Solid Cell Nests and Cyst Heterogeneity in Thyroglossal Duct Cysts

Adriana Handra-Luca^{1,2,3} · Ibtissem Radhouani¹

Received: 27 December 2016 / Accepted: 1 March 2017 / Published online: 27 March 2017
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To the Editor,

Solid cell nests (SCN) have been recently reported in the thyroid tissue surrounding thyroglossal duct (TGD) [1]. We had the occasion to study a TGD cyst showing SCNs in the surrounding thyroid tissue in a woman (39 years) presenting with a 1.5 cm midline neck cyst. Besides the TGD-cyst which was multilocular consisting in milimetric and centimetric cysts (lined by squamous, glandular non-ciliated and ciliated-type epithelia), atrophic thyroid follicular remnants were seen at the periphery of one of the largest cyst. Interestingly one of the squamoid SCN showed a cystic change on serial sections (Fig. 1).

This observation lead us to the hypothesis that cysts in the TGD-type lesions may be heterogeneous, some of them originating possibly from « thyroid-type » SCNs (as defined for ectopic thyroid by Carmeselle-Teijerio et al.) equivalent of type 3 SCNs in the Asioli et al. classification [2, 3]. Of interest might be the fact that 4 skin nodules (bilateral buttocks and right arm measuring 0.5–2 cm of which one with epidermic cyst histology) and 3 pedunculated skin tags (left neck, right axilla, abdominal) were also identified. Whether a same precipitating event might have determined the cystic change in the TGD, thyroid SCN and

cutaneous lesions, all lined by a squamous-type epithelium, remains difficult to precise.

We would also like to point out another feature we have observed, that of HBME1 expression in the TGD-cysts, in both the ciliated respiratory-type epithelium and the mature and/or immature, squamous epithelium. While in the respiratory type epithelium, CK5/6, along with P63, was expressed only in basal cells, lacking in the cubocylindric ciliated cells, in the squamous-type lined cysts, CK5/6 and P63 were expressed in both basal and suprabasal spindle-shaped cells. The relevance of the HBME1 positivity is difficult to explain. However, HBME1 is also expressed in benign lesions such as thyroid SCNs [4].

A third peculiarity seen in the case we report was the presence of several facial bone variants, several reminiscent of cysts, however not suggestive of an already identified syndrome (Fig. 2). Besides a right Haller cell, a left Onodi cell and concha bullosa; bilateral maxillary septa, nasal septal latero-deviation as well as right nasal cone hypoplasia and frontal sinus agenesis were noted. To mention would be that hypoplasia of nasal bones is reported in cystic fibrosis patients [5]. Of interest would be that the patient was known with Langerhans histiocytosis in relationship with spontaneous pneumothorax and emphysema since the age of 24 years, however without bone location and without histological confirmation available.

A fourth peculiarity was that of laryngeal mixoid nodules, resected at the same time as the TGD cyst, which was located, on the neck CT-scan, at the level of the right thyroid cartilage. Whether related to the indacaterol or tiotropium treatments the patient had for the lung disease or developmental remains matter of debate.

In conclusion, TGD cysts may be heterogeneous, some originating from SCNs of the pericystic thyroid tissue. The relevance of HBME1 expression in both the SCNs

✉ Adriana Handra-Luca
adriana.handra-luca@aphp.fr; adriana.handra-luca@hotmail.com

¹ APHP GHU Avicenne, Bobigny, France

² Université Paris Nord Sorbonne Cité, Bobigny, France

³ Service d'Anatomie pathologique, APHP GHU Avicenne, Université Paris Nord Sorbonne Cité, 125 rue Stalingrad, deck 52047, secretary office 55601, 93009 Bobigny, France

Fig. 1 The resected specimen showed several cysts (*asterisks*), a focus of atrophic thyroid (*white asterisk*) and several solid cell nests (*grey arrow*), some of them cystic (**a–f** are photos of serial sections of a same block). To note would be the cystic appearance of the SCN on figure (**b** and **e**, serial sections). HBME1 was expressed in

the cyst lining and in a peri-cystic solid cell nest (**g** and **h**, respectively). The cyst epithelium, either of squamous type or of respiratory type, expressed HBME1, cytokeratin 5/6 and p63 (**i, j, k** and **l, m, n**, respectively)

Fig. 2 The bone variants consisted in Haller cell (**a**: white arrow), Onodi cells (**b**: orange arrow), concha bullosa (**a**: orange arrow), bilateral maxillary sinus septa (**c**: blue arrow), nasal cone hypoplasia (**a**, **d**: green arrow) and, bilateral nerve V protrusion (**d**: yellow arrows)

(including cystic) and in the TGD cyst lining, of respiratory or squamous type, whether indicating a coelomic origin or not, remains matter of debate.

Acknowledgements The author thanks Pr F Pagni, Dr S Wei, Dr MI Cherif, Pr JY Brillet and Pr C Prost. We acknowledge Dr B Villette

and Dr S Benzakin involved in the management of the case. We also thank K Dahmane, J Peron, C Van Vetteren, F Spindler, L Jovanovic, F Bouchard, I Pluchard, N Delva, S El-Sayeh, M Rodrigues, J Raleche, V Ipotesi, N Jalem, C Almeida, A Maillard, I Alexandre, MC Portenier, M de Souza and the NCA/Avicenne, BIUM, CMDP/APHP and PathologyOutlines.com teams.

Compliance with Ethical Standards

Conflict of interest The authors declare that they don't have conflict of interest.

Ethical Approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. The study deals with tissue studies. The patient-related data are de-identified.

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