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Case Report

Transduodenal resection of periampullary neuroendocrine tumor: A case report

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ABSTRACT

Introduction and importance: Periampullary tumors are characterized as tumors that emerge nearby to the major papilla in the duodenum. They are rare lesions with an incidence rate of 0.4–0.48 per 100,000. Neuroendocrine tumors (NETs) constitute only 3% of all duodenal tumors. Their proximity to the major and minor papillae along with the gastric outlet raises a surgical challenge.

Case presentation: Our patient is a 40-year-old, male, medically free. He presented with history of a testicular mass. A CT scan of the abdomen and pelvis was done and showed a large retroperitoneal lymph node. A biopsy of the epididymal mass showed no evidence of malignancy. Excision of the left para-aortic mass revealed a metastatic lymph node of a well-differentiated neuroendocrine tumor. Further evaluation by gallium-68 PET-CT scan showed a periampullary neuroendocrine tumor. The decision to operate was concluded in a multidisciplinary team meeting, and intraoperatively the duodenum showed a well-defined mass between the first and second part of the duodenum which was excised via a *trans*-duodenal submucosal approach. A frozen section confirmed a negative margin. The final histopathology report showed a grade 2 metastatic well-differentiated neuroendocrine tumor. The latest follow-up was 3 years post-op via fluorine-18 fluorodeoxyglucose PET-CT and it showed no FDG avid disease at the duodenum or pancreases with no FDG avid lymphadenopathy or distant metastasis.

Conclusion: Periampullary tumors that fall under certain parameters could be resected via transduodenal local resection. This procedure yields equivalent results to more invasive surgeries, such as a Whipple's procedure, with less morbidity.

1. Introduction

Periampullary tumors (PATs) are tumors that emerge within 2 cm of the major papilla in the duodenum and are categorized into four types: pancreatic, biliary, duodenal, and ampullary [1]. The incidence of PATs is 0.4–0.48 per 100,000, although that has been increasing in recent years [2]. Their proximity to the major and minor papillae along with the gastric outlet raises a surgical challenge [3,4]. Duodenal neuroendocrine tumors (d-NETs) arise from the enterochromaffin cells of the gastroenteropancreatic neuroendocrine system, accounting for 2.6% of all NETs [5]. The incidence of NETs has been increasing in recent years due to advancements in imaging modalities, with studies showing that 3% of all duodenal tumors are NETs [5,6]. We report a case of a 40-year-old male who was incidentally found to have a periampullary NET and was managed by transduodenal submucosal excision. This article was reported in line with the SCARE guideline [7].

1.1. Description of the case

A 40-year-old male who is medically free presented to another facility with a history of a testicular mass. Ultrasound (US) revealed a right isoechoic epididymal mass with minimal peripheral calcification.

The referring facility thought of malignancy as their top differential diagnosis. Thus, they proceeded with a computed tomography (CT) scan of the abdomen and pelvis which showed a large retroperitoneal aortocaval soft tissue mass inseparable from the pancreatic head and

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uncinate process (Fig. 1); which was perceived as a large retroperitoneal lymph node consistent with the history of epididymal mass. Consequently, the patient was referred to our hospital, which is a specialized center, for further care.

In our hospital, the patient underwent further investigations. The routine laboratory tests and tumor markers were within the normal range. Additionally, the patient underwent abdominal magnetic resonance imaging (MRI) which showed a large retroperitoneal left para-aortic mass inseparable from the small bowel and the uncinate process of the pancreas. This finding was suggestive of retroperitoneal lymph nodes as the patient is known to have testicular mass the other differential diagnosis is a primary tumor from the abdomen. A biopsy of the epididymal mass showed findings suggestive of a previous infection with no evidence of malignancy.

The case was discussed in our multidisciplinary team (MDT), and the decision to excise the left para-aortic mass was made. The patient underwent a midline laparotomy incision by a hepatobiliary surgeon which was carried down to the peritoneum, kocherization of the duodenum was done, and the mobile mass was exposed. The duodenum, aorta, and head of the pancreas were abutted, and the lesion was released layer by layer until the mass was completely enucleated. The immunohistochemical staining showed positivity of synaptophysin and chromogranin (Fig. 2 A). The mitotic activity was estimated by Ki-67 as 1–2% and the mitotic count was 2/10 HPFs. The final histopathology report showed a grade 2 metastatic well-differentiated neuroendocrine tumor.

Further evaluation by gallium-68 positron emission tomography integrated with computed tomography (Ga-68 PET/CT) revealed suspicious gallium avid duodenal lesion (Fig. 3). The case was rediscussed again in the MDT meeting and Whipple's procedure was planned.

The patient underwent a midline laparotomy, adhesiolysis was done, and intra-operative assessment of the lesion showed a well-defined mass between the first and second part of the duodenum. A longitudinal incision of the duodenum and submucosal resection of the lesion was done. A frozen section was sent and affirmed a negative margin of 1.5 mm. After that closure of the mucosal defect and closure of the longitudinal incision of the duodenal in a transverse fashion, pyloric exclusion with stapler line was done, and a loop of jejunum 40 cm from the duodenojejunal junction was used to create a loop gastrojejunostomy, a leak test was done and secured. The gross examination of the lesion showed a well-circumscribed lesion with a white firm homogenous cut surface, and the was measuring 2 × 0.9 × 0.8 cm. There was no lymphovascular or perineural invasion. The immunohistochemical staining showed the positivity of synaptophysin and chromogranin. The mitotic activity was estimated by Ki-67 1–2% and the mitotic count was <2/10 HPFs (Fig. 2B–D). The final histopathology report showed a grade 2 metastatic well-differentiated neuroendocrine tumor with a pathological stage of T2N1M0.

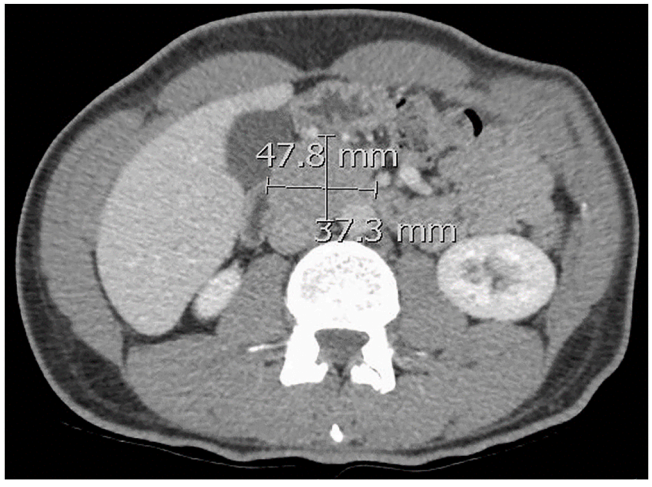

Fig. 1. Computed tomography (CT) scan of the abdomen (axial view) showing a large retroperitoneal aortocaval soft tissue mass inseparable from the pancreatic head and uncinate process.

The patient was followed-up for 3 years post-operatively and the last fluorine-18 fluorodeoxyglucose positron emission tomography integrated with computed tomography (18F-FDG PET/CT) showed no evidence of recurrence or metastasis.

2. Discussion

Neuroendocrine neoplasms are a heterogeneous group of tumors that originate from the highly distributed cells of the neuroendocrine system that may be found throughout the body [6]. The incidence of the duodenal NETs is low accounting for almost 2–3% of all tumors [6]. Ampullary NETs are even more rare, accounting for less than 0.3% of all gastroenteropancreatic (GEP) NETs, but they tend to be more aggressive than tumors arising from the duodenum with a high metastatic potential regardless of the size of the tumor [8].

The diagnosis of GEP-NETs is tricky, however, Naswa et al. reported Ga-68 PET/CT is a highly sensitive and specific modality for the diagnosis of GEP NETs [9]. In their study Ga-68 PET/CT was used to evaluate 109 patients with GEP-NETs and showed 78.3% sensitivity and 92.5% specificity for the primary tumor, while sensitivity and specificity for metastases was 97.4% and 100% respectively [8].

In addition to being a diagnostic challenge, surgical resection remains to be the treatment of choice regarding NETs, which adds a second layer of complexity. The shared blood supply of the retroperitoneal duodenum with the pancreas makes surgical management quite difficult, and controversy arises regarding the optimal surgical approach [2,4,8,10,11].

When resecting a d-NET, the controversy resides in two main surgical options; the first being a pancreaticoduodenectomy (PD), commonly referred to as a Whipple procedure. Conventionally, a PD consists of the removal of the head of the pancreas, duodenum, portion of the stomach, gallbladder, and a portion of the bile duct [2,10]. The remaining parts are then anastomosed to restore the normal flow of the ingested contents, digestive enzymes, and bile [12]. Thus, making this procedure not only complex and demanding, but also a time-consuming procedure that carries significant risks and serious complications, including delayed gastric emptying (DGE), pancreatic fistula, bleeding, intra-abdominal collection, and pulmonary complications [12]. Moreover, chronic complications such as exocrine insufficiency, and diabetes mellitus have been reported to occur post PD, and although the mortality rates after the procedure have been remarkably decreasing over the past decades, it still carries significant postoperative morbidity and complication rates ranging between 30 and 60% [2,13]. Postoperative risk notwithstanding, PD remains a preferred option by many surgeons due to the ability to examine surrounding lymph nodes allowing for accurate staging and appropriate oncological resection of tumors [11].

The second surgical option is a transduodenal local resection. This procedure was first proposed in 1899 however it fell out of practice and has only recently been readmitted as a considerable treatment option, that up to 2013 only 3 cases had been reported in the English literature and currently only around 4–6% of resected ampullary tumors have been done through transduodenal resection. A transduodenal local resection is a feasible procedure in patients with periampullary neuroendocrine tumors and is considered an organ-preserving operation with lower morbidity and mortality compared to PD [13,14]. To achieve satisfactory clinical outcomes after transduodenal local resection, three criteria must be taken into consideration: absence of lymph node metastasis, tumor size ≤2 cm, and negative resection margin. As such, when opting for transduodenal local resection, intraoperative histological evaluation of marginal status and lymph node metastasis through frozen biopsy should be considered to obtain satisfactory results. Performing a transduodenal local resection outside of these parameters may

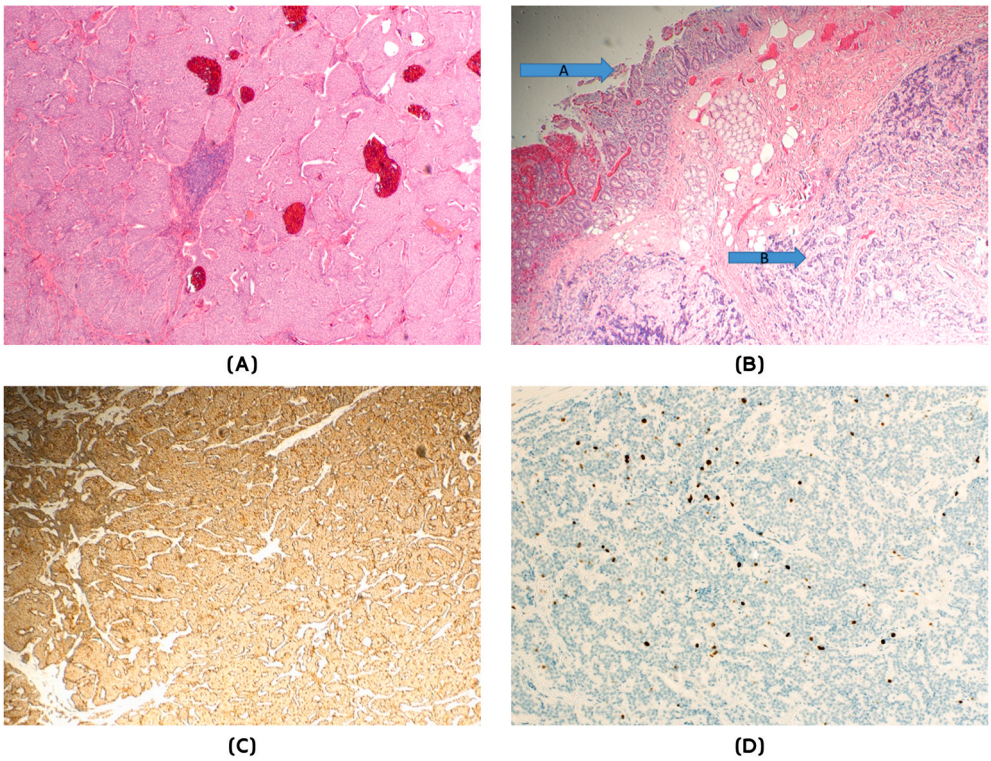

Fig. 2. Histopathological examination of the duodenal mass showing. (A) H/E 4X of peritoneal lymph node with metastatic neuroendocrine tumor. (B) The specimen with Hematoxylin and Eosin (H&E) staining of the duodenal mucosal mass (magnification power: ×4). (C) Positive Immunohistochemistry staining (synaptophysin stain) of the mass. (D) The nuclear protein Ki67 index is low 1–2%.

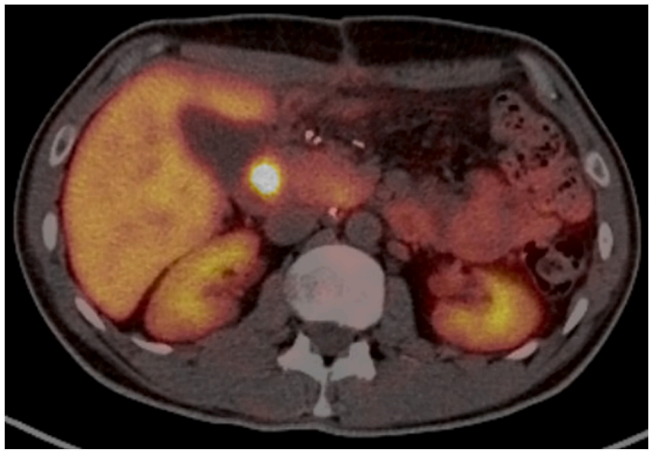

Fig. 3. A gallium-68 positron emission tomography integrated with computed tomography (Ga-68 PET/CT) showed a suspicious gallium avid duodenal lesion.

not yield optimal results and follow-up surgeries such as PD may be required [14,15].

Papalampros et al. reported that early postoperative complications following non-Whipple operations (i.e; pancreas preserving duodenectomy and transduodenal ampullectomy) was evident in 54% of patients. In the transduodenal group specifically, the complications were in the form of pleural effusion, atelectasis, pneumonia, fever, duodenal leak, and delayed gastric emptying [16]. As a matter of concern, this procedure is known to have inconsistent rates of recurrence varying from 0 to 40% [17]. However, when compared to PD, it has similar survival rates, a shorter operative time and hospital stay, less medical cost, and less blood loss [14]. Thus, for patients who have met the criteria, transduodenal local resection is believed to be a less invasive, safe, and an alternative for PD [13].

3. Conclusion

A transduodenal local resection is an appropriate form of therapy in the treatment of periampullary tumors. This procedure has been proven to be less morbid and could potentially provide equivalent clinical outcomes for early detected tumors compared to radical PD, which was in the past considered as the standard treatment for both benign and malignant tumors. Despite PD showing serious complications and mortality rates, it is indicated in fit patients with an advanced malignant process or large benign tumors that have not met the criteria for transduodenal resection.

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This study did not receive any sources of funding.

Ethical approval

This type of study does not require any ethical approval by our institution.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Drafting and writing the article: Abdullah AlQatari, Rania Fallata, Alaa Al Abdrabalnabi, and Miral Mashhour. Study concept and revising

the article: Abdullah AlQattan. Final approval: Abdulwahab AlShahrni.

Research registration

None.

Guarantor

Dr. Abdulwahab A. AlShahrni.
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Declaration of competing interest

None.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2021.103126.

References

[1] M.D. Chandrasegaram, A.J. Gill, J. Samra, T. Price, J. Chen, J. Fawcett, N. D. Merrett, Ampullary cancer of intestinal origin and duodenal cancer - a logical clinical and therapeutic subgroup in periampullary cancer, World J. Gastrointest. Oncol. 9 (2017) 407–415, https://doi.org/10.4251/wjgo.v9.i10.407.

[2] F.C.H. Wong, E.C.H. Lai, D.T.M. Chung, C.N. Tang, Robotic transduodenal excision of ampullary tumour, HepatoBiliary Surg, Nutridate 6 (2017) 312–316, https://doi.org/10.21037/hbsn.2016.12.04.

[3] S. Downs-Canner, W.J. Van der Vliet, S.J.J. Thoolen, B.A. Boone, A.H. Zureikat, M. E. Hogg, D.L. Bartlett, M.P. Callery, T.S. Kent, H.J. Zeh, A.J. Moser, Robotic Surgery for benign duodenal tumors, J. Gastrointest. Surg. 19 (2015) 306–312, https://doi.org/10.1007/s11605-014-2668-0.

[4] G.E. Cervoni, T. Singer, C. Decicco, J.F. Critchlow, T.S. Kent, A.J. Moser, Minimally invasive resection of duodenal tumors, J. Vis. Surg. 5 (2019), https://doi.org/10.21037/jovs.2019.02.01, 20–20.

[5] P. Dewan, S.P. Bhat, H.L. Kishan Prasad, R. Ballal, K. Sajitha, Neuroendocrine carcinoma of duodenum—an uncommon tumour at an unusual site, Indian J. Surg. Oncol. 10 (2019) 199–203, https://doi.org/10.1007/s13193-018-0834-7.

[6] G.A. Watson, Y. Ahmed, S. Picardo, S. Chew, S. Cobbe, C. Mahony, J. Crotty, F. Wallis, M.J. Shelly, P. Kiely, O.B. Ipadeola, V. Healy, N. Osman, R.K. Gupta, Unusual sites of high-grade neuroendocrine carcinomas: a case series and review of the literature, Am. J. Case Rep. 19 (2018) 710–723, https://doi.org/10.12659/AJCR.908953.

[7] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, for the SCARE Group, The SCARE 2020 guideline: updating consensus surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230.

[8] D. Tamburrino, G. Spoletini, S. Partelli, F. Muffatti, O. Adamenko, S. Crippa, M. Falconi, Surgical management of neuroendocrine tumors, Best Pract. Res. Clin. Endocrinol. Metabol. 30 (2016) 93–102, https://doi.org/10.1016/j.beem.2015.10.003.

[9] N. Naswa, P. Sharma, A. Kumar, A.H. Nazar, R. Kumar, S. Chumber, C. Bal, Gallium-68-DOTA-NOC PET/CT of patients with gastroenteropancreatic neuroendocrine tumors: a prospective single-center study, Am. J. Roentgenol. 197 (2011) 1221–1228, https://doi.org/10.2214/AJR.11.7298.

[10] J.M. Cloyd, E. George, B.C. Visser, Duodenal adenocarcinoma: advances in diagnosis and surgical management, World J. Gastrointest. Surg. 8 (2016) 212, https://doi.org/10.4240/wjgs.v8.i3.212.

[11] G.G. Baptiste, L.M. Postlewait, C.G. Ethun, N. Le, M.C. Russell, D.A. Kooby, C. A. Staley, S.K. Maithel, K. Cardona, Is there an optimal surgical approach to neuroendocrine tumors of the ampulla? A single institution experience over 15 years, Am. Surg. 82 (2016) 637–643, https://doi.org/10.1177/000313481608200731.

[12] S.A.M. Karim, K.S. Abdulla, Q.H. Abdulkarim, F.H. Rahim, The outcomes and complications of pancreaticoduodenectomy (Whipple procedure): cross sectional study, Int. J. Surg. 52 (2018) 383–387, https://doi.org/10.1016/j.ijsu.2018.01.041.

[13] R.C. Zhang, X.W. Xu, D. Wu, Y.C. Zhou, H. Ajoodhea, K. Chen, Y.P. Mou, Laparoscopic transduodenal local resection of periampullary neuroendocrine tumor: a case report, World J. Gastroenterol. 19 (2013) 6693–6698, https://doi.org/10.3748/wjg.v19.i39.6693.

[14] Y. Gao, Y. Zhu, X. Huang, H. Wang, X. Huang, Z. Yuan, Transduodenal ampullectomy provides a less invasive technique to cure early ampullary cancer, BMC Surg. 16 (2016) 1–8, https://doi.org/10.1186/s12893-016-0156-z.

[15] Y.K. Jung, S.S. Paik, D. Choi, K.G. Lee, Transduodenal ampullectomy for ampullary tumor, Asian J. Surg. 44 (2021) 723–729, https://doi.org/10.1016/j.asjsur.2020.12.021.

[16] A. Papalampros, D. Moris, A. Petrou, N. Dimitrokallis, I. Karavokyros, D. Schizas, I. Delladetsima, T.N. Pappas, E. Felekouras, Non-whipple operations in the management of benign, premalignant and early cancerous duodenal lesions, Anticancer Res. 37 (2017) 1443–1452, https://doi.org/10.21873/anticanres.11468.

[17] C. Heise, E. Abou Ali, D. Hasenclever, F. Auriemma, A. Gulla, S. Regner, S. Gaujoux, M. Hollenbach, Systematic review with meta-analysis: endoscopic and surgical resection for ampullary lesions, J. Clin. Med. 9 (2020) 3622, https://doi.org/10.3390/jcm9113622.