PAPILLARY CYSTADENOMA LYMPHOMATOSUM

A RARE TERATOID OF THE PAROTID REGION

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In a diagnostic material of several hundred thousand cases examined by me since 1895, there have been over seven hundred mixed tumors of the parotid gland, and over five hundred branchial cysts of the cervical region, and two cases only of a form of tumor of the parotid region which I must believe to be of very great rarity, not only because of its extremely low incidence in my material, but also because of the silence in the literature regarding it. These cases were as follows:

Case I. No. 1431-L-AD. Mr. J. McD., age about 45. Tumor from lower pole of parotid, about half as large as a hen’s egg. Slowly growing for many years. Pathologic Diagnosis: Papilliferous cystadenoma, the papillae filled with lymphoid tissue with many germ centers. The epithelium of the cyst spaces is a stratified columnar epithelium of respiratory type. It is undoubtedly a congenital disturbance of development, a teratoid cystadenoma. It may represent an accessory auditory structure. Not malignant.

Case II. No. 4887-L-AF. Mr. C., age 60. A completely encapsulated tumor 2 cms. in diameter, removed from beneath the lower pole of the left parotid gland. Tumor has been there 20–30 years, but has increased in size more rapidly recently. Pathologic Diagnosis: A congenital papillary cystadenoma, lined with respiratory epithelium, the papillae containing much lymphoid tissue, with germ centers. A congenital disturbance of development; may represent accessory ear structure (Eustachian tube mucosa). Not malignant, and should not recur if all removed.

These two tumors are practically identical in structure, so that they may be described as one. They were wholly encapsulated, elastic, oval tumors, with little or no surrounding in-
flammatory reaction, and no evidences of infiltration. They were regarded as enlarged lymphnodes. On section they were found to be cystic, the cyst-spaces filled with papillae. The fluid content was small, serous rather than mucoid. Throughout the papillae whitish nodules, corresponding to

![Image](image-url)

**Fig. 1. Case I. Papilliferous Cystadenoma with Lymphoid Follicles and Germ Centers in the Papillae, which are Covered with a Stratified Columnar Epithelium, Partly Ciliated and Partly Showing a Cuticular Border.**

the germ centers, could be seen. Microscopically, the structure of the two tumors is practically identical. Inside the capsule is a narrow zone of lymphoid tissue containing germ centers, but no evidences of lymph sinuses, or a division into cortical and medullary portions. This lymphoid tissue is arranged in many-branched papillae covered with stratified columnar epithelium, having a well-defined cuticular border showing cilia distinctly over the greater part. In no place was the epithelium of a squamous cell type. The uppermost
layer of epithelium was of a tall columnar type, with deeply-staining nuclei at the outer edge of the cells. Beneath the columnar cell layer there were 2–3 layers of polygonal or cuboidal cells having more lightly staining nuclei and a smaller amount of cytoplasm. There was no evidence of mucin-

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**Fig. 2. Higher Power View of Portion of Preceding.** Albuminous precipitate in cyst spaces between papillae.

formation in the columnar layer, but clear vacuoles appeared here and there in the rather coarsely granular cytoplasm of the basal layers. Few mitoses were seen in the epithelium. The cilia were very distinct in many places; in others the cells appeared as if bordered by a hyaline cuticular zone about \( \frac{1}{2} - \frac{1}{4} \) as thick as the whole epithelial layer. Between the papillae there was a rather coarsely granular albuminous precipitate containing a few leukocytes and desquamated cells. This stained red with eosin, but was not dense enough or sufficiently hyaline to be regarded as colloid, and showed no con-
centric lamination. No trace of mucin was found between the branching and anastomosing papillae. On the whole there was very little space between the latter for fluid, the greater part of the intracapsular space being filled with the closely packed papillae. The epithelium rested upon a very narrow, almost imperceptible, basement membrane. The remainder of the stroma consisted of lymphoid tissue, a delicate stroma packed with lymphocytes, and showing numerous large germ centers. The latter were especially developed in the broadest portions of the papillae, which were roundish and plump. The narrower papillae without germ centers were villus-like

![Image](image_url)

**Fig. 3. Higher Power View of Portion of Papilliferous Cystadenoma, Showing Character of Epithelium and Lymphoid Tissue in Papillae.**

or spatulate in form. The tissue was not very vascular; the arterial supply small, and the veins not large and were thin-walled. No lymphatic trunks were found piercing the capsule. No evidence of malignancy was present in either tu-
mor; the growth was typical and regular, stratified ciliated columnar epithelium upon a lymphadenoid submucosa, without any evidence of infiltration. These two cystic tumors resembled branchial cysts in every way, except for the character of the epithelium, ciliated stratified columnar instead of a stratified squamous epithelial lining. Branchial cysts have the same subepithelial layer of lymphadenoid tissue and germ centers. While many of them are papillomatous they do not

Fig. 4. High Power View of Papilla.

as a rule show such a degree of intracystic papilliferous growth as in the case of these two cysts. In both cases the parotid, showing some degree of pressure atrophy, was entirely outside of the cyst wall and wholly detached from it.

It is evident from the structure described that we are dealing here with a heterotopia of mucous membrane from the pharyngeal entoderm, representing either the upper respiratory tract or the Eustachian tube. The type of mucosa repre-
presented in this tumor is precisely like that of part of the Eu-
stachian tube which has a tall stratified ciliated columnar epi-
thelium with many lymphoid follicles beneath the epithelium. 
In no other part of the respiratory tract is there such a close

![Image of papillary cystadenoma lymphomatous]

FIG. 5. CASE II. LOW POWER VIEW OF PAPILLIFEROUS CYSTADENOMA, SHOWING LYMPHOID TISSUE IN PAPILAE.

resemblance between the normal mucosa and the mucosa lin-
ing these cystic tumors. Bearing out this resemblance is the oc-
currence in my material of a polypoid tumor of the Eustach-
chian tube which had identically the same structure as these 
two tumors except that it was not enclosed in a cyst wall, but 
presented a papillomatous growth covered with stratified cili-
ated epithelium, the stroma of the papillae containing diffuse 
lymphoid tissue with large and numerous germ centers. This 
polyp of the Eustachian tube is the only structurally closely 
related growth to these two paraparotid cysts that has oc-
curred in my service. No nasal or nasopharyngeal polyp,
out of the many examined in my service, has shown the same close resemblance. I am, therefore, inclined to believe that these two papilliferous cystadenomas with lymphoid stroma represent a developmental disturbance of the ear—accessory Eustachian tube anlage, which has grown slowly through the years, assuming neoplastic tendency rather late in life.

I have been unable to find descriptions of similar cystic lymphoid papillary adenomas in any of the special works on neoplasms. They are not mentioned by Ewing. In the liter-

Fig. 6. Higher Power View of Preceding, Showing Character of Epithelium and Lymphoid Tissue of Stroma.

ature I have found only one report of what are apparently identical tumors. Albrecht and Arzt (1) describe two cases which, according to the description given and the illustrations given of their structure, are without any doubt wholly similar to mine. One was a tumor the size of a small apple from the left parotid region of a man 64 years of age, the second from
the submaxillary region of a girl of twelve years. Both were papillary cystadenomas with stratified columnar epithelium with a cuticular border, and a lymphoid stroma containing lymph follicles and germ centers. These tumors were regarded by Albrecht and Arzt as heterotopias of pharyngeal entodermal anlage into lymph nodes. Clinically and anatomically they presented the picture of lymphomas, microscopically they were interpreted as papillary cystadenomas enclosed within typical lymph node tissue. I differ in my interpretation of the lymphoid tissue, as being an essential part of the misplaced tissue (Eustachian tube mucosa), and not as representing a lymph node into which the epithelial elements alone have been misplaced. Further, Albrecht and Arzt describe the epithelium covering the papillae in their two tumors as non-ciliated but with a cuticular border. Their
illustrations make it evident that a ciliated border was present.

As the branchial cysts represent heterotopias or dystopias of pharyngeal mucosa (squamous epithelium and lymphoid tissue), so these two papillary cystadenomas represent a dystopia of Eustachian tube mucosa (ciliated stratified columnar cells and lymphoid tissue), to the parotid region. This latter form is very much rarer than the branchial cyst; it is also much rarer than thyreoglossal remains, which are found near the middle of the neck, have a single layer of columnar cells, without lymphoid tissue and usually show thyroid acini in the cyst wall. As to the clinical significance of such a papillary cystadenoma lymphomatous, its excessive rarity makes it slight. In three of the known four cases the cyst began to show neoplastic tendency at the age of 45
in one case, 60 in a second and 64 in the third case; hence the growth is very slow. There is, of course, the possibility of a malignant transformation of such heterotopic tissue, and the development of an adenocarcinoma. Such an event has not been observed as yet, although it is possible that some of the adenocarcinomas developing primarily in the parotid region may have had such an origin.

In conclusion, this paper presents descriptions of two cases of *papilliferous cystadenoma lymphomatosum*, a very rare teratoid of the parotid gland region, representing a heterotopia or dystopia of pharyngeal entoderm, resembling in structure most closely the mucosa of the cartilaginous portion of the Eustachian tube.

**REFERENCE**