

Fine Needle Aspiration Cytologic Diagnosis of Isolated Hepatic Metastasis of Adenoid Cystic Carcinoma of the Body of Tongue: A Case Report

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Citation

N Siddaraju, Q Chahwala, P Jothilingam, B Badhe, S Kumar. *Fine Needle Aspiration Cytologic Diagnosis of Isolated Hepatic Metastasis of Adenoid Cystic Carcinoma of the Body of Tongue: A Case Report*. The Internet Journal of Otorhinolaryngology. 2008 Volume 8 Number 2.

Abstract

Isolated liver metastasis from adenoid cystic carcinoma (ACC) is uncommon with only a few cases being documented in the literature. Here, we report a case of ACC presenting with an isolated hepatic metastasis, primarily diagnosed by fine needle aspiration biopsy (FNAB).

A 50-year-old male presented with a firm, nodular, non-tender hepatomegaly. An ultrasound (US) guided FNAB from the hepatic nodule revealed cytologic features consistent with adenoid cystic carcinoma (ACC), with a prominent cribriform pattern. On further history, it was found that the patient was on follow-up with radiotherapy for histologically proven ACC of the body of the tongue.

Our case emphasizes the use of simple FNA technique in an early and unambiguous diagnosis of isolated hepatic metastasis of ACC. The body of the tongue as a primary site of ACC is unusual.

INTRODUCTION

Adenoid cystic carcinoma (ACC) is the most common malignant tumor of the submandibular, sublingual and minor salivary glands.^{1,2} The other primary sites of ACC include the parotid gland,^{1,2} mucous glands of the upper respiratory tract, lacrimal glands, lung, prostate and the breast.³ In the oral cavity, palate is the most common site. It has also been reported within the body of the tongue, a very rare presentation for a salivary gland neoplasm.^{4,5,6} The age group reported for these tumors ranges between 20 and 84 years, with a median of 52 years;⁷ ACC occurring in the oral cavity has a slight male predilection.⁸ Clinically these tumors are characterized by slow growth, multiple recurrences, long clinical course, early perineurial spread and late metastasis.^{7,9} Histologically, the tumor can be classified into tubular, cribriform and solid types,^{8,10} of which the cribriform pattern is the most common and the solid pattern, the least.⁸ ACC with solid pattern is associated with a more aggressive clinical behavior.^{8,9,10} The commonest site of metastasis of ACC is the lung followed by the brain and bone. Rare sites such as stomach, toe, choroids, skin,¹ and kidney^{3,9} have also been reported.

Metastasis to liver is rare; when it occurs, it is generally seen as a part of the wide spread metastasis. An isolated hepatic metastasis from ACC is highly unusual.¹

Here, we report a case of ACC of the tongue presenting with an isolated hepatic metastasis, diagnosed primarily by FNAB technique.

CASE

Clinical Summary: A 50-year-old male presented in the outpatient department of Surgery with a history of heaviness and pain in the right hypochondrium and loss of appetite of 2 months' duration. Except for a firm, non-tender hepatomegaly, there were no other significant clinical findings. Laboratory investigation including the blood tests, chest radiography and liver function tests were unremarkable. However, an ultrasonography (US) of the abdomen revealed heterogeneous hypoechoic mass involving the right lobe of the liver.

Material and methods: An ultrasound (US) guided FNA from the hepatic nodule was performed using a 23 gauge needle attached to a 10 mL syringe. The material obtained was blood mixed, particulate. Both air dried and wet fixed

smears were prepared and stained with Papanicolaou and May-Grünwald Giemsa (MGG) techniques respectively.

Results (cytologic findings): Smears were cellular, with MGG stained smears showing a striking presence of homogenous, brightly magenta colored ground substance in the form of hyaline globules and finger-like stroma, along with rare sheets and a few discrete, as well as tiny groups of hepatocytes, some of which revealed granules of greenish bile pigment (figure1). Cellular morphology was well appreciated in Papanicolaou smears in which they were seen as monomorphic cells exhibiting high nuclear to cytoplasmic (N: C) ratio, with scanty cytoplasm and small rounded nuclei having coarsely granular chromatin and inconspicuous nucleoli. These cells were seen chiefly, encircling and adhering to pale, translucent, cyanophilic to eosinophilic hyaline globules. Also seen in the background were many dispersed, neoplastic naked nuclei, along with hepatocytes (figure2). Based on these cytomorphologic features, a diagnosis of hepatic metastasis from adenoid cystic carcinoma (ACC) was offered. On further history, it was found that the patient was on follow-up with radiotherapy for carcinoma of the tongue. Three years previously the patient had presented with growth in the anterior 2/3rd of the tongue which was histologically proven to be an ACC with a prominent cribriform pattern. Further examination and investigation revealed no other lesion elsewhere in the body, favoring an isolated hepatic metastasis.

Figure 1

Figure 1: Smear showing hyaline globules of varying size in association with bile pigment laden hepatocytes in a hemorrhagic background (MGG stain, X400)

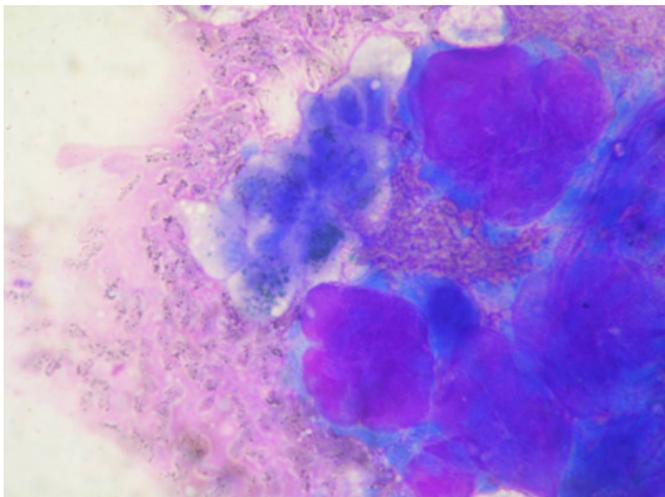
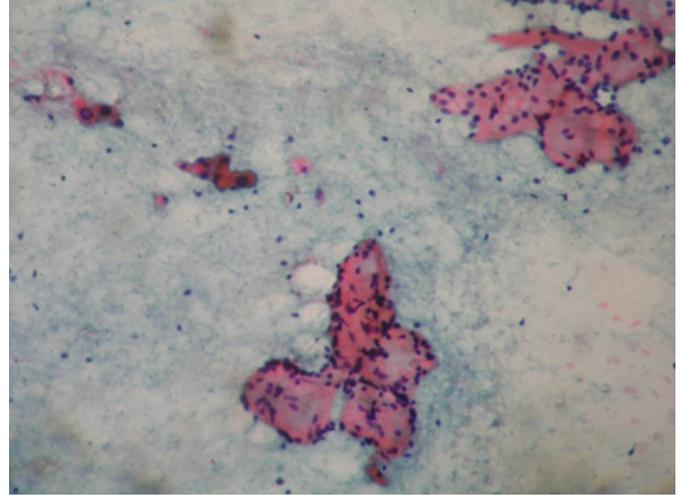


Figure 2

Figure 2: Smear showing pale, eosinophilic, translucent hyaline globules encircled by monomorphic neoplastic cells. A few naked nuclei and two tiny groups of hepatocytes are also seen in the background (Papanicolaou stain X200)



DISCUSSION

Histologically, the cribriform pattern of ACC is characterized by sieve like appearance with islands of tumor cells containing many, small, round pseudocystic structures of varied size. A tubular pattern of ACC exhibits a less complex cribriform appearance. On cytology, the pseudocystic appearance of ACC is reflected as hyaline spheres (globules) and finger-like structures (cylinders) surrounded by monomorphic basal cells exhibiting scanty rim of cytoplasm, high nucleus to cytoplasmic (N:C) ratio and nuclear moulding.^{8,11} The single cells seen in the background are generally seen as discrete naked nuclei.⁸ All these features were highly apparent in our case; this prompted an easy diagnosis.

An ACC with a solid pattern is said to predict a more aggressive behavior with increased distant metastasis and decreased survival;⁹ histologically, it is characterized by solid nests of tumor cells with absent or very few cyst-like spaces, and more frequent mitoses. Cytology of solid pattern is less often documented in the literature;⁸ it is diagnostically more challenging due to its cytologic resemblance to tumors such as basal cell adenoma⁸ and small cell neuroendocrine carcinoma.¹¹ Despite the obvious absence of solid pattern, our case presented with distant metastasis. In one of the recent studies by Sung et al, the rates of distant metastasis reported for tubular, cribriform and solid variants of ACC were 42%, 48% and 64% respectively.⁹

Cytologically, ACC occurring at primary locations needs to be distinguished from other benign and malignant neoplasms manifesting with hyaline globules.^{8,11} As the liver is not a known primary site for ACC, diagnosing hepatic metastasis from an ACC is easy, especially, when the cytomorphology reflects a classic cribriform pattern, as evident in the present case. The other significant finding in our case was that the morphologic features of both the primary and metastatic lesions were identical; this was evident by the review of histopathologic slides of the primary tongue lesion diagnosed, 3 years prior to the development of metastasis.

Cases identical to ours have been rarely reported by other authors. Qureshi et al² as well as, Harish and Gouri¹ documented cases of hepatic metastasis from ACC of the parotid gland, while Zeiden and colleagues¹² reported it from the lacrimal gland. Similar to our case, the histologic pattern of the primary lesion and hepatic metastasis in their cases were identical. In these cases, the time interval between the surgical treatment of primary lesion and the development of hepatic metastasis varied from 3 to 20 years.^{1,2,12} In the present case, hepatic metastasis developed 3 years after the surgery was performed for primary in the tongue. Harish and Gowri¹ stressed on the use of FNAB in the diagnosis of hepatic metastasis of ACC. Srivastava et al³ reported a case of metastatic ACC in kidney, diagnosed by FNAB.

The distant metastasis appears to progress very slowly, remaining stable for a long period. Thus, resection of solitary and even multiple nodules is said to be justifiable. Because of its unpredictable behavior, many authors have stressed on the need of a long term follow up. If liver metastasis is solitary and the disease free interval is long, surgical resection is recommended in an effort to improve survival.² The solitary hepatic metastatic focus in the case reported by Qureshi and colleagues² was successfully resected.

To conclude, owing to its characteristic cytomorphologic expression, the cytodiagnosis of metastasis from an ACC with a prominent cribriform pattern is quite straight forward, especially in a site such as liver, where primary ACC is unheard of. Our case emphasizes the use of simple FNA technique in an early and precise diagnosis of isolated hepatic metastasis of ACC. Primary site in the present case

was the body of the tongue which is an extremely uncommon site for ACC.

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References

1. Harish K, Gouri SRM: Adenoid cystic carcinoma of the parotid metastasizing to liver: case report. *BMC cancer*; 2004, 4:41 doi:10.1186/1471-2407-4-41
2. Qureshi SS, Nadkarni MS, Shrikhande SV, Desai S, Deodhar K, Ramadwar M, Shukla PJ: Hepatic resection for metastasis from adenoid cystic carcinoma of parotid gland. *Indian J Gastroenterol*.2005;24:29-30
3. Srivastava S, Jaiswal R, Agarwal A, Singh PK, Singh SN: Cytological diagnosis of adenoid cystic carcinoma of the parotid metastatic to kidney and lung. 2007; 24:201-202
4. Greer Jr R, Said S: Adenoid cystic carcinoma. In: *Silverberg's Principles and Practice of Surgical Pathology and Cytopathology*. Fourth edition. Edited by Silverberg SG, DeLellis RA, Frable WJ, LiVolsi VA and Wick MR. Elsevier. China 2006, pp1189-1190
5. Pino Rivero V, Pentoja Hernandez CG, Gonzalez Polamino A, Pardo Rpmero G, Trinidad Ramos G, Blasco Huelva A: Adenoid cystic carcinoma of tongue. Report of a case and literature review. *An Otorrhinolaryngol Ibero Am*. 2006;33:449-54
6. Carrasco Ortiz D, Aldape Barrios B: Adenoid cystic carcinoma of the base of the tongue. Presentation of a case. *Med Oral Patol Oral Cir Bucal*.2006;115:E417-420
7. Spiro RH, Huvas AG, Strong EW: Adenoid cystic carcinoma of salivary gland origin. A clinicopathologic study of 242 cases. *Am J Surg* 1974;128:512-529
8. Auclair PL, Ellis GL, Stanley MW: Adenoid cystic carcinoma. In: *Silverberg's Principles and Practice of Surgical Pathology and Cytopathology*. Fourth edition. Edited by Silverberg SG, DeLellis RA, Frable WJ, LiVolsi VA and Wick MR. Elsevier. China 2006, pp1236-1240
9. Sung MW, Kim KH, Kim JW, Min YG, Seong WJ, Roh JL, Lee SJ, Kwon TK, Park AW: Clinicopathologic predictors and impact of distant metastasis from adenoid cystic carcinoma of the head and neck. *Arch Otolaryngol Head Neck Surg*.2003;129:1193-1197
10. Rosai J: Major and minor salivary glands. In: *Rosai and Ackerman's Surgical Pathology*, vol. 1, Ninth edition, Edited by Rosai J. Mosby, an imprint of Elsevier. St Louis.2005; pp 873-916
11. Orell SR, Sterret GF, Walters MN-I, Whitaker D: Head and neck; salivary glands. In: *Manual and Atlas of Fine Needle Aspiration Cytology*. Third edition. Edited by Orell SR, Sterret GF, Walters MN-I, Whitaker D. Churchill Livingstone, Edinburgh, London.1999-pp 40-72
12. Zeidan BA, Hilal MA, Al-Gholmy M, El-Mahallawi H, Pearce NW, Primrose JN: Adenoid cystic carcinoma of the lacrimal gland metastasizing to the liver: report of a case. *World Journal of Surgical Oncology*.2006;4: 66 doi:10.1186/1477-7819-4-66

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