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## LYMPHOEPITHELIAL LESIONS OF SALIVARY GLANDS

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**INTRODUCTION:** A lymphoepithelial lesion is one in which the lymphoid and epithelial components predominate, creating the distinctive histology picture for that lesion. There is a close connection between lymph tissue and the epithelium that covers it. This connection is known as lymphoepithelioma, and the lesions that result from it are called lymphoepithelial lesions.

Salivary gland lesions are frequently observed in the para-oral regions, and due to their mode pattern, the parotid glands are the most frequently impacted. A loose collection of lymphoid aggregates called the parotid gland experiences encapsulation, which traps the lymph nodes inside the gland.

It could be classified as

- 1) a Warthin's tumour (cyst adenolymphoma, papillary cystadenoma lymphomatosum).
- 2) Noncancerous lymphoepithelial lesions, such as adenolymphoma, lymphoepithelioma, and Mikulicz's disease and syndrome, Sjogren's disease.
- 4) A harmless lymphoepithelial cyst.
- 5) Malignant lymphoepithelial lesion (lymphoepithelial carcinoma)<sup>1</sup>.

**WARTHIN'S TUMOR:** Hildebrand first identified it as a congenital disorder in 1895. Two cases of papillary cystadenoma with lymphoid component were reported by Albrecht and Arzt in 1910. The lesions to be similar to pharyngeal endoderm. Only a few cases of heterotopic salivary gland adenomas were reported by Nicholson in 1923. Warthin reported a few cases in 1929 and named or created the term papillary cystadenoma lymphomatosum, which is still used today.

**HISTOGENESIS:** Many studies have been conducted to understand its pathogenesis, which has been the subject of debate. Hildebrand thought it was a remnant of the brachial pouches and a type of lateral cervical cyst. Heterotopic salivary rests in parotid lymph nodes were suggested as the origin by Albrecht and Arzt. According to Warthin, it develops from heterotopic pharyngeal endoderm. Ectopic eustachian tube endoderm had been proposed by Hevenor and Clark as the tissue of origin.

Other histogenesis theories have:

- Orbital inclusions as a component.
- Homotypic oncocytes
- Glands that produce sebum
- Heterotopic lymphoid endothelium,

The parotid ductile epithelium in lymphnodes close to the parotid gland was revealed to be the tissue of origin by the embryology-based studies of Thompson and Bryant. Azzopardi and Smith's histochemical analyses further suggested that these tumours originated from salivary ducts in lymphoid stroma. Azzopardi and Hou demonstrated the theory of enclaved parotid epithelium in a lymph node.

Allegra proposed a delayed hypersensitivity as the primary cause, suggesting the following series of events based on the immunohistochemical investigations: - oxyphilic metaplasia of striated ducts, - papillary formations with secretion, - cyst formation, - basophil and histiocyte infiltration of the basement membrane that results in a fully delayed hypersensitive response. According to Hsu et al histochemical 's studies, the lymphoid component of the tumour is merely an exacerbated secretary immune response. The currently accepted theory, however, states that salivary gland tissue that became trapped inside the Para parotid and intra-parotid lymph nodes during development is what gives rise to the tumour.

**CLINICAL FEATURES:** The average patient age at the time of discovery was 56 years old, with 82% of patients falling between the ages of 41 and 70. It displays a preference for men in a 5:1 ratio. Less than 10% of cases exhibit symptoms like pain, pressure, or a sudden increase in tumour size, and the majority of cases are asymptomatic. The parotid gland makes up 6–10% of all parotid gland tumours and is the most frequently affected site or gland. It is typically bilateral and affects the lower pole or apex of the parotid gland. Submandibular gland involvement is also occasionally observed. However, due to the proximity of the parotid gland's apex to the submandibular gland and the parotid gland's involvement, it is doubtful whether it could be classified as a primary lesion. The tumour is clearly defined, typically superficial and soft or fluctuant, or firm to the touch if it is deep and on a gland.

**MACROSCOPIC APPEARANCE OF THE TUMOR:** The surgical specimen is typically an oval or spherical mass covered by a usually intact thin, tough capsule. The surface of the tumour is lobulated, smooth, and red-gray. It has a range of consistencies, from fluctuant with cystic space to firm and rubbery. The fluid in the cystic compartments has a range of viscosities and can be clear, serous, mucoid, have a brown tint, or be semisolid caseous material. The cyst's lining is shaggy and asymmetrical, with numerous indicating papillae extending from the cyst wall. The grey solid areas have white nodules that represent lymphoid follicles.

**MICROSCOPIC FEATURES:** A distinctive and pathologic picture is created when lymphoid matrix and papillary projections of eosinophilic epithelial cells form cystic spaces.

**EPITHELIAL COMPONENT:** Two layers of uniformly spaced epithelial cells make up this arrangement. The cystic space is approximated by tall columnar cells. The centrally located, darkly stained, pyknotic nuclei of the columnar cells are close to the luminal space. Cuboidal and polygonal cells with nuclei that have noticeable nucleoli make up the inner layer. Both cell layers have cytoplasm that is clearly eosinophilic and finely granular. When using phosphotungstic acid—hematoxylin stain to demonstrate the density of mitochondria, staining intensity varies. The cystic spaces where the epithelial papilla protrude are frequently filled with uniform eosinophilic granular material that stains with periodic acid Schiff stain. The lymphoid stroma and epithelial cells are separated by a thin layer of basement membrane.

**LYMPHOID COMPONENT:** With germinal centres, sporadic sinusoids, and subcapsular spaces, the lymphoid component appears reactive; however, the amount of lymphoid stroma is variable and may only contain a scant lymphoid infiltrate. Oncocytosis, squamous metaplasia, mucous cell, prosoplasia, and infrequently sebaceous gland inclusions are the most common histologic variations. Sheets and nests of disorganised oncocytic cells with lost papillary formation are the hallmarks of oncocytosis. It is important to remember that focal oncocytosis is not a distinct neoplasm or a malignant degeneration. Squamous metaplasia frequently manifests as focal areas with flattened luminal surfaces and a loss of regular columnar cells.

In the tissue of the salivary gland, mucous cells are typically present and interdigitated between the epithelial cells. It may exhibit spontaneous necrosis and squamous metaplasia if inflammation is present. Studies using immunohistochemistry on luminal epithelial cells show basal cell layer IgA and peanut agglutinin reactivity. Negative findings for Warthin's tumour include IgA secretory component and series on S-100 protein in salivary gland tumours<sup>3</sup>.

### **DIFFERENTIAL DIAGNOSIS:**

- Sebaceous lymphadenoma.
- Acinic cell carcinoma.
- Mucoepidermoid carcinoma.

**ULTRASTRUCTURAL FEATURES:** When compared to the normal striated ducts of the parotid gland, which range in size from normal to three times larger with pleomorphic forms, they do not have packed mitochondria. Instead, the majority of epithelial cells exhibit prominent mitochondria throughout the cytoplasm, and those cells that are close to the cystic luminal exhibit densely packed mitochondria. The lamellar sheaves of the aberrant mitochondria are more numerous, longer, and tightly packed. Cristea can form spherical concentric rings or rouleaux, or they can be haphazardly arranged in villous forms. If tonofilaments are present, they are rarely seen and usually show significant metaplasia. The presence of true cilia is confirmed by electronic microscopy, and microvilli are frequently seen on the apical surfaces of epithelial cells. Kim et al. used cytochemical studies to demonstrate the existence of lysosomal granules that contain acid phosphatase and are connected to a lytic process.

**TREATMENT AND PROGNOSIS:** Surgery is typically used to treat this condition. This tumour is well encapsulated, so it is said to be extremely rare to recur. But because it is multifocal, it is challenging to assess accurately. It is extremely uncommon for either the lymphoid or epithelial component to develop malignant transformation<sup>4</sup>.

**BENIGN LYMPHOEPITHELIAL LESION [BLL] [MIKULICZ'S DISEASE]:** According to Batsakis, a lymphoepithelial lesion is an advanced epithelial alteration that is characterized by a lymphocytic infiltration of the salivary parenchyma. The lesion has gone through several stages of chronic inflammation to reach this stage. Batsakis used the terms chronic lymphoepithelial sialadenitis, chronic punctate parotitis, or chronic non-specific sialadenitis to describe a gland or part of a gland that has been completely or nearly completely replaced by a chronic inflammatory infiltrate and in which only islands of metaplastic ducts are visible<sup>5</sup>.

**PATHOGENESIS:** The etiology of the lesion is not clear. Mikulicz suggested that it was the result of an infectious or parasitic process penetrating the gland locally rather than hematogenously. Morgan and Castleman considered it as primary lesion of the salivary duct system with secondary infiltration. Bernier suggested that it is neither a neoplasm nor a lesion in which epithelium plays a aggressive or dominant role. These are autoimmune disease in which the patient's own salivary gland tissue becomes antigenic.

**CLINICAL FEATURES:** The BLL typically manifests as an asymptomatic or barely noticeable firm swelling of the parotid gland. Any salivary gland can experience them. The enlargement is non-tender, nodular or diffuse in distribution, and there is no evidence of attachment to the nearby structures on palpation. The size of enlargements can be unilateral or bilateral and range, but they are typically only a few cm in diameter. Sometimes, a fever, an upper respiratory infection, an oral infection, a tooth extraction, or another local inflammatory disorder precedes the onset of the lesion<sup>6</sup>.

**HISTOLOGICAL FEATURES:** An orderly lymphocytic infiltration of the salivary gland tissue, the destruction or replacement of the acini, and the persistence of islands of epithelial cells—likely representing the remnants of gland ducts—are the hallmarks of the lesion. Although the lymphoid element is typically diffuse, occasionally one can see actual germinal centres. The epithelial components can be solid nests or clumps of ill-defined epithelial cells that Morgan and Castleman referred to as "epi-myoeptithelial islands" as the disease progresses or ducts exhibiting cellular proliferation and loss of polarity. It has been proposed that both ductal cell and peripheral myoeptithelial cell proliferation causes these islands to form. Eosinophilic and hyaline material deposition in epithelial islands is also seen in advanced lesions.

**DIFFERENTIAL DIAGNOSIS:** The disease's differential diagnosis includes illnesses like malignant melanoma, chronic sialadenitis, papillary cystadenoma lymphomatosum, and uveoparotitis.

**TREATMENT:** Treatment with surgical excision is the best option. It is not advised to undergo radiotherapy because it may cause cancer. Positive prognosis.

**SJOGREN'S SYNDROME** [ Sicca syndrome, Gougerat-Sjogren syndrome]: Since 1965, it has been defined as the triad of (a) kerato conjunctivitis sicca, initiated by lesions in the lacrimal glands, (b) xerostomia with or without salivary gland involvement due to damage to the salivary glands and mucosa glands of the oral cavity, and (c) a connective tissue disease typically affecting the thyroid and ovaries. In 1925, Giugerot recognized a generalized condition that involved dryness. However, primary Sjogren's syndrome, also known as the sicca complex, and secondary Sjogren's syndrome, are now recognized as two distinct medical conditions<sup>7</sup>.

**ETIOLOGY:** The precise cause of the syndrome is unknown, but a number of causes, including altered immune responses and combinations of extrinsic & intrinsic factors as well as genetic, hormonal, infectious, and immunologic factors, have been proposed. HIV may be an etiologic agent based on the clinical similarities between Sjogren's and the salivary disease associated with HIV. It is unknown whether some Sjogren's syndrome (SS) patients have anti-HIV antibodies. Additionally, it has been proposed that these antibodies might be induced by a retrovirus related to HIV or that they might be cross-reacting auto-antibodies.

Additionally, laboratory results point to an autoimmune etiological role. Patients with Sjogren's syndrome who have salivary glands have been shown to have Epstein-Barr virus. However, normal people's salivary glands have also contained viruses. Therefore, it is believed that the Epstein-Barr virus may not be the primary cause of this condition. Consequently, its function is probably secondary. Xerostomia is the most frequent presenting complaint when symptoms are present. 30% to 50% of patients experience enlargement of the salivary glands. Any gland may be affected; the parotid glands are most frequently affected. Affected glands may be unilateral or bilateral.

**CLINICAL FEATURES:** It displays a preference for women in a ratio of 10 to 1. Over 50 is when it starts to happen. The main oral symptoms are dryness; however, some people may also experience difficulty swallowing, issues with dentures, changes in taste, an increase in caries, chronic symptoms of burning oral mucosa, or an inability to speak continuously for more than a few minutes. The oral symptoms typically begin slowly and may advance gradually.

Indicators of chronic erythematous candidiasis include angular cheilitis, which is characterised by dry, sticky oral mucosa surfaces, primary or recurrent dental caries in cervical or incisal locations, no saliva or cloudy saliva expressed from the major salivary gland ducts, patchy or generalised oral mucosal erythema, and no or little saliva<sup>8</sup>.

**SALIVARY GLANDS:** The major salivary glands may develop firm, diffuse, non-tender, or slightly tender enlargements, typically bilaterally. In early or mild cases, the glands may indurate slightly without enlarging. The size of chronic enlargement slowly fluctuates. At a significant salivary gland duct orifice, a cream-colored exudate might be visible. There is hypergammaglobulinemia. There are numerous other autoantibodies that can be shown.

**OCULAR SYMPTOMS:** The term "keratoconjunctivitis sicca" refers to a specific condition that can occur alone or be linked to sarcoidosis and involves both a decrease and qualitative changes in the tear film. Discharge and photophobia are symptoms.

Extra glandular: The primary form of SS is present in about 50% of patients, who typically do not progress to other connective tissue diseases. Raynaud's phenomenon is one of the extra glandular characteristics that patients with primary SS can acquire. The central nervous system may become affected, along with primary biliary cirrhosis, diffuse interstitial lung disease, interstitial nephritis, chronic atrophic gastritis, peripheral neuropathies, and inflammatory vascular disease.

**HISTOPATHOLOGY:** The primary salivary gland exhibits three different types of histologic changes,

A significant lymphocytic infiltration of the gland replaces all acinar structures while leaving the lobular pattern intact.

b) The development of epi-myoe epithelial islands as a result of both ductal epithelial cell and myoe epithelial cell proliferation.

c) Merely the glands shrinking after the lymphocytic infiltration.

Additionally, according to research by Bertram et al., 85% of SS patients had changes in their accessory salivary glands on their lips that were similar to those in their major glands. Therefore, they hypothesised that a labial mucosal biopsy would aid in making the diagnosis of the condition. However, patients with SS and progressive systemic sclerosis (scleroderma) showed a different pattern in labial biopsy.

- Fibrosis with lymphocytes
- Fibrosis without lymphocytes
- Normal salivary gland are all variations.

The fibrosis may reflect features of systemic sclerosis than being a manifestation of SS<sup>9</sup>.

**MINOR SALIVARY GLANDS:** Chisholm et al has demonstrated that while foci of lymphocytes are not typically found in the minor salivary glands, they are prevalent in a significant portion of patients with SS. These foci will be visible in a lip biopsy if salivary gland tissue is present, and evidence suggests that their size and number are related to how severe the disease is. In mild cases, there are only a small number of these foci, which are densely packed with plasma cells. In serious cases, the foci are larger, more numerous, and contain fewer plasma cells.

**LABORATORY FINDINGS:** Three tests are primarily used to assess salivary gland function:

- Salivary scintigraphy
- Minor salivary gland biopsy
- Parotid flow rate

a) Parotid flow rate: Saliva is collected by stimulating the gland with lemon juice every 30 seconds for 10 minutes. The saliva is then collected by placing a Lashley, Carlson-Crittendon, or other specially fabricated cup over the stensons duct orifice. At least 5ml of secretions from each gland should be within the normal range.

b) Minor salivary gland biopsy: The major salivary glands are challenging to biopsy and may cause facial nerve paralysis when combined with parotid biopsy. A minor salivary gland biopsy is much less difficult and reveals histologic signs of disease.

Stool scintigraphy

c) A decrease in the total uptake of the isotope by the salivary glands, slow uptake, or abnormal results from two of the three tests in a patient undergoing salivary scintigraphy demonstrate the involvement of the salivary glands<sup>10</sup>.

**TREATMENT:** Sjogren's syndrome does not have an effective symptomatic treatment. which also recommends using fluoride frequently to prevent tooth decay and ocular lubricants for keratoconjunctivitis and saliva substitutes for mouth dryness.

Only in cases of extreme enlargement and if it is problematic is surgery advised. Radiotherapy was once recommended, but it is no longer supported because it is believed to cause malignant transformation.

**COMPLICATIONS:** An association with lymphoma malignancy complicates the prognosis for Sjogren's syndrome.

**LYMPHO-EPITHELIAL CYST:**

**BRANCHIAL CLEFT CYST:** Typically, lymphoepithelial cysts are small, elevated, submucosal tumours that range in colour from soft pink to yellow. The floor of the mouth, the ventral and posterolateral surfaces of the tongue, the soft palate, the anterior palatine pillar, and the buccal vestibule are the most frequent sites. Most lesions are asymptomatic, but

a few patients have complained of swelling or discharge. Males are more frequently affected than females, with their size rounded from 1 to 10 mm and swelling of the oral mucosa of normal colour, with the exception of when they were large when they were yellow or white<sup>11</sup>.

**PATHOGENESIS:** According to Bhasker and Bernier's theory, salivary gland epithelium that is imprisoned in the neck nodes during embryogenesis experiences cystic changes. They suggested that the lesion be referred to as a lympho epithelial cyst rather than a branchial cyst because it is thought that the ectopic glandular epithelium found in the lymphoid tissues of the oral mucosa may undergo cystic changes and develop into a lympho epithelial cyst. The lymphoepithelial cyst, which forms in oral tonsils when the crypt opening plugs up, was described by Knapp as a pseudocyst. Due to an accumulation of desquamated cells and keratin, the blocked crypt widens and expands.

**CLINICAL FEATURES:** Benign lymphoepithelial cysts of the parotid gland are typically well-circumscribed, asymptomatic masses in the superficial position of the gland. Rarely, they may be associated with facial paralysis and occasionally they may be tender or painful masses. Cysts feel firm or rubbery to the touch, but they could also be compressible. Their average diameter is 2.5 cm, and they typically have sizes between 0.5 and 0.6 cm. Bilateral occurrences are extremely rare.

**GROSS FINDINGS:** These are unilocular, fluctuant, and ovoid with a sharp border. Many cysts have small protrusions and an impact granular appearance to the luminal surface, which is caused by reactive lymphoid follicles in the cyst wall. Most of these cysts contain soft, yellow-white caseous material, though some are fluid-filled. The luminal surface is shiny and can range from yellow brown to white.

**MICROSCOPIC FEATURES:** Squamous cuboidal, columnar, and pseudostratified ciliated types of epithelium may line the lymphoepithelial cyst of the parotid gland. Squamous epithelium that has been stratified lines the majority of the cysts. The lymphoid tissue typically surrounds the entire cyst and displays typical germinal centres, but in some instances, only a diffuse, dense infiltrate of lymphocytes may be visible. Mostly desquamated parakeratotic cells make up the lumen. The defining characteristic of a lymphoepithelial cyst is the abundance of lymphoid tissue.

**DIFFERENTIAL DIAGNOSIS:** It should be distinguished from mucoepidermoid carcinoma, acinic cell carcinoma, Warthin's tumour, and metastatic carcinoma when it affects the salivary gland. Localized and conservative excision is the method of treatment.

**LYMPHOEPITHELIAL CARCINOMA:** Lymphoepithelial carcinomas have similar histomorphology growth patterns and architectural characteristics to early lymphoepithelial lesions. Minor salivary gland origin tissue tumours are extremely uncommon and most often occur in the parotid gland and submandibular gland. The epithelial foci found in the benign lymphoepithelial lesion, according to Morgan and Castleman in 1953, may have been ductal remnants undergoing reactive myoepithelial proliferation. To describe this structure, they coined the term epi-myoeplithelial lands<sup>12</sup>.

**CLINICAL FEATURES:** The ratio is slightly less than two to one, with females being more affected than males. The primary physical finding is an indurated mass in the parotid region, which accounts for 90% of parotid gland involvement. Patients' complaints of pain, discomfort, and drainage account for about half of the cases. Less than 20% of people may experience facial nerve paralysis.

**MACROSCOPIC APPEARANCE:** The solid tumour is 2-3 cm in diameter, but it may be larger. The tumour in the parotid gland is firm, fixed, and typically bounded. The terms grossly encapsulated, partially circumscribed, multinodular, and infiltrate have all been used to describe tumours. The tumor's c-cut section varies from firm grey to tan to yellow grey.

**MICROSCOPIC FEATURES:** Lymphoepithelial carcinoma has oval and irregular elongated epithelial islands that rarely anastomose and are isolated by a benign lymphoid stroma, similar to benign lymphoepithelial lesions at lower magnifications. Ducts and acini can also be seen. The epithelial cells appear polygonal to spindled at higher magnification, without keratinization or luminal formation. Large nuclei make it simple to see some vesicular mitosis.

**HISTOCHEMICAL FINDINGS:** Cytokeratin S-100 protein and muscle specific actin protein are expressed in epithelial component of lympho epithelial carcinoma. Diagnostic electron microscopy has shown round nuclei with prominent nucleoli, marginated chromatin, and a paucity of cytoplasmic organelles to demonstrate desmosomes and tonofilaments.

**DIFFERENTIAL DIAGNOSIS:** The differential Diagnosis of the disease includes Squamous carcinoma & Mucoepidermoid carcinoma with lymphoid infiltration.

**TREATMENT:** It is treated with surgery or surgery in combination with radiotherapy.

**CONCLUSION:** A lymphoepithelial lesion is one in which the lymphoid and epithelial components predominate, creating the distinctive histology picture for that lesion. There is a close connection between lymph tissue and the epithelium that covers it. This connection is known as lymphoepithelioma, and the lesions that result from it are called lymphoepithelial lesions. Hence more studies must be conducted based on the literature available.

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