

or equal to 2 mm in eight patients. The pattern of large cysts, particularly located in the lower portions of the lungs, with a multiseptated and lobulated appearance, was previously described in the literature.⁷ Among cystic lung diseases, lymphangiomyomatosis was the greatest challenge in the differential diagnosis of BHDS, especially when associated with tuberous sclerosis complex.^{1,6}

Our study presented some limitations, in that it was retrospective and patients were examined with a variety of tomographic techniques due to the different institutions involved in the study. Despite these limitations, we believe that this variation did not significantly impact on the results of the study. In conclusion, pulmonary cysts in imaging studies may be the initial finding in patients with BHDS and the careful analysis of its characteristics may suggest the presence of BHDS, highlighting the role of CT in early diagnosis. Knowledge about symptoms of BHDS by pulmonologists and radiologists is fundamental in patient care, as it leads to the early detection and management of complications, particularly renal malignancy.

Conflict of interest

The authors have no conflicts of interest to declare.

References

1. Gupta N, Seyama K, McCormack FX. Pulmonary manifestations of Birt–Hogg–Dubé syndrome. *Fam Cancer*. 2013;12:387–96.
2. Dal Sasso AA, Belém LC, Zanetti G, Souza CA, Escuissato DL, Irion KL, et al. Birt–Hogg–Dubé syndrome. State-of-the-art review with emphasis on pulmonary involvement. *Respir Med*. 2015;109:289–96.
3. Agarwal PP, Gross BH, Holloway BJ, Seely J, Stark P, Kazerooni EA. Thoracic CT findings in Birt–Hogg–Dube syndrome. *AJR Am J Roentgenol*. 2011;196:349–52.
4. Toro JR, Pautler SE, Stewart L, Glenn GM, Weinreich M, Toure O, et al. Lung cysts, spontaneous pneumothorax, and genetic

associations in 89 families with Birt–Hogg–Dubé syndrome. *Am J Respir Crit Care Med*. 2007;175:1044–53.

5. Menko FH, van Steensel MA, Giraud S, Friis-Hansen L, Richard S, Ungari S, et al. Birt–Hogg–Dubé syndrome: diagnosis and management. *Lancet Oncol*. 2009;10:1199–206.
6. Tobino K, Gunji Y, Kurihara M, Kunogi M, Koike K, Tomiyama N, et al. Characteristics of pulmonary cysts in Birt–Hogg–Dubé syndrome: thin-section CT findings of the chest in 12 patients. *Eur J Radiol*. 2011;77:403–9.
7. Tobino K, Hirai T, Johkoh T, Kurihara M, Fujimoto K, Tomiyama N, et al. Differentiation between Birt–Hogg–Dubé syndrome and lymphangiomyomatosis: quantitative analysis of pulmonary cysts on computed tomography of the chest in 66 females. *Eur J Radiol*. 2012;81:1340–6.

A.A.D. Sasso^a, G. Zanetti^a, C.A. Souza^b, D.L. Escuissato^c, K.L. Irion^{d,e}, M.D. Guimarães^f, A. Soares Souza Jr^g, D. Penha^g, E. Marchiori^{a,*}

^a *Federal University of Rio de Janeiro, Rio de Janeiro, Brazil*

^b *The Ottawa Hospital, University of Ottawa, Ottawa, Ontario, Canada*

^c *Federal University of Paraná, Curitiba, Brazil*

^d *Liverpool Heart and Chest Hospital, Liverpool, United Kingdom*

^e *Royal Liverpool University Hospital NHS Trusts., Liverpool, United Kingdom*

^f *A.C. Camargo Cancer Center, São Paulo, Brazil*

^g *Faculdade de Medicina de São José do Rio Preto, São Paulo, Brazil*

* Corresponding author.

E-mail address: edmarchiori@gmail.com (E. Marchiori).

<http://dx.doi.org/10.1016/j.rppnen.2017.02.003>
2173-5115/

© 2017 Sociedade Portuguesa de Pneumologia. Published by Elsevier España, S.L.U. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

Pulmonary placental transmogrification: The last 16 years in a reference centre



Dear Editor,

Pulmonary placental transmogrification (PPT) is an extremely rare benign lesion of the lung, first described by McChesney in 1979.¹ Histopathologically it resembles immature placental tissue, although it does not bear any biological and biochemical properties of a placenta.² This condition has been described in patients with emphysema associated with cigarette smoking, congenital bullous emphysema and hamartomas of the lung.³

We present this retrospective study of all cases of pulmonary hamartoma (103) and bullous emphysema (410) diagnosed in our Pathology Department of the *Centro Hospitalar Lisboa Norte, EPE* between 01/01/2000 and

31/12/2015 (a Portuguese reference medical centre with a long-standing tradition of cardio-thoracic surgery). Histologic slides of all these cases were retrospectively reviewed for the presence of villus-like papillary projections and/or placenta-like structures. Placental transmogrification was identified in 3 of 513 cases (0.58%): one case (1/103, 0.97%) with clinical information of hamartoma and two of emphysema (2/410, 0.49%). The features of these cases are summarized in [Table 1](#).

Conducting a review of the medical literature, we have found only 30 cases since the first description of PPT. The age of presentation is between 24 and 72 years with a clear male predominance.

From a clinical point of view, placental transmogrification may cause nonspecific symptoms such as dyspnoea and/or pneumothorax, as with two of our patients.

Radiologically, PPT can appear as a large bullous emphysema lesion and rarely as a solitary pulmonary lung nodule, such as hamartoma. In our study, we present both; two cases

Table 1 Clinical, radiological and pathological features of PPT.

Sex; age (years)	M; 55	M; 54	M; 61
Symptoms	Dyspnoea	Dyspnoea	None
Physical examination	Decreased breath sounds	Decreased breath sounds	Normal function
Radiological diagnosis	Bullae at left upper lobe	Bullae at right upper lobe	Nodule
Tumour size (cm)	6.5	8	1.9
Intervention	Wedge surgical resection	Wedge surgical resection	Wedge surgical resection
Pathology	Emphysematous changes with structures similar to the chorionic villi	Emphysematous changes with structures similar to the chorionic villi	Hamartoma with structures similar to the chorionic villi

M = male; CT = computed tomography scan.

of emphysematous and one of hamartomatous presentation. Kim et al.⁴ have classified the radiological findings into three patterns: the most common is bullous emphysema manifestation; next, it is a mixed pattern of thin-walled cystic lesion and nodule; and in the third, the radiography shows a solitary nodule pattern, which is rare. Differential diagnosis of the lesion includes bullous emphysema, particularly giant bullous emphysema and solitary pulmonary lung nodules, such as pulmonary hamartoma. PPT usually has a unilateral giant bullous change, however, bullous emphysema generally demonstrates a diffuse bilateral lung involvement.⁴ From the radiological standpoint there is no technique or specific image that is able to differentiate the lesion from PPT. Clinically, the preoperative diagnosis by imaging is very difficult. Therefore, the definitive diagnosis is always provided by pathological studies.

Histologically, it is characterized by the development of peculiar papillary structures that resemble immature placental chorionic villi (Fig. 1). These villus-like structures correspond to alveolar walls enlarged with thin or dilated capillaries. The epithelium covering the papillary projections may be flat, cuboidal, or columnar type, and some of the cells are ciliated. Immunohistochemically, these cells are positive for TTF-1 antibody, indicating that they originate from respiratory epithelium or represent an extension from adjacent lung epithelium.²

The pathogenesis of the lesion is not clear. In 1995, Fidler et al.⁵ suggested that PPT was a histologic variant of giant bullous emphysema. Some investigators further speculate that it may be related to a lymphatic or vascular abnormality, based on the observation of the frequent lymphatic or vascular proliferation and dilation of vascular channels.⁶

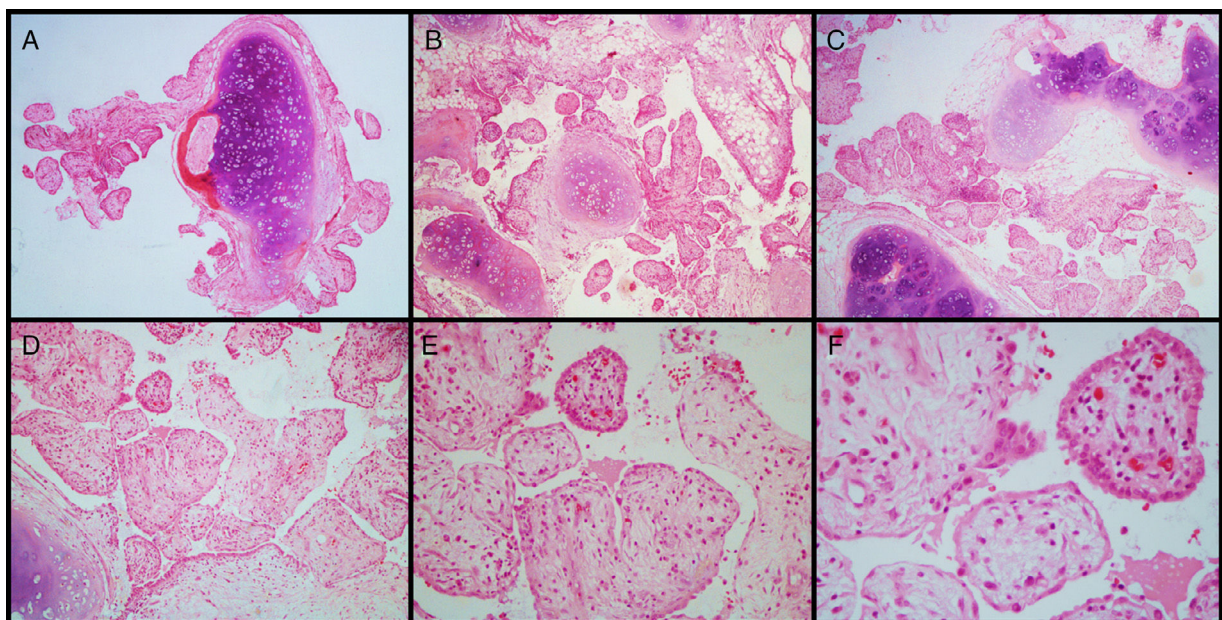


Figure 1 Histologic examination of PPT. On histologic examination, hamartoma is manifested by mature mesenchymal tissues but with abnormal configurations. These elements are represented by hyaline cartilage, fibrous tissue, adipocytic components and bone, with varying degrees of epithelial invagination. During the growth of hamartoma, prominent entrapped epithelial foldings showed peculiar papillary projections that resembles placental chorionic villi. The stroma of these placental villus-like structures contained fibroadipose tissue, blood vessels and variable number of inflammatory cells (i.e. plasma cells and lymphocytes) diffusely dispersed. The lining epithelium consisted of cuboidal and ciliated columnar cells (H&E – A, B and C – 40 \times , D – 100 \times , E – 200 \times , F – 400 \times).

Other authors have suggested that the process is like a congenital malformation with slow progression and not a sequela of pre-existing emphysema, but no PPT has been reported in children.^{4,6} McChesney has thought that placental transmogrification might represent an unrecognized pulmonary hamartoma. The exact relationship between this and hamartoma, has not yet become clear.¹ Xu et al.² found that all pulmonary hamartomas show varying degrees of development of papillary structures, lined by a single layer of epithelial cells or with epithelial invagination into stroma. Depending on the size of papillary growth, hamartomas can have minimal epithelial clefting, with a small number of epithelial projections, up to those with a large number of tall villous-like projections. This exaggeration of epithelial clefting may be an explanation for the papillary pattern. In our opinion it probably results from the development of oedema, fibrosis, and chronic inflammation in the residual alveolar tissues.

PPT is regarded as an unusual benign lesion curable by surgical resection. As Brevetti et al.⁷ described, we believe that these lesions are best treated by minimal resection, leaving as much normal lung tissue as feasible and if possible avoiding a lobectomy. After surgery, all our patients have been followed-up by monitoring imaging techniques. There was no evidence of recurrent disease or the appearance of new tumours in other locations, during a follow-up period that ranged between 36 and 108 months (average, 72 months).

As far as we know, this is the first comprehensive and updated work carried out in Portugal, also the first series of PPT in this country, the first largest series of a single centre in Europe and the second in the medical English literature. Just 30 cases have been described in the literature, all associated with emphysema or hamartoma. Our study is the first and the only centre that has presented this lesion in its two forms (hamartomatous and emphysematous).

Conflict of interest

None declared.

Acknowledgements

We thank Paula Plim for the English review.

References

1. McChesney T. Placental transmogrification of the lung: a unique case with remarkable histopathologic feature. *Lab Invest.* 1979;40:245–6.
2. Xu R, Murray M, Jagirdar J, Delgado Y, Melamed J. Placental transmogrification of the lung is a histologic pattern frequently associated with pulmonary fibrochondromatous hamartoma. *Arch Pathol Lab Med.* 2002;126:562–6.
3. Marchevsky AM, Guintu R, Koss M, Fuller C, Houck W, McKenna RJ. Swyer–James (MacLeod) syndrome with placental transmogrification of the lung: a case report and review of the literature. *Arch Pathol Lab Med.* 2005;129:686–9.
4. Kim JW, Park IH, Kwon W, Eom MS, Kim YJ, Oh JH. Placental transmogrification of the lung. *Korean J Radiol.* 2013;14:977–80.
5. Fidler ME, Koomen M, Sebek B, Greco MA, Rizk CC, Askin FB. Placental transmogrification of the lung, a histologic variant of giant bullous emphysema. *Clinicopathological study of three further cases.* *Am J Surg Pathol.* 1995;19:563–70.
6. Theile A, Müller KM. Placentoid malformation of the lung. *Pathologie.* 1998;19:134–40.
7. Brevetti GR, Clary-Macy C, Jablons DM. Pulmonary placental transmogrification: diagnosis and treatment. *J Thorac Cardiovasc Surg.* 1999;118:966–7.

S. Ortiz*, F. Tortosa

Department of Pathology, Centro Hospitalar Lisboa Norte, EPE – Hospital de Santa Maria, Lisbon Portugal

* Corresponding author.

E-mail address: santi.orfer@gmail.com (S. Ortiz).

<http://dx.doi.org/10.1016/j.rppnen.2017.02.007>
2173-5115/

© 2017 Sociedade Portuguesa de Pneumologia. Published by Elsevier España, S.L.U. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

CT-guided transthoracic lung diagnostic procedures: A 5 year experience



Computerized tomography (CT)-guided transthoracic diagnostic procedures are effective and safe techniques, and may help avoid thoracic surgery for the diagnosis of intrathoracic lesions. Fine needle aspiration (FNA) provides material for cytological analysis, core biopsy (CB) provides a sample for histologic examination.

The aim of our study was to evaluate the diagnostic accuracy and complication rate of both FNA and CB in pulmonary lesions suspected of malignancy.

A retrospective analysis of CT-guided transthoracic interventional procedures completed in the Radiology Department, between January 2008 and January 2013, was

performed. Patients with a pulmonary lesion suspected of malignancy, on chest CT, were included in the study.

All patients underwent a coagulation study. Anticoagulant and antiplatelet therapy had been previously interrupted. An axial acquisition prior to the procedure was obtained. The CT equipment was Philips® Brilliance 16P. A 22G spinal needle was used for the FNA, and an 18G semi-automatic needle *tru-cut* gauge system for CB. The shortest intraparenchymal needle path was chosen. The usual number of needle passes was 2 (1–3). The occurrence of complications, including pneumothorax and bleeding was identified through CT images obtained immediately after needle removal. After the procedure, patients remained under observation in the Department of Pulmonology for 6 h, with clinical evaluation before discharge.

The results of the procedures were compared with the diagnosis obtained from the integration of clinical evolution,