# BENIGN LYMPHOEPITHELIAL CYSTS (ECTOPIC SALIVARY GLAND TISSUE) IN LYMPH NODES

## Report of Two Cases

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YSTIC changes in a cervical lymph node containing ectopic salivary gland tissue are rarely seen by the pathologist and are highly confusing to the surgeon; this is the report of two such cases.

## Case Reports

Case 1. A 42-year-old woman, a school teacher, had been treated at the Cleveland Clinic for several years for an anxiety tension state and mucous colitis. On July 22, 1958, during a routine physical examination a 0.5-cm. firm, solitary nodule was palpated in the right lobe of the thyroid gland. In the right digastric triangle adjacent to the right submaxillary salivary gland a 2.0-cm. firm mass believed to be a lymph node was felt. The results of the remainder of the physical examination and of the laboratory and roentgen studies were within normal limits.

On July 30, 1958, a lobectomy of the right side of the thyroid was performed. There was no lymphadenopathy in the vicinity of the thyroid gland. The nodule in the thyroid was diagnosed as papillary carcinoma. In view of this diagnosis, it was considered advisable to start desiccated thyroid therapy immediately. During this treatment, from August through January, the submaxillary node remained unchanged. On January 26, 1959, the node was excised. The patient has continued the thyroid therapy for the papillary carcinoma and has done well to the date of this writing.

The gross excised specimen was soft, ovoid, and reddish-tan, measuring 1.1 cm. in its greatest dimension. A thin fibrous capsule was noted. The cut surface was moist, tan, and stippled with yellow puncta that were less than 0.1 cm. in diameter.

The microscopic findings confirmed the encapsulated nature of the lesion. A peripheral sinus and lymphoid follicles characteristic of a lymph node were found in the peripheral or cortical areas. The hilus of the node was also included in sections and contained afferent and efferent lymphatics. A small amount of salivary gland tissue was present immediately adjacent to the node at the hilus. Scattered throughout the lesion were numerous cystic structures of various sizes (Fig. 1). The largest ones measured up to 1.5 mm. in diameter, and the smaller cysts were approximately 100 to 200  $\mu$  in diameter. Most of the smaller cysts were arrayed in small clusters. Each cyst was lined with squamous epithelial cells, this pavement epithelium ranging from 1 cell to 6 cells in thickness (Figs. 2 and 3). In some cysts a delicate fibrous enveloping layer was present, and the epithelial cells appeared to rest upon this. However, in many

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Fig. 1. Photomicrograph of a section of a cervical lymph node, showing numerous large and small cysts scattered throughout. Lymph follicles and a portion of peripheral sinus are at the left. A small island of salivary gland tissue is outside the node at the hilus. Hematoxylin-cosin—methylene blue stain; magnification X 12.

cysts this fibrous layer was discontinuous. Many of the cysts contained amorphous acidophilic material, and in a few there was infiltration by histiocytes and neutrophils. No transmural migration of lymphocytes was noted. Occasional small solid islands of epithelial cells were also present.

**Case 2.** A 59-year-old man was examined for a mass, posterior to the left ear, which had been present one year. During the past three months the patient had noted discomfort upon turning his head to the left. There were no other symptoms related to the mass. An irregular, movable, nontender, 3.5-cm. mass was palpated adjacent to the posterior border of the left parotid gland. This mass appeared to extend into the space behind the mandible and posteriorly beneath the sternocleidomastoid muscle. On March 25, 1960, this area was explored surgically. Several lymph nodes were removed from the area adjacent to the posterior border of the parotid gland. No other masses were present.

The largest node measured 2.0 cm. in diameter; the smallest was 0.3 cm. in diameter. The gross and microscopic pathologic findings were identical to those described for the specimen in case 1.

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Fig. 2. Photomicrograph of a section of a cervical lymph node, showing a small cyst and squamous epithelial island just above the peripheral sinus of the node. Masson trichrome stain; magnification X 120.

## Discussion

Three groups of lymph nodes are associated with the parotid salivary gland: one superficial, one within, and one deep to the gland.<sup>1</sup> These are true lymph nodes possessing both subcapsular and medullary sinusoids and having afferent and efferent lymphatics.<sup>2</sup> Neisse<sup>3</sup> demonstrated numerous true lymph nodes in the parotid gland of a 120-mm. fetus. He believed that later the nodes become encapsulated and separate from parotid tissue to form preparotid nodes, some of them containing salivary tissue. In an examination of the parotid glands of 14 newborn infants, he found an average of from 8 to 14 lymph nodes in each parotid, with salivary tissue in every one. Bernier and Bhaskar<sup>2</sup> reported 11 cases of ectopic salivary gland tissue in lymph nodes. They applied the term *benign lymphoepithelial cyst* to these cases.

The ectopic salivary tissue in lymph nodes can give rise to a variety of lesions characterized by pathologic changes in both the epithelial and the lymphoid tissue. Godwin<sup>4</sup> proposed the general term *benign lymphoepithelial lesion* for these cases. In an exhaustive review of the subject, based upon the examination of 186 case records from the Armed Forces Institute of Pathology, Bernier and Bhaskar<sup>2</sup> presented convincing evidence that the following benign tumors arise in ectopic salivary gland tissue in lymph nodes both within and outside the parotid gland.

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Fig. 3. Photomicrograph of a section of a cervical lymph node, showing the characteristic squamous epithelial lining of one of the cysts. Hematoxylin-cosin—methylene blue stain; magnification X 430.

- I. Single or multiple cysts in lymph nodes (benign lymphoepithelial cyst)
- II. Papillary cystadenoma lymphomatosum (Warthin's tumor)
- III. Adenoma lymphomatosum

IV. Mixed tumor in lymph nodes (pleomorphic adenoma lymphomatosum) They based their conclusions upon the fact that many parotid and cervical lymph nodes contain ectopic acinar and ductal elements of salivary tissue. The case for papillary cystadenoma lymphomatosum is based on the demonstration of lymph node structure in uninvolved areas of the lesion, and on the occurrence in other locations of tumors that have an identical epithelial component but lack the lymphocytic element. The same authors<sup>5</sup> suggest that branchial cysts that are not connected to the skin or the pharynx may arise from ectopic epithelial tissue in cervical lymph nodes.

Our cases are in group I of the Bernier and Bhaskar<sup>2</sup> classification of benign lymphoepithelial lesions. We have presented these cases because of the rarity of the lesion and also to point out the significance of the finding of ectopic salivary gland tissue in a lymph node.

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## Summary

1. Lymph nodes of the parotid salivary gland and other cervical areas may contain ectopic salivary gland tissue.

2. Benign cystic and solid lymphoepithelial tumors may occur in such lymph nodes.

3. Two cases of benign lymphoepithelial cysts (ectopic salivary gland tissue) in submaxillary lymph nodes are presented.

### References

- 1. Gray, H.: Anatomy of the Human Body. 25th ed., edited by C. M. Goss. Philadelphia: Lea & Febiger, 1948, 1478 pp., p. 703.
- 2. Bernier, J. L., and Bhaskar, S. N.: Lymphoepithelial lesions of salivary glands; histogenesis and classifications based on 186 cases. Cancer 11: 1156-1179, 1958.
- Neisse, R.: Ueber den Einschluss von Parotisläppchen in Lymphknoten. Anat. Hefte (Wiesb.) 10: 287-306, 1898.
- Godwin, J. T.: Benign lymphoepithelial lesion of parotid gland (adenolymphoma, chronic inflammation, lymphoepithelioma, lymphocytic tumor, Mikulicz disease); report of 11 cases. Cancer 5: 1089-1103, 1952.
- 5. Bhaskar, S. N., and Bernier, J. L.: Histogenesis of branchial cysts; report of 468 cases. Am. J. Path. 35: 407-423, 1959.