대한세포병리학회지 제 15권 제 1호
The Korean Journal of Cytopathology
15 (1) : 56-59, 2004

아가미 암종의 세침흡인 세포학적 소견
- 1예 보고-

가톨릭대학교 임상병리과

맹 이 소 · 김 경 미 · 이 안 희 · 강 창 석

= **Abstract** =

Fine Needle Aspiration Cytology of the Branchiogenic Carcinoma
-Report of A Case-

Leeso Maeng, M.D., Kyoung-Mee Kim, M.D., Anhi Lee, M.D., and Chang Suk Kang, M.D.

Department of Clinical Pathology, The Catholic University of Korea,
College of Medicine, Seoul, Korea

Development of an invasive squamous cell carcinoma within a branchial cleft cyst (branchiogenic carcinoma) is very rare and this case is the first description of cytologic findings in Korea. A 62-year-old man presented with a 5-month history of an enlarged right neck mass. Fine needle aspiration cytology of the mass showed low cellularity and the background containing proteinaceous debris, inflammatory cells, and histiocytes suggested a cystic nature. Abundant lymphocytes, a few anucleated squames, and rare atypical squamous cells with pyknotic nuclei and abnormal keratinization were also noted. After excision of the mass, histologic findings were consistent with a branchiogenic carcinoma. We recommend to raise the possibility of carcinoma (primary or metastatic) when we observe a few dyskeratotic cells in a cystic mass within the neck area, especially in an old male.

Key words: Branchiogenic carcinoma, Fine needle aspiration cytology, Neck, Branchial cleft cyst

책임저자 : 김 경 미
주 소 : (403-026) 인천 부평구 부평동 665
전 화 : 032-510-5538
팩 스 : 032-510-5881
E-mail address : kkmkys@catholic.ac.kr

INTRODUCTION

The existence of primary carcinomas arising from branchial cleft cysts has been controversial since 1882 when von Volkmann[1] introduced the concept of "branchiogenic carcinoma". In 1950, Martin et al.,[2] established stringent criteria for establishing the diagnosis of a primary branchial cleft carcinoma. A few reports of branchiogenic carcinoma have been described in the English literature.[3-6] We present an additional case of this very rare disease with cytologic findings.

CASE

A 62-year-old man presented with a progressively enlarging right neck mass for 5 months. He had a similar swelling, when he was a child, which had spontaneously resolved. On physical examination, a 3.0cm sized, nontender, firm, movable mass was found on the anterior border of the sternocleidomastoid (SCM) muscle. There was no other lymphadenopathy. The oropharyngeal examinations were normal and the fiberoptic nasolaryngoscopy revealed no hypopharyngeal or laryngeal lesions. A computed tomography (CT) scan of the neck revealed a 3.0x3.5x2 cm sized, round, rim-enhancing cystic lesion in the left submandibular region, between the submandibular gland and SCM muscle (Fig. 1).

Fig. 1. Contrast-enhanced CT of the neck. It shows an about 3 x 3.5 cm sized, round, rim-enhancing cystic lesion in the left submandibular region, between the submandibular gland and the sternocleidomastoid muscle. It has an internal thick septum-like soft tissue density with a small nodular thickening in the enhanced wall.

Cytologic findings

Fine needle aspiration of the mass showed low cellularity and the background containing proteinaceous debris, inflammatory cells, and histiocytes suggested cystic contents (Fig. 2). In addition, there were abundant lymphocytes, a few anucleated squames and a few atypical squamous cells with pyknotic nuclei and abnormal keratinization (Fig. 2 inset). Differential diagnoses included branchial cleft cyst, epidermal inclusion cyst, and cystic squamous cell carcinoma.

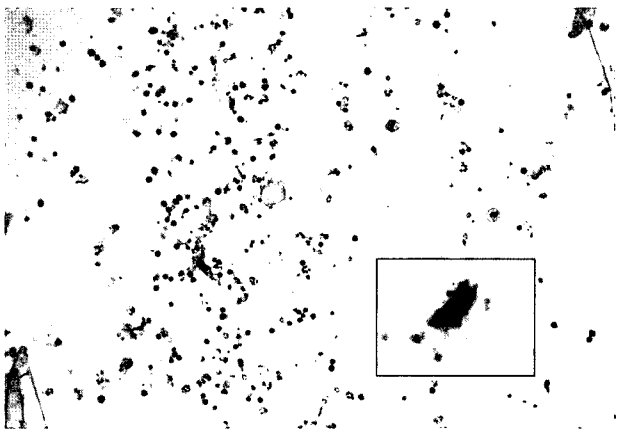

Fig. 2. Fine needle aspiration cytologic findings. It shows low cellularity with abundant lymphocytes and a few anucleated squames in proteinacious background, consistent with cystic content (Papanicolau). Inset. There is an atypical squamous cell with pyknotic nuclei and abnormal keratinization (Papanicolau).

Histologic findings

Direct laryngoscopy, bronchoscopy, and esophagoscopy with guided biopsies from multiple sites in the upper aerodigestive tract were followed by an excision of the cervical lesion. The mass appeared cystic but had a mural

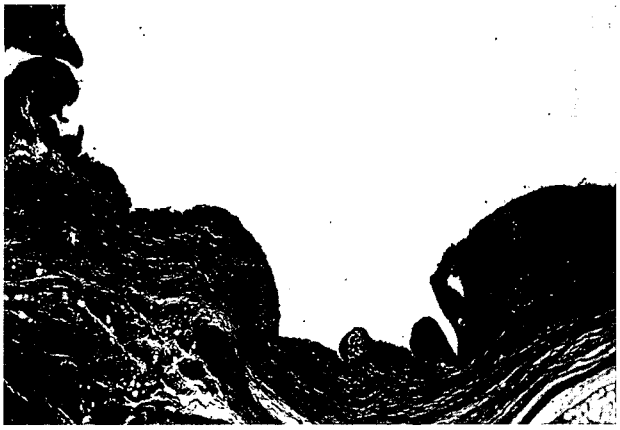

Fig. 3. Histologic findings. The cystic wall is lined by stratified squamous epithelium with gradual transition from dysplasia to invasive squamous cell carcinoma.

nodule on the fibrotic wall. The surrounding lymphoid elements did not show peripheral lobulation, internodular trabeculae, or perinodal sinuses. Small adjacent lymph nodes were also removed and showed no tumor component. The final histopathologic examination of the mass revealed a cystic structure lined by stratified squamous epithelium with gradual transition from dysplasia to invasive squamous cell carcinoma (Fig. 3). The patient underwent a full oncologic workup including a CT scan of the chest and abdomen. A primary source of carcinoma was not found. He refused the additional complete neck dissection and radiotherapy. He has remained disease-free for 11 months.

DISCUSSION

In differential diagnosis, we considered branchial cleft cyst and malignant cystic squamous lesion including branchiogenic carcinoma and metastatic squamous cell carcinoma of unknown origin. The malignant squamous lesion could be recognized by the following cytologic features: increased nuclear/cytoplasmic ratio, irregularity of nuclear outline and nuclear hyperchromatism. The branchial cleft cysts showed essentially benign squamous cells with only mild nuclear atypia. The background of the malignant aspirates showed more necrotic debris but fewer polymorphonuclear cells than that of the branchial cleft cysts. In our case, only a few dyskeratotic cells are the clue of malignant squamous lesion. Branchiogenic carcinoma is difficult to prove and to differentiate from cystic metastasis of squamous cancer from Waldeyer' ring epithelium.[5,6] Furthermore, some cases show only a few deceptively bland squamous cells and squame, simulating those in a branchial cleft cyst.[7] True branchiogenic carcinoma is exceedingly rare.[3-5] Khafif et al.[4] reviewed the English literature and found 26 cases of carcinoma in a lateral neck cyst with histologic evidences of branchiogenic carcinoma and no identifiable primary. But only 6 cases satisfied the third criterion of Martin et al.[2] in which patients must have survived and have been followed up by periodic examinations for at least 5 years without the development of any other lesions that could be the primary tumor. Khafif et al[4] proposed two other critieria: the absence of an identifiable primary cancer by a thorough evaluation, including endoscopy, CT scan of thehead and neck, and appropriate biopsies; and histologic identification of a cystic structure partially lined by normal squamous or pseudostratified columnar epithelium with gradual transition to invasive squamous cell carcinoma. In the present case, the patient has only 11 months of disease-free period, but satisfied the proposal by Khafif et al.[4] Especially, the histologic finding of dysplastic epithelium next to frankly invasive carcinoma in our patient strongly supports the origin of the carcinoma from a branchial cyst. With our experience, it is noteworthy to remind Troxel' suggestion.[8] First, don't make a diagnosis of branchial cyst without first considering the possibility of metastatic cystic squamous cell carcinoma, regardless of patient age and clinical diagnosis. Second, look carefully for cytologic atypia in the lining squamous epithelium. If present, alert the clinician to the need for thorough work-up to exclude an occult primary in the head or neck. Third, hesitate to make a FNA diagnosis of "consistent with branchial cyst" based on a few bland squamous cells in sparsely cellular aspirate or to make a diagnosis of "nondiagnostic due to

sparse cellularity", which leads to biopsy.

In the neck area, especially in the male patient, when we observe a few dyskeratotic cells in the cystic background, it is better to diagnose "nondiagnostic" or raise the possibility of carcinoma, either primary or metastatic.

REFERENCES

1. Von Volkmann R. Das tiefe branchiogege halskarcinom. *Zentralbl Chir.* 1882;9:49-63.
2. Martin H, Morfit HM, Ehrlich HE. Case for branchiogenic cancer (malignant branchioma). *Ann Surg* 1950;132:867-87.
3. Singh B, Balwally AN, Sundaram K, Har-el G, Krgin B. Branchial cleft cyst carcinoma: Myth of Reality. *Ann Otol Rhinol Laryngol* 1998;107:519-24.
4. Khafif RA, Prchep R, Minkowitz S. Primary branchiogenic carcinoma. *Head Neck* 1989;11:153-63.
5. Micheau C, Klijanienko J, Luboinski B, Richard J. So-called branchiogenic carcinoma actually cystic metastases in the neck from a tonsillar primary. *Laryngoscope* 1990;878-83.
6. Verma K, Mandal S, Kapilar K. Cystic change in lymph nodes with metastatic squamous cell carcinoma. *Acta Cytol* 1995; 39:478-80.
7. Frierson JR HF. Cysts of the head and neck sampled by fine-needle aspiration: sources of diagnostic difficulty. Editorial. *Am J Clin Pathol* 1996:559-60.
8. Troxel DB. Diagnostic errors in surgical pathology uncovered by a review of malpractice claims, PART VI Urinary bladder and branchial cleft cyst. *Int J Surg Pathol* 2002;9:227-9.