg Engl* 2010, **92:**W4-W6.
22. Seelig MH, Schonleben K: **Intra-abdominal ectopic thyroid presenting as a pancreatic tumour.** *Eur J Surg* 1997, **163:**549-551.
23. Eyuboglu E, Kapan M, Ipek T, Ersan Y, Oz F: **Ectopic thyroid in the abdomen: report of a case.** *Surg Today* 1999, **29:**472-474.
24. Ma A, Liu H: **Ectopic thyroid of the pancreas: A case report and literature review.** *Medicine (Baltimore)* 2017, **96:**e8707.
25. Kondo T, Katoh R, Omata K, Oyama T, Yagawa A, Kawaoi A: **Incidentally detected liver metastasis of well-differentiated follicular carcinoma of the thyroid, mimicking ectopic thyroid.** *Pathol Int* 2000, **50:**509-513.
26. Salam M, Mohideen A, Stravitz RT: **Ectopic thyroid presenting as a liver mass.** *Clin Gastroenterol Hepatol* 2012, **10:**xxx.
27. Strohschneider T, Timm D, Worbes C: **[Ectopic thyroid gland tissue in the liver].** *Chirurg* 1993, **64:**751-753.
28. Rajabi P, Eftekhari SM, Rouhani E, Baradaran A: **Ectopic Thyroid in Stomach; a Case Report.** *Iran J Pathol* 2018, **13:**103-105.
29. Takahashi T, Ishikura H, Kato H, Tanabe T, Yoshiki T: **Ectopic thyroid follicles in the submucosa of the duodenum.** *Virchows Arch A Pathol Anat Histopathol* 1991, **418:**547-550.
30. Gungor B, Kebat T, Ozaslan C, Akilli S: **Intra-abdominal ectopic thyroid presenting with hyperthyroidism: report of a case.** *Surg Today* 2002, **32:**148-150.
31. Campora M, Trambaiolo AC, Valle L, Fiocca R, Mastracci L, Grillo F: **Abdominal Ectopic Thyroid Tissue: The Man From Istanbul.** *Int J Surg Pathol* 2019**:**1273703608.
32. Roth LM, Miller AR, Talerman A: **Typical thyroid-type carcinoma arising in struma ovarii: a report of 4 cases and review of the literature.** *Int J Gynecol Pathol* 2008, **27:**496-506.
33. Yilmaz F, Uzunlar AK, Sogutcu N: **Ectopic thyroid tissue in the uterus.** *Acta Obstet Gynecol Scand* 2005, **84:**201-202.
34. Hoda SA, Huvos AG: **Struma salpingis associated with struma ovarii.** *Am J Surg Pathol* 1993, **17:**1187-1189.
35. Tamaki S, Miyakura Y, Someya S, Ishikawa H, Kakizawa N, Hasegawa F, Tanaka H, Kikugawa R, Tsujinaka S, Tanaka A, et al: **Laparoscopic resection of retroperitoneal ectopic thyroid tissue.** *Asian J Endosc Surg* 2017, **10:**331-333.

Figures

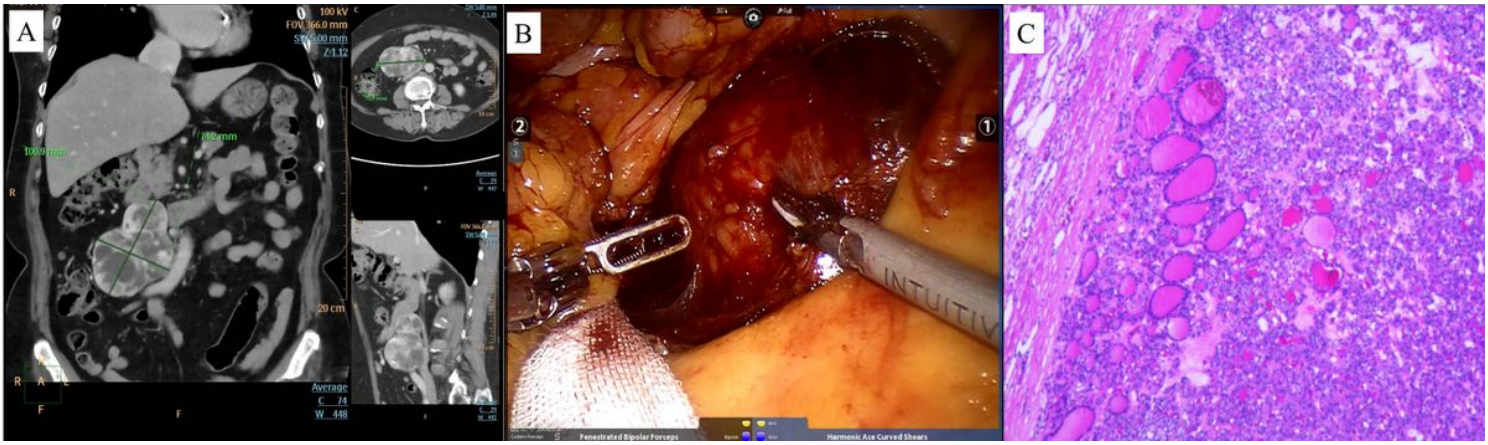

Figure 1

Contrast-enhanced computed tomography of the abdomen revealed a retroperitoneal mass with a size of 100.9 mm×74.2 mm×75.9 mm (A). The patient underwent Da Vinci robotic surgery and the retroperitoneal mass was completely resected (B). The pathologic diagnosis of the mass (C).