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Intraductal tubulopapillary neoplasm (ITPN) of the pancreas: A new entity different from intraductal mucinous neoplasm of the pancreas

Neoplasia tubulopapilar intraductal pancreática (ITPN): una nueva entidad diferente a la neoplasia mucinosa intraductal del páncreas

Intraductal tubulopapillary neoplasms (ITPN) of the pancreas were considered a rare subgroup of intraductal papillary mucinous neoplasms (IPMN) until the new classification was published by the World Health Organization (WHO) in 2019.[1] First described in 2009,[2] ITPN represent less than 1% of all exocrine pancreatic neoplasms and 3% of intraductal neoplasms of the pancreas.[3] They are considered premalignant lesions, associated with possible progression to invasive carcinoma. Although the actual incidence and oncological prognosis are unknown, ITPN seem to present better biological behavior than other intraductal papillary mucinous neoplasms.[4]

We present a case of ITPN: a 41-year-old male patient who, in the context of sudden abdominal pain, underwent a computed tomography scan, which discovered a solid 4 × 2 cm lesion in the tail of the pancreas. Magnetic resonance cholangiopancreatography and endoscopic ultrasound were performed, which confirmed said lesion and demonstrated that it had no communication with the main pancreatic duct. There was also no dilation of the Wirsung duct, no infiltration of neighboring structures, and no vascular compromise. The endoscopic ultrasound-assisted biopsy reported ductal adenocarcinoma.

Suspecting a malignant neoplasm, we conducted a laparotomy, finding a solid tumor in the body-tail of the pancreas that invaded the splenic hilum, without affecting adjacent vascular structures (Fig. 1A). We performed anterior radical anterograde modular pancreatosplenectomy (RAMPS), reinforcing the pancreatic stump with a continuous absorbable V-lock 3/0 suture.

The pathological study identified an ITPN with an associated intrapancreatic invasive carcinoma measuring 1.5 cm (T1cN0) (Fig. 1A & 1B), without the V600E mutation of the BRAF gene. Immunohistochemistry showed patchy positivity for CK7, CK19, MUC-1, MUC-6, and MUC-5AC (Fig. 1), while showing negativity for BCL-10 (Fig. 1C), chromogranin, synaptophysin, beta-catenin, CD10, MUC-2, Her-Par1 and KRAS.

The patient evolved favorably. He presented a type A pancreatic fistula, which was controlled conservatively (Dindo-Clavien II), and he was discharged on the 8th postoperative day. Adjuvant therapy was administered with Folfirinox for 6 months, and the patient is disease-free.

According to the latest WHO classification from 2019, ITPN are separate from IPMN, and their main difference is the absence of a KRAS mutation.[1] Other typical immunohistochemical features are positivity for CK7 and/or CK19 as well as negativity for trypsin, MUC2, MUC5AC, and fascin.[3] This type of neoplasm was first described in 2009 by Yamaguchi et al.[2] Since then, most publications report isolated clinical cases, and the most extensive multicenter case series was published by Basturk et al.,[5] with 33 cases. Even more exceptional is the case of ITPN positive for MUC-5AC, as only one case has been published, in 2015.[6] Therefore, the case that we present in this manuscript is the second ITPN with positivity for MUC-5AC and the first case published in Spanish.

There is little evidence available regarding the prognosis of these neoplasms. In the series by Basturk,[5] among the 22 patients in whom follow-up was reported, a survival rate of 100% was observed in cases with tumors without an invasive component, and 71% in patients with an invasive carcinoma, with a median follow-up of 48 months. Most authors postulate a favorable outcome after surgical resection of ITPN in terms of survival compared with the survival results obtained after resection of other IPMN, which, according to some series, is

80%–100% if there is no invasive component and 40%–60% when there is.[7]

ITPN are a different entity than IPMN. Despite the few cases published to date and little follow-up, they seem to generally have better survival than IPMN. The creation of an international registry on ITPN would make it possible to determine the diagnostic perspective and treatment of these patients in order to propose a therapeutic algorithm, while also providing more reliable data on the intermediate- and long-term prognosis. Until then, it may be appropriate to follow the current IPMN clinical guidelines.

Conflicts of interest

None.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at http://dx.doi.org/10.1016/j.cireng.2022.10.008.

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