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Isolated Langerhans islets: Potential pitfall in solid and pseudopapillary neoplasm resection?

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To the Editor,

Sir

Solid and pseudopapillary tumors of the pancreas are frequently treated by surgical enucleation (or limited resection) as recently pointed out by Wang et al. [1]. Isolated Langerhans islets (also called naked Langerhans islets, islet hyperplasia, islet aggregates, subclinical adult nesidioblastosis) (ILI) are rarely reported as associated lesions [2].

We had recently the opportunity to detect several groups of ILI in the peripancreatic tissues of a distal pancreatectomy for solid and pseudopapillary tumor (SPPT) both in peritumor and peripancreatic adipose tissue location. The peripancreatic tissues of the surgical specimen, present at proximity of the pancreas resection limit, were included entirely for microscopy analysis. The ILI foci (present on 4 blocks) were located in the peritumor capsule and in the adipose tissue (Supplementary Material Figs. 1 and 2). They were composed exclusively of Langerhans islets/LI (chromogranin-positive) and did not contain pancreatic acini or ducts. The LIs were of varied forms and sizes, most small. To mention would be LIs of 2–3 cell size. The nodules showed scant or moderate fibrosis and no acini or ducts (including on cytokeratin CK7 immunohistochemistry). There were 6 foci (1–4 mm in size) in the adipose tissue, 2 of which at the sub-serosal/resection limit site. The ILI-foci were completely surrounded by adipose tissue. One of the ILI-focus was in continuity with a peritumoral capsular ILI, resulting in a total length of 7 mm. One group contained nerves and ganglion cells. Peritumor foci (4) measured 0.5–4 mm. For one of the block specimen, the 2 foci observed on the HE and chromogranin slides, became contiguous over a zone of 4 mm on the CK7 and Ki67 stained slides. On these slides, there was a zone of ILI confluence measuring 1–1.5 mm (Supplementary Fig. 2). Ki67 was expressed by very rare nuclei. Nuclear atypia were also exceedingly rare.

Here we report isolated Langerhans islet foci in both peritumor and peripancreatic adipose tissue location. The main clinical relevance resides in the potential misdiagnosis with lymph node or tumor foci, in particular for evaluating the tumor extension and for orienting the surgical resection limit. To note would be that foci of ILI, some of 1–2 mm, were at less of 1 mm from the resection limit. A second important issue, although of limited clinical impact at the moment of surgical resection, was the differential diagnosis of the zone of confluent ILI with a neuroendocrine microadenoma [3].

The histogenesis of such lesions is difficult to precise [4–9]. While for the peritumor ILI, the hypothesis of compression-related lesions may be accepted [4,9–12], for the intra-adipose ILI, other hypothesis could be associated. The hypothesis of a hamartomatous, ectopic or developmental lesion might be sustained by the presence of adipocytes at direct contact of some LI as well as the presence of nerves and ganglion cells between the ILI foci.

In conclusion, we report ILI located not only in pancreatic peritumor tissues but also in the peripancreatic adipose tissue. Foci of ILI were also detected at the resection limit. Tumor compression may in part explain the histogenesis of such lesions. The number of ILI and the presence of adipose cells, nerves and ganglion cells (without pancreatic duct or acini) may suggest ectopic or hamartoma-type lesions, the evolutive potential of which remains to be further investigated.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.ejso.2018.12.002.

References

[1] Wang X, Chen YH, Tan CL, Zhang H, Xiong JJ, Chen HY, et al. Enucleation of pancreatic solid pseudopapillary neoplasm: short-term and long-term outcomes from a 7-year large single-center experience. Eur J Surg Oncol 2018;44:644–50.
[2] Wilcox R, Noffsinger A. Naked islets of Langerhans are frequently found in patients with pancreatic neuroendocrine tumors. Lab Invest 2009;89(Suppl):325A.
[3] Kwon JH, Kim HJ, Park DH, Lee YJ, Heaphy CM, Klöppel G, et al. Incidentally detected pancreatic neuroendocrine microadenoma with lymph node metastasis. Virchows Arch 2018. https://doi.org/10.1007/s00428-018-2407-0. Jul 12, [Epub ahead of print].
[4] Bartow SA, Mukai K, Rosai J. Pseudoneoplastic proliferation of endocrine cells in pancreatic fibrosis. Cancer 1981;47:2627–33. 1981.
[5] Rosai J. Rosai and Ackerman's surgical pathology. Philadelphia PA: Expert Consult. Elsevier; 2011.
[6] La Rosa S, Sessa F. Pancreatic neuroendocrine neoplasms. Berlin: Springer; 2015.

https://doi.org/10.1016/j.ejso.2018.12.002

[7] Goossens A, Gepts W, Saudubray JM, Bonnefont JP, Nihoul-Fekete, Heitz PU, et al. Diffuse and focal nesidioblastosis. A clinicopathological study of 24 patients with persistent neonatal hyperinsulinemic hypoglycemia. Am J Surg Pathol 1989;13:766–75.
[8] Klöppel G, Anlauf M, Raffel A, Perren A, Knoefel WT. Adult diffuse nesidioblastosis: genetically or environmentally induced? Hum Pathol 2008;39:3–8.
[9] Hruban RH, Pitman MB, Klimstra DS. AFIP Atlas of tumor pathology 4th series fasc 6. Washington DC: AFIP series; 2007.
[10] Suda K, Komatsu K, Hashimoto K. A histopathological study on the islets of Langerhans and ductal epithelial metaplasia in atrophic lobuli of pancreas. Acta Pathol Jpn 1976;26(5):561–72.
[11] Suda K, Tsukahara M, Miyake T, Hirai S. A morphologic analysis of 'naked' islets of Langerhans in lobular atrophy of the pancreas. Pathol Int 1994;44:618–23.
[12] Kimura W. Histological study on pathogenesis of sites of isolated islets of Langerhans and their course to the terminal state. Am J Gastroenterol 1989;84:517–22.

Adriana Handra-Luca[a,b,*]
[a] *APHP GHU Avicenne, Bobigny, France*
[b] *Universite Paris Nord, UFR SMBH Bobigny, France*

[*] Service d'Anatomie pathologique APHP GHU Avicenne, Universite Paris Nord Sorbonne Cite, 125 rue Stalingrad, 93009, Bobigny, France.
E-mail addresses: adriana.handra-luca@aphp.fr, adriana.handra-luca@hotmail.com.

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