A Case of Pleomorphic Adenoma Manifested as a Subcutaneous Nodule

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Among tumors covered with skin over pre-or infra-auricular areas of the face, pleomorphic adenoma arising from parotid gland is relatively common to occur, but many dermatologists can miss the correct diagnosis of it. Clinically, it is painless, slowly growing mass which may suggest to make diagnosis.

We experienced a case of pleomorphic adenoma occurring on the left cheek as a single mass in a 36-year-old man. Total surgical excision was done. Histopathological findings of tumor mass revealed a mixture of epithelial cells, myoepithelial cells, ductal and cartilagenous structure. PAS stain, alcian blue stain, carcinoembryonic antigen and vimentin and S-100 protein in the immunohistochemical staining showed positive results. (Ann Dermatol 4(2) 77–82, 1992)

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Pleomorphic adenoma grows slowly, sometimes appearing to remain unchanged for years, and it is not ordinarily a fatal disease. The possible development of malignancy is the chief danger which increases with the passing years. Tumors that are completely removed surgically do not recur and the patient is cured. With limited surgical removal, there is risk of local recurrence. Subsequent surgery becomes more difficult because the recurrence comes out to be often multiple and widespread within the area of the previous operation.

We report herein a case of pleomorphic adenoma which have not been reported in Korean dermatological literature under this name and should be considered in the categories of tentative diagnoses of benign tumor occurring on the face.

REPORT OF A CASE

A 36-year-old man visited our clinic for the evaluation of the skin tumor which was painless skin-colored, walnut-sized single mass on the anteroinferior part of the left auricle for about 26 years. The patient noticed first accidentally a painless bean-sized nodule on the left cheek, and it grew slowly every year. His past and family histories were non-contributory. Physical examination showed a 3×4cm, movable hard mass on the anteroinferior part of the left auricle (Fig. 1), and laboratory findings of CBC, LFT, chest P-A, urinalysis, and stool exam. were within normal limits.

Under the impression of an epidermal cyst, surgical intervention and histopathological studies were performed for the final diagnosis of the tumor mass. Grossly, the removed tumor mass consisted of 3 lobules surrounded by thin, delicate capsule and the color of cut surface was yellow or yellowish-white (Fig. 2, 3). Microscopically, it showed a characteristic mixture of epithelial cells, myoepithelial cells, ductal structure, and cartilaginous area (Fig. 8). Alcian blue stain and immunohistochemical staining with carcinoembryonic antigen (CEA), vimentin, and S-100 protein showed positive (Fig. 9). Electron microscopic findings revealed actin microfilaments, tonofilaments and myoepithelial cell (Fig. 10). The fea-
Fig. 1. Movable hard tumor mass on the left cheek.

Fig. 2. Tumor mass composed of 3 lobes was well encapsulated, yellowish, moderately hard in consistency.

Fig. 3. The cut surface of the mass showed solid, white or yellowish-white and semitranslucent cartilage-like area.

Fig. 4. The section consisted of epithelial and myoepithelial cells increased cellularity and myxoid stroma (H&E, ×100).

Fig. 5. It revealed a mixture of ductal structure, chondroid structure and epithelial cell infiltration (H&E, ×200).

Fig. 6. On the focus of chondroid formation, it showed hydropic clear cells scattered in the chondroid matrix (H&E, ×100).
Fig. 7. On higher power view, it showed epithelial cells, myoepithelial cells in the stroma (H&E, x400).

Fig. 8. On PAS staining with diastase treatment, it revealed PAS-positive and diastase-resistant.

Fig. 9. Alcian blue stain(a), and immunohistochemical staining with CEA(b), vimentin(c) and S-100 protein(d) showed positive results; CEA stained at ductal area, vimentin & S-100 protein stained at myoepithelial area.
Pleomorphic adenoma is a circumscribed tumor characterized microscopically by its pleomorphic or mixed appearance, and its clearly recognizable epithelial tissue intermingled with areas of mucoid, myxoid, or chondroid appearance.

It is painless, nonulcerated, slowly growing mass behind the angle of the jaw, below and in front of the ear which may suggest to make diagnosis. It is moderately firm and usually movable and smooth-surfaced at first, but it often becomes nodular and less mobile as it grows. The tumor is a common benign tumor of salivary glands, especially parotid gland and submaxillary gland. However, its development in the minor salivary glands of the oral cavity, pharynx, and lacrimal glands and in the skin, is well known.

It occurs about 10 times more common in the parotid gland than in the submandibular gland, and is rare in the sublingual gland. In the review of over 4,000 cases by Rauch et al. 8, 92.5% arose in the major salivary glands and 6.5% in the minor salivary glands. It occurs most frequently in women around the age of 40.

Distribution in cases of chondroid syringoma by Hirsch et al. 3 showed that chondroid syringoma involved everywhere but the region of the head and neck was the most common involved (80%). Histologically similar tumors occurred in the lacrimal glands and skin are so called mixed tumor of skin or chondroid syringoma. Thackray et al. 4 showed the varieties of pleomorphic adenomas in microscopic findings: duct area, myxoid and chondroid area, solid area resembling a myoma, cylindromatous area, neumatous area, area of squamous epithelium, calcification area, and area of deposits of crystalline materials. He said that myxoid and chondroid area is characteristic of pleomorphic adenoma and it is responsible for its alternative name of "mixed tumor". So we thought that chondroid syringoma is one of the categories of pleomorphic adenoma.

The neoplastic glands have a lining composed of two cell types: a luminal layer of cuboidal cells and a peripheral layer of flattened cells.

Two types of mucin are formed by pleomorphic adenomas, one is epithelial and the other is connective tissue type. The former is characterized by its high content of neutral glycoprotein (PAS positive diastase resistant, and mucicarmine posi-
tive), and the latter contains highly sulfated glycosaminoglycans (alcan blue positive, mucicar- 
mime negative)\textsuperscript{13}.

Electron microscopic findings indicated that pleomorphic adenomas are myoepithelial origin, 
\textit{e.g.}, perinuclear tonofilaments, ectoplasmic actin microfilaments, and remnants of basement 
membrane\textsuperscript{10, 11}.

Immunohistochemically, the ductal epithelial 
component is positive for keratin, epithelial mem-
brane antigen, secretory component, CEA, lyso-
yzyme, epithelial Ig A and M, lactoferrin\textsuperscript{10, 12, 13}. 
But, amylase is usually absent\textsuperscript{13}. The myo-
epithelial component is immunoreactive for 
keratin, actin, vimentin, S-100 protein, and 
laminin\textsuperscript{10, 12, 14, 15, 16}. The latter marker is also 
strongly expressed in the cartilaginous area and in 
a subtype of the epithelial ductal cells. The im-
munohistochemical profile of the epithelial com-
ponent is similar to that of the normal intercalated 
duct cells.

On PAS staining with diastase treatment and 
alcan blue stain in our case, the matrix of the 
tumor revealed positive staining. Also immuno-
histochemical stainings with CEA, vimentin, S-100 
protein showed positive results. The results indi-
cate that the tumor cells of pleomorphic ade-
oma show a bipolar differentiation capability of 
both epithelial and mesenchymal origins, although 
normal myoepithelial cells show only mesen-
chymal characteristics.

Immunohistochemical stainings, light and electro-
n microscope findings contribute to more pre-
cise diagnosis and a better understanding of the 
histogenesis of this tumor.

Some pleomorphic adenomas are extremely cell-
ular and these may be confused with malignant 
tumors. However, the rarity of mitotic figures and 
absence of necrosis are of help in the differential 
diagnosis with true malignant neoplasms. It is 
histologically benign, but clinically malignant be-
cause of frequent recurrent rate directly related 
to the adequacy of the initial surgical procedure\textsuperscript{1}.

Recurrence is very high if the tumor is removed 
by a simple excision. This is because small nod-
ules attached by threadlike filaments of main mass 
may be present surrounding the tumor. If the 
tumor is enucleated, these small remnants will be 
left behind and will provide the nidus for recur-
rence. Also, rupture during the removal of a 
tumor with spillage of tumor fragments into the 
wound causes the common type of recurrence 
cases. Because of these, long-term follow-up is 
necessary to properly evaluate complete surgical 
excision.

Total surgical removal is almost always curative. 
For many tumors located in the superficial lobe 
of the parotid gland, the standard surgery is a su-
perficial parotidectomy with preservation of the 
facial nerve. The incidence of recurrence follow-
ning this procedure is almost zero, in contrast to 
a much higher recurrence rate for simple enucleation\textsuperscript{2}. And so, recurrence of pleomor-
phic adenoma after surgical excision usually reflects 
surgical error.

A pleomorphic adenoma unusually metastasize 
to the lungs, bone, or other organs, with appear-
ing as benign as the original tumors by hemat-
ologic metastasis\textsuperscript{4}. Encapsulated pleomorphic 
adenoma is completely benign and does not un-
dergo malignant transformation, but Tortoledo\textsuperscript{7} 
reported that malignant change developed within 
and already existing benign pleomorphic adenoma. 
Suzuki et al.\textsuperscript{17} reported one recurrent case, 
another malignant case. Sim et al.\textsuperscript{18} reported a 
case in which a solitary pulmonary metastasis 
arose from a pleomorphic adenoma of the parotid 
gland.

In our case, there was no metastatic evidence 
to other organs. Because we thought that this case 
arose from the skin, we removed the tumor mass 
without salivary gland capsule. But we have not 
detected any signs of recurrence of this tumor.

In Korea, Haw et al.\textsuperscript{10} reported "a case of 
mixed tumor" in the central portion of the cheek 
in the dept. of Dermatol. and Bang et al.\textsuperscript{20} and 
Lee et al.\textsuperscript{21} reported cases in the dept. of 
Otolaryngol.

In the dermatologic field, we emphasize that the 
tumors of salivary glands should be included in 
the diagnosis of the tumors on the face.

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