Cases Report

Disappearance of Primitive Basaloid Cells after External Irradiation in Adenoid Cystic Carcinoma of the Lacrimal Gland

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Objective: *To study the histopathological findings of basaloid adenoid cystic carcinoma after high dose orbital irradiation.*
Material and Method: *The histopathology study was done in a 38-year-old Thai female patient who had basaloid adenoid cystic carcinoma of the right lacrimal gland confirmed by incisional biopsy. After a 60-Gy external irradiation, the mass decreased in size and finally could not be palpated, but subsequent orbital imaging still showed infiltrative soft tissue mass at the right lacrimal gland with bony defects at posterosuperior and lateral walls without extraorbital extension. Orbital exenteration and lateral orbitectomy was performed. The specimen was submitted for histopathological examination.*
Results: *Microscopic examination revealed periglandular infiltration with chronic inflammation of the palpebral lobe of lacrimal gland. The orbital lobe was extensively replaced with fibrous tissue. There was only a small area of cribiform pattern of the adenoid cystic carcinoma left; no basaloid pattern was seen in the tissue.*
Conclusion: *The primitive cells (basaloid pattern) disappear after orbital irradiation for adenoid cystic carcinoma of the lacrimal gland. Replacement with fibrous tissue is seen.*

Keywords: *Adenoid cystic carcinoma, Lacrimal gland, Basaloid pattern, External beam radiation*

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Adenoid cystic carcinoma is the most common malignant epithelial tumor of the lacrimal gland[1].The treatment of choice for this devastating tumor is orbital exenteration alone or combined with other modalities. Isolated radiation therapy is not efficacious in treating the malignancy[1].

Histopathologic patterns have been described into five categories: cribiform, basaloid, tubular, sclerosing and comedocarcinomatous[1-3].The basaloid pattern contains the most primitive cell type and has the gravest prognosis[2-4] since it has been found to reduce estimated disease-free survival when the tumor is discovered half or more of the biopsy specimen[4]. Histopathological change of the pattern after high dose irradiation has never been described. The authors herein report a case of basaloid adenoid cystic carcinoma in which the primitive cells disappear after the external beam high dose irradiation.

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Case Report

A 38 year-old female presented with pain and proptosis on the right eye for three months (Fig. 1). Visual acuity was normal in both eyes. A firm mass without tenderness, size 1.5x1 cm, could be palpated at the superolateral area of the orbit. No lymphadenopathy was detected. Computerized tomogram of the orbit showed an infiltrative mass involving lacrimal

Fig. 1 The patient at presentation with her right proptotic eye

gland with bony erosion of the lacrimal gland fossa. The differential diagnosis included adenoid cystic carcinoma, malignant mixed tumor, lymphoma and other infiltrative diseases. The provisional diagnosis was established by an incisional biopsy. The histopathological findings revealed adenoid cystic carcinoma with basaloid pattern in majority (Fig. 2a, 2b). The patient denied surgical treatment and the decision of treatment went to the external beam irradiation. After a high dose radiation of 60 Gy within a six-week period, the tumor decreased in size and finally could not be palpated. Unfortunately, the infiltrative mass was still detectable by subsequent orbital computerized tomogram. The patient eventually underwent orbital exenteration and lateral orbitectomy. Histopathology by serial sections showed extensive areas of fibrosis, with only a small area of cribiform pattern left as tumor remnant in the orbital lobe of the lacrimal gland. There was no detectable basaloid pattern in the tissue (Fig. 3a, 3b). There was periglandular infiltration with chronic inflammation of the palpebral lobe. The surgical margin was free of tumor.

Discussion

In addition to the orbital exenteration as the treatment of choice for adenoid cystic carcinoma of the lacrimal gland, there are other options such as intra-arterial chemotherapy proposed by Meldrum et al as an adjunctive therapy to the surgical treatment[5]. Proton beam irradiation promises benefit in long-term follow up period[6]. As far as the authors are aware, there are no previous reports describing histopathological change after isolated high dose irradiation. In the presented case, the basaloid pattern totally disappeared and only a small area of cribiform pattern could be found in the tissue after the radiation therapy. The gland was replaced by fibrous tissue. These findings raise questions whether basaloid pattern is more sensitive to radiation than cribiform pattern and whether a high dose of irradiation can eradicate this pattern of the tumor.

In conclusion, disappearance of the basaloid pattern of the adenoid cystic carcinoma of the lacrimal gland could be seen after a high dose external beam

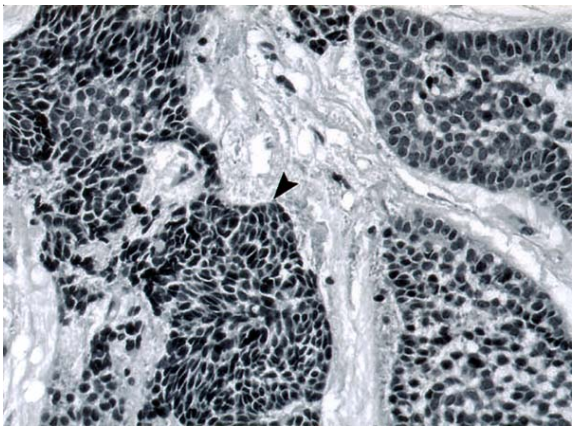

Fig. 2a Photomicrograph of the biopsy specimen shows well-defined lobules of basophilic cells (arrow) representing the basaloid pattern of the tumor. (Hematoxylin-eosin stain)

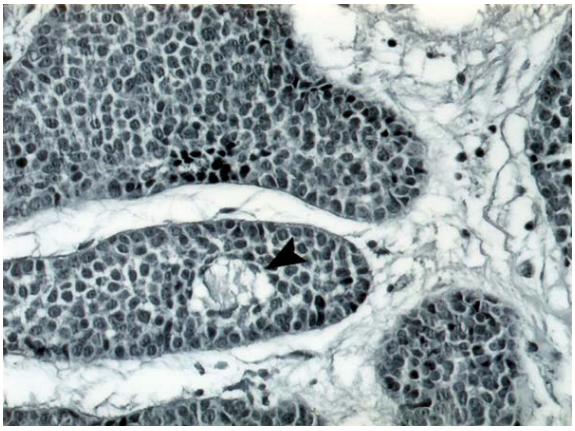

Fig. 2b Special staining of the biopsy specimen shows some areas of mucin production within the intercellular space (arrow), representing some areas of the tumor differentiation. (Mucicarmine stain)

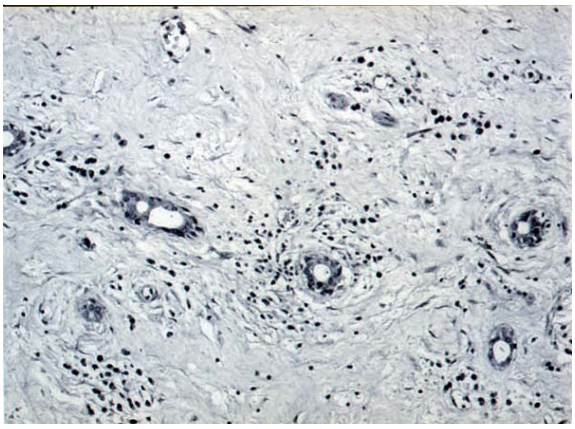

Fig. 3a Low power photomicrograph of the exenterated specimen shows extensive area of fibrosis with some chronic inflammation. Areas of cri-biform pattern are seen (Hematoxylin-eosin stain)

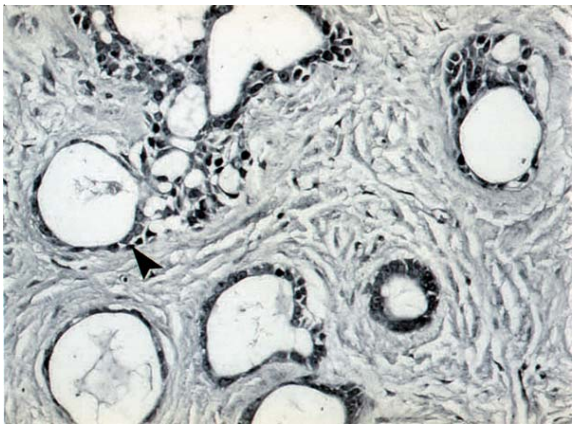

Fig. 3b High power photomicrograph of the exenterated specimen shows cribiform pattern of the adenoid cystic carcinoma (arrow) (Hematoxylin-eosin stain)

irradiation and the tumor areas were replaced by fibrous tissue.

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การสลายตัวไปของ primitive basaloid cell หลังการให้รังสีรักษาในมะเร็งต่อมน้ำตาชนิด adenoid cystic

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วัตถุประสงค์: เพื่อศึกษาลักษณะทางพยาธิวิทยาของเนื้องอก basaloid adenoid cystic carcinoma ภายหลังการใช้รังสีรักษา

วัสดุและวิธีการ: ศึกษาในผู้ป่วยหญิงไทยอายุ 38 ปี ที่พบว่าเป็นมะเร็งต่อมน้ำตาชนิด basaloid adenoid cystic carcinoma ด้วยการตัดบางส่วนของชิ้นเนื้อไปตรวจ ภายหลังการใช้รังสีรักษาปริมาณสูง 60 Gy ก้อนเนื้อยุบลงจนคลำไม่พบ แต่จากภาพถ่ายทางรังสีพบยังมีก้อนอยู่บริเวณต่อมน้ำตาร่วมกับมีการทำลายของกระดูกเบ้าตาบริเวณด้านหลังส่วนบนและด้านข้าง จึงได้ทำการผ่าตัดควักลูกตาและเนื้อเยื่อในเบ้าตาออก ร่วมกับการตัดกระดูกเบ้าตาด้านข้าง แล้วส่งชิ้นเนื้อตรวจทางพยาธิวิทยา

ผลการศึกษา: การตรวจชิ้นเนื้อทางจุลภาคพบมีการอักเสบเรื้อรังที่บริเวณ palpebral lobe ของต่อมน้ำตาส่วนบริเวณ orbital lobe นั้นเต็มไปด้วยเนื้อเยื่อพังผืด มีเพียงบางบริเวณเท่านั้นที่มีมะเร็งต่อมน้ำตาแบบ cribiform pattern หลงเหลืออยู่ แต่ไม่พบว่ามีชนิด basaloid pattern เหลืออยู่เลย

สรุป: เซลล์มะเร็งส่วนที่ primitive หรือ basaloid pattern ในโรคมะเร็งต่อมน้ำตาชนิด adenoid cysticนั้นสลายตัวไปหลังจากการใช้รังสีรักษา โดยมีการแทนที่ด้วยเนื้อเยื่อพังผืด