

Metastatic Carcinoma ex Pleomorphic Adenoma in Pleural Effusion

Hsiao-Ching Chen*

Department of Pathology, Hsinchu MacKay Memorial Hospital, Taiwan

Abstract

Carcinoma ex pleomorphic adenoma (CXPA) arising from primary or recurrent pleomorphic adenoma is characterized by the presence of longstanding palpable mass with growth spurt mainly in the parotid region. Overall, patients with CXPA have a poor prognosis. Pleural effusion involved by CXPA is precedingly rare, compared to that common dissemination from pulmonary or mammary origin. Here we reported a metastatic CXPA in pleural effusion from an 82-year-old male. The clinical presentation, cytological morphology, and immunohistochemistry confirmed the diagnosis. When dealing patients with prolonged history of pleomorphic adenoma, underwent recent rapid growth of malignant transformation and malignant cells presented in the pleural effusion, metastatic CXPA should be seriously considered.

Keywords: Carcinoma ex pleomorphic adenoma (CXPA) • Pleural effusion • Immunohistochemistry

Introduction

Carcinoma ex pleomorphic adenoma (CXPA) defined as: a pleomorphic adenoma in which an epithelial malignancy is derived, is a rare, aggressive, poorly understood malignant neoplasm of salivary gland [1]. CXPA constitutes approximately 3.6% of all salivary gland tumors, 5% of pleomorphic adenomas undergo malignant transformation, and 11.6% of all malignant salivary gland neoplasms. It is a distinctive but relatively common high-grade adenocarcinoma arising from the excretory ductal epithelium of the major salivary gland, especially the parotid gland [1-3]. The clinical findings typical of this tumor include history of a slow growing, ulcerated, painless mass that enlarges rapidly; aggressive and infiltrative behavior with local recurrence, distant metastasis, or significant mortality [1-5].

Metastasis of salivary gland neoplasms to pleural effusion is an unusual finding. Cases of mucoepidermoid carcinoma involving peritoneal and pericardial fluid and adenoid cystic carcinoma metastasizing to the pleural fluid had been reported before [6-8]. In this article, metastatic CXPA in pleural effusion presented here and highlighted the diagnostic challenges on a cytology specimen.

Case Report

An 82-year-old male presented with receiving excision of pleomorphic adenoma at right parotid 35 years ago and an enlarged lymph node was found at right supraclavicular area recently. CT revealed a 2 cm heterogeneously enhanced tumor at right parotid tail. Fine needle aspiration was then performed. The Pap-stained smears showed a mixed population of cells composed of benign and malignant elements. Some clusters showed a metachromatic stroma in a background. The benign epithelial cells component

***Address for Correspondence:** Hsiao-Ching Chen, Department of Pathology, Hsinchu MacKay Memorial Hospital, 690, section 2 Guangfu Rd, Hsinchu-300, Taiwan, Tel- 886-3-6889595, Fax: 886-3-6110850, E-mail: apple531.joyce@gmail.com

Copyright: © 2023 Chen HC. This is an open-access article distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited.

Received: 15 April, 2023, Manuscript No. jch-22-82233; **Editor Assigned:** 17 April, 2023, PreQC No. P-82233; **Reviewed:** 28 April, 2023, QC No. Q-82233; **Revised:** 03 May, 2023, Manuscript No. R-82233; **Published:** 10 May, 2023, DOI: 10.37421/2157-7099.2022.13.688

appeared as small round to oval cells with bland oval nuclei. The malignant population consisted of large cells with large pleomorphic nuclei having coarse chromatin and high nucleo-cytoplasmic ratio. Malignant change occurring in a background of pleomorphic adenoma was suggested. Based on the above-mentioned cytological features, malignant transformation from previous right parotid tumor was diagnosed (Figure 1).

Sequential study revealed multiple distant metastases to rib, T-spine, mediastinal lymph node and subsequently pleural effusion was sent for cytological study and cell block was made at the same time for further investigation.

The cytology report showed some clusters of plump hyperchromatic oval neoplastic cells with vesicular nuclei and pinkish cytoplasm (Figures 2 and 3). The tumor cells showed positive staining for CK7, lysozyme and occasionally positive for CEA (Figure 4) while negative for TTF-1, S100 and actin. The features suggested metastatic CXPA, compatible with salivary duct origin.

He received salvage radiotherapy with partially shrinkage of the tumor. General condition became worse after radiotherapy and he passed away after completion of radiotherapy due to pneumonia.

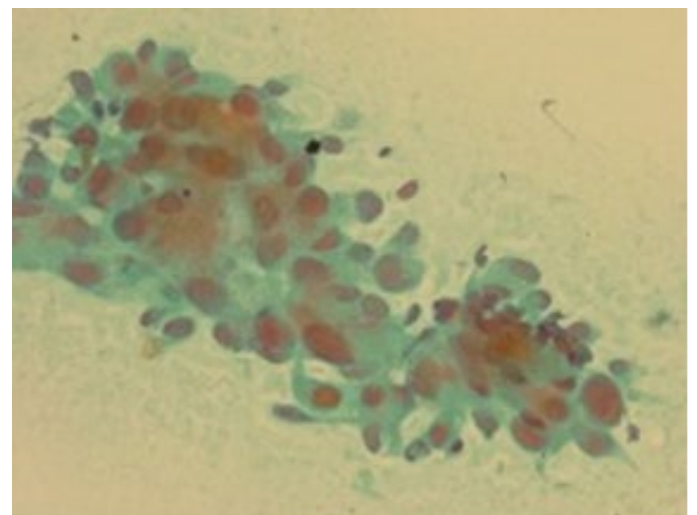


Figure 1. FNA smear showing malignant epithelial cells with pleomorphic nuclei, coarse chromatin and increased N/C ratio (Papanicolaou's stain, original magnification x20).

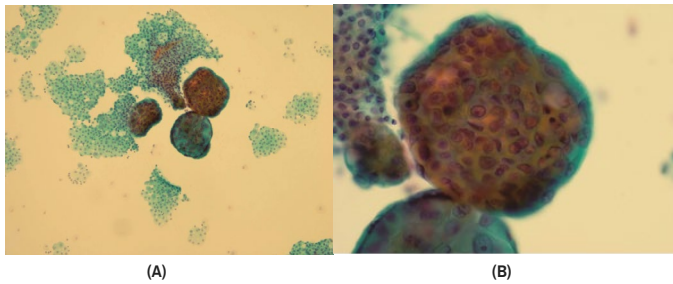


Figure 2. Pleural effusion cytology showing ball-like clusters of neoplastic cells (Papanicolaou's stain, original magnification x10 in A, x40 in B).

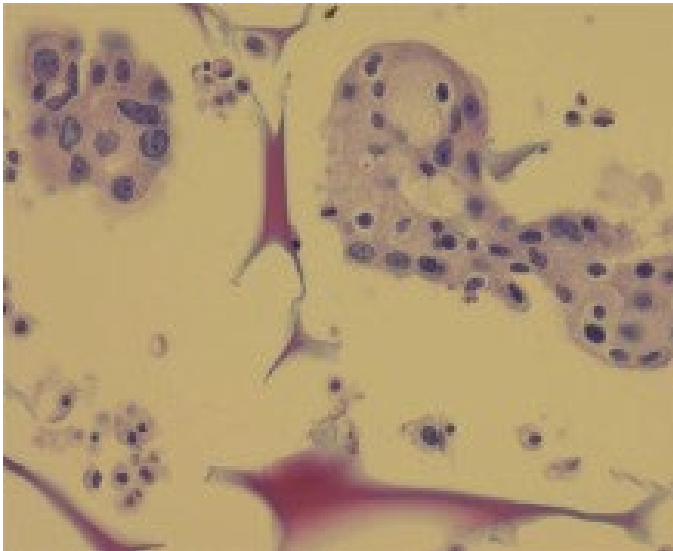


Figure 3. Cell block preparation illustrating clusters of plump hyperchromatic neoplastic cells occasionally with lumens within the clusters (H&E, original magnification x20).

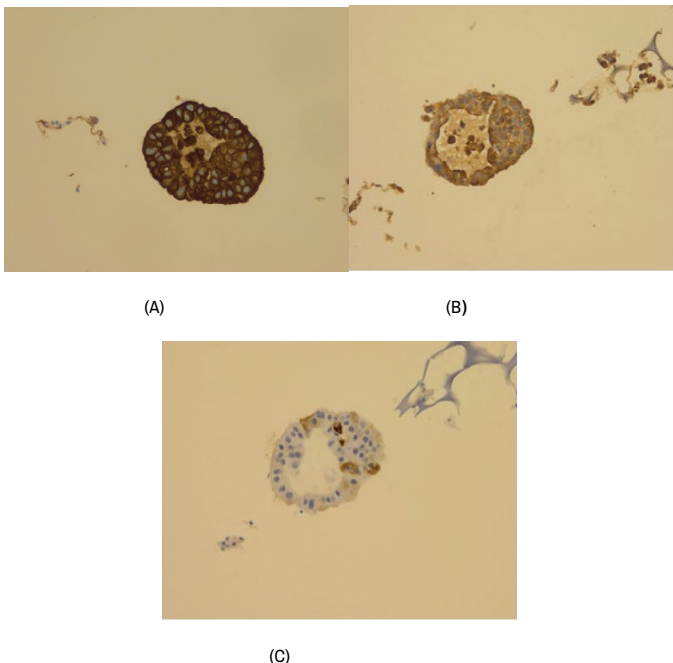


Figure 4. Immunohistochemical stain showing positive staining for CK7 (A) Lysozyme, (B) and (C) CEA (original magnification x20).

Discussion

We thought that our case was a carcinomatous transformation in pleomorphic adenoma involved by pleural fluid. Our case had all the features of malignant transformation along with local and distant metastasis. Multiple

distant metastases have been reported, including lung, breast, gingiva, vagina and inguinal lymph node [9-14]. Immunoprofiles were believed to be of potential assistance in the diagnosis of salivary gland tumors and in the prediction of histogenesis. The typical clinical presentation in a patient with CXPA is a longstanding history of pleomorphic adenoma and a sudden period of rapid growth. CXPA usually is a more poorly circumscribed mass than benign pleomorphic adenoma; is most likely to occur within the major salivary gland, especially the parotid gland; and typically occurs in patients in the sixth to eight decades of life [4,5].

CXPA has been named as carcinoma ex mixed tumor, carcinoma ex adenoma and carcinoma ex benign pleomorphic adenoma [4,5].

The differential diagnosis of a slow growing parotid mass that has recently exhibited a growth spurt should raise the suspicion of a CXPA. A CXPA can be mistaken for a benign PA. It can also be misdiagnosed as other benign and malignant salivary gland tumors. Cytologic differential diagnosis included mucoepidermoid carcinoma, a high-grade salivary gland adenocarcinoma and metastatic adenocarcinoma which are difficult to classify should include CXPA in its differential [15,16].

Conclusion

The diagnosis of metastatic CXPA in pleural effusion is possible on cytology if presence of unequivocal malignancy in association with features of PA. Correlation with clinical history and careful scrutiny interpretation of the cytomorphologic features help arriving at a conclusive diagnosis. This critical information is necessary for the clinician to plan out the therapeutic modalities for a better outcome.

Conflict of Interest

None.

References

1. Barnes, Leon, John Eveson, Peter Reichart and David Sidransky, et al. "World Health Organization classification of tumours: pathology and genetics of head and neck tumours." (2005).
2. Delgado, Ruby, Frank Vuitch and Jorge Albores-Saavedra. "Salivary duct carcinoma." *Cancer* 72 (1993): 1503-1512.
3. Lewis, Jean E., Barbara C. McKinney, Louis H. Weiland and Jorge A. Ferreiro, et al. "Salivary duct carcinoma: Clinicopathologic and immunohistochemical review of 26 cases." *Cancer: Interdisciplinary International Journal of the American Cancer Society* 77 (1996): 223-230.
4. Antony, Joyce, Vinod Gopalan, Robert A. Smith and Alfred KY Lam, et al. "Carcinoma ex pleomorphic adenoma: A comprehensive review of clinical, pathological and molecular data." *Head Neck Pathol* 6 (2012): 1-9.
5. Gupta, Avigeeet, Sina Koochakzadeh, David M. Neskey and Shaun A. Nguyen, et al. "Carcinoma ex pleomorphic adenoma: A review of incidence, demographics, risk factors, and survival." *Am J Otolaryngol* 40 (2019): 102279.
6. Buyukkurt, Selim, Aytakin Altintas, Derya Gumurdulu and Handan Zeren, et al. "Mucoepidermoid carcinoma of the parotid gland with ovarian and peritoneal metastasis." *J Obstet Gynaecol Res* 34 (2008): 271-273.
7. Torre, W., M. Comellas and M. Cuesta. "Massive pleural effusion as isolated manifestation of metastatic spread of salivary adenoid cystic carcinoma." *Respir Med* 91 (1997): 169-170.
8. Florentine, Barbara D., Tania Fink, Sali Avidan and Diana Braslavsky, et al. "Extra-salivary gland presentations of adenoid cystic carcinoma: A report of three cases." *Diagn Cytopathol* 34 (2006): 491-494.
9. Monaco, Sara E., Walid E. Khalbuss, Elena Ustinova and Alicia Liang, et al. "The cytomorphologic spectrum of salivary gland type tumors in the lung and mediastinum: A report of 16 patients." *Diagn Cytopathol* 40 (2012): 1062-1070.

10. Zhang, Yaxia, Carmen R. Gomez-Fernandez, Merce Jorda and Parvin Ganjei-Azar et al. "Fine-needle aspiration (FNA) and pleural fluid cytology diagnosis of benign metastasizing pleomorphic adenoma of the parotid gland in the lung: A case report and review of literature." *Diagn Cytopathol* 37 (2009): 828-831.
11. Hayes, Malcolm M., David Lesack, Christophe Girardet and Marina Del Vecchio, et al. "Carcinoma ex-pleomorphic adenoma of the breast. Report of three cases suggesting a relationship to metaplastic carcinoma of matrix-producing type." *Virchows Arch* 446 (2005): 142-149.
12. Bernabé, Daniel G., Luiz A. Veronese, Glauco I. Miyahara and Sebastião Conrado-Neto, et al. "Gingival metastasis from salivary duct carcinoma of the parotid gland." *J Periodontol* 79 (2008): 748-752.
13. Gabril, M. Y. and G. M. Yousef. "Salivary duct carcinoma presenting with vaginal metastasis: Case report." *European J Gynecol Oncol* 28 (2007): 415-417.
14. Bhalla, Ritu, Douglas C. Parker and Talaat S. Tadros. "Salivary duct carcinoma metastatic to inguinal lymph node: A case report of salivary duct carcinoma with distant metastasis diagnosed by fine-needle aspiration." *Diagn Cytopathol* 34 (2006): 41-44.
15. Pitman MB. "Mucoepidermoid carcinoma ex pleomorphic adenoma of the parotid." *Acta Cytol* 39 (1995): 604-606.
16. Nigam, Sonu, Neeta Kumar and Shyama Jain. "Cytomorphologic spectrum of carcinoma ex pleomorphic adenoma." *Acta cytologica* 48 (2004): 309-314.

How to cite this article: Chen, Hsiao-Ching. "Metastatic Carcinoma ex Pleomorphic Adenoma in Pleural Effusion." *J Cytol Histol* 13 (2023): 688.