

CASES OF PATHOLOGIC INTEREST

DONALD A. KERR, A.B., D.D.S., M.S.

THE cases in this report were selected from our routine material because of some interesting clinical or pathologic feature. They have little that is new to add to our clinical and pathologic knowledge, but perhaps will refresh for us some of the clinical and pathologic possibilities.

The history in most of the cases is very brief, because material was sent to the laboratory from practitioners, and sometimes no history was available.

CASE 1.—Complex, composite odontoma.

A 38-year-old white woman complained of recurrent headaches and pains in the cervical portion of the neck for one year. The patient had no other complaint. History and physical findings were essentially negative. Roentgenographic examination revealed an area of increased density about the size of a walnut in the upper third molar region. The mass was removed under local anesthesia. Recovery was uneventful. For one year the patient has been free of headache and pain in the neck.



Fig. 1.

Fig. 1 demonstrates the character of the tumor removed. It was a complex composite odontoma of the type frequently seen in this region. It was composed of irregular masses of dentine surrounded by masses of enamel, enamel matrix, and some cementum.

Tumors of this type are fairly common in this region. This one is of interest because of the referred character of the symptoms and the complete relief of symptoms following removal.

*Assistant Professor of Pathology, Schools of Dentistry and Medicine, University of Michigan.

CASE 2.—Globulomaxillary cyst.

Two weeks previous to his appearance in the clinic, the patient noticed swelling and pain on the right side of the midline of the maxilla. The area was incised and drained by his dentist. The swelling was relieved, but the discomfort continued. Roentgenographic examination revealed a cyst above the right lateral incisor and cuspid. This extended to the maxillary sinus. A similar cyst was seen above the left lateral incisor. At operation, it was found that the cyst on the right side communicated with the maxillary sinus. On the left side there was no sinus involvement.

The cystic lining removed was composed of a thin epithelium of a columnar type, which appeared in most areas to be two cells in thickness and represented an attenuated respiratory epithelium. The epithelium covered a dense connective tissue containing numerous inflammatory infiltrations. The inflammatory process was much more severe in the membrane removed from the right side, but in all other respects the cystic membranes were essentially alike.

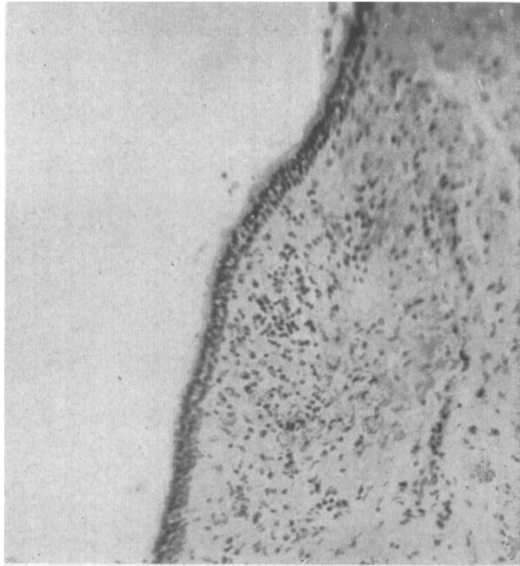


Fig. 2.

Fig. 2 shows the character of the cystic membrane removed from the left side.

These findings are characteristic of the globulomaxillary cysts, which are developmental inclusions at the line of union between the globular (median palatine) process and the maxillary process.

This case is reported to call our attention to the not infrequent bilateral character of this condition.

CASE 3.—Epuloid giant-cell tumor of the tongue.

A white man, aged 35 years, had a lesion on the right lateral aspect of the tongue of three months' duration. The lesion was pedunculated and not painful. Recently the lesion appeared to change in character and began to infiltrate the tongue. It produced some discomfort. Clinical examination revealed, at approximately the junction of the middle and posterior thirds of the tongue, on the right lateral aspect and extending to the dorsal and inferior surfaces, a firm, rubbery, ulcerated lesion, 3 cm. in diameter. There was no fixation of the tongue and the floor of the mouth was not invaded. All of the teeth on the right side were missing except the mandibular second molar which came in direct contact

with the lesion of the tongue. A biopsy of the lesion was reported "epuloid giant cell tumor of the tongue." Right hemiglossectomy was done.

The pathologic specimen revealed a lesion of the character and location described clinically. Microscopically, below the ulcerated surface was a marked fibroblastic and angio-



Fig. 3A.

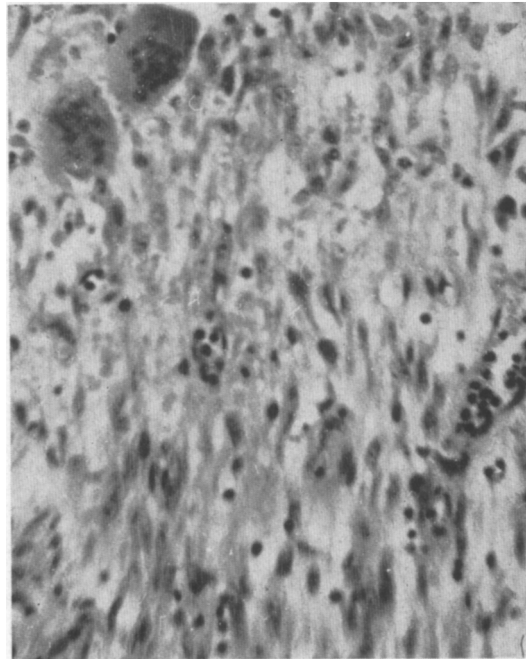


Fig. 3B.

blastic proliferation producing a very young immature connective tissue. The fibroblasts, although typical, were very large. The angioblastic proliferation formed many small, thin-walled, vascular channels. This zone of proliferation was very narrow and rapidly blended into an area which was characterized by the presence of numerous large multinucleated giant cells in an actively proliferating vascular stroma. The nuclei of the giant cells were of the same size and morphology as those of the proliferating fibroblasts, while the cytoplasm had a deeper staining quality. There appeared areas of transition from fibroblasts to giant cells. This resulted in a very cellular tissue which was invading and replacing the tongue muscle. In and around the areas of proliferation there was much old blood pigment in the form of hemosiderin. Many inflammatory cells of polymorphonuclear type were present beneath the area of ulceration.

Figs. 3A and 3B show the character of this pathologic change. It appears to be similar to the changes found in the giant-cell epulis associated with the jaws.

Treatment in this case was perhaps too radical, as this lesion is of local significance only. Recovery was uneventful and the patient has had no recurrence.



Fig. 4.

CASE 4.—Lymphangioma cysticum.

A white female infant, 17 months of age, was brought to the clinic because of an enlarged and bleeding tongue. The mother stated the child's tongue had always been large and had protruded from the mouth for the past seven months. Examination revealed a normal healthy child except for the enlarged protruding tongue which almost filled the oral opening. Its surface was dry and cracked and had some tendency to hemorrhage. There was a slight swelling of the submaxillary region. After injection with lipiodol, a multilocular cyst could be visualized in the floor of the mouth. A biopsy was taken, which showed the changes were those of a lymphangioma cysticum. By three separate surgical procedures, the tumors of the tongue and floor of the mouth were removed. The patient was discharged and appeared normal except for the small size of the tongue.

Congenital lymphangioma cysticum of the tongue, floor of the mouth and neck region, is not rare. When the tongue is involved extensively, there is very often an extension of the process into the neck region. This case is included because of the location of the dilated lymph spaces. Nearly all of the cystic areas were situated just beneath the epithelium. The entire process, although widespread, was located in the submucosa. There were large dilated lymphatic spaces in the connective tissue papillae which produced a thinning of the epithelium. In the deeper tissues, there were only a very few slightly dilated lymphatic spaces. This superficial terminal involvement is not the usual finding.

CASE 5.—Adenocarcinoma arising in mixed salivary gland tumor.

A 58-year-old white farmer presented himself with the chief complaints of swelling and discomfort in the floor of the mouth. The history revealed that a small nodule had been removed from the floor of the mouth by his dentist two years previously. The pathologic diagnosis was aberrant salivary gland. The mass then had recurred and gradually increased in size until there was a hard and movable mass present in the floor of the mouth on the left side, extending from the midline posteriorly for a distance of from 6 to 8 cm. Bimannual examination revealed a small egg-sized mass in the left side of the floor of the mouth. There was no fixation of the tongue. Physical examination was otherwise negative.

Microscopic sections of this tumor showed a marked variation in morphology. A large portion of the mass was composed of epithelial cells, which in some areas formed rather regular glandlike spaces of varying size. The cells were large with a vesicular nucleus and a rather intense-staining abundant pink cytoplasm. The boundary of the glandlike spaces was usually 2 to 3 cells in thickness. The spaces contained a mucinous substance. Near the capsule the pseudoglands were small and showed evidence of infiltration. In other areas, the epithelial cells were in small masses and cords. The masses were surrounded by a mucinous material. Where arranged in cords, the groups of cells resembled slightly the zona medullaris of the adrenal gland. Some of these characteristics are shown in Fig. 5A. In still another area the cells became very atypical and assumed a pleomorphic spindle shape. The cells were compactly arranged and had a slight amount of mucinous material associated with them. This character is shown in Fig. 5B.

This case is of interest for two reasons. First, it represents the rather rare pseudoadenomatous type of mixed salivary gland tumor so named because of the pseudoglands formed, and also because of its close resemblance to the pseudoadenomatous basal-cell carcinoma. Second, this neoplasm is of interest because of its malignant character, which is expressed by the ability to infiltrate, and marked atypical and cellular character in some areas. Adenocarcinoma arising in mixed salivary gland tumors are not a common finding.

CASE 6.—Papillary cystadenoma lymphomatosum.

Man, 51 years of age. "Has had this gland for 8 years—cervical gland." No further history was obtainable.

The mass grossly looked like enlarged lymph nodes. On section, they were found to be cystic, the cystic spaces filled with papillae. There was a small amount of serous fluid present. Throughout the papillae were whitish nodules.

Microscopically there was much lymphoid tissue present, arranged in branched papillae and masses with few germinal centers. The papillae were covered with a stratified or pseudostratified columnar epithelium. In some areas, it had a cuticular border. There was no evidence of mucin formation in the epithelial layer. Between the papillae was a coarsely granular albuminous precipitate containing a few leucocytes and desquamated cells. The remainder of the stroma consisted of lymphoid tissue, a delicate stroma, packed with lymphocytes, some showing germinal centers. The growth was typical and regular, stratified ciliated columnar epithelium upon a lymphadenoid submucosa, without any evidence of infiltration. This resembled very much the branchial cyst except for the character of the epithelium. (Fig. 6.)

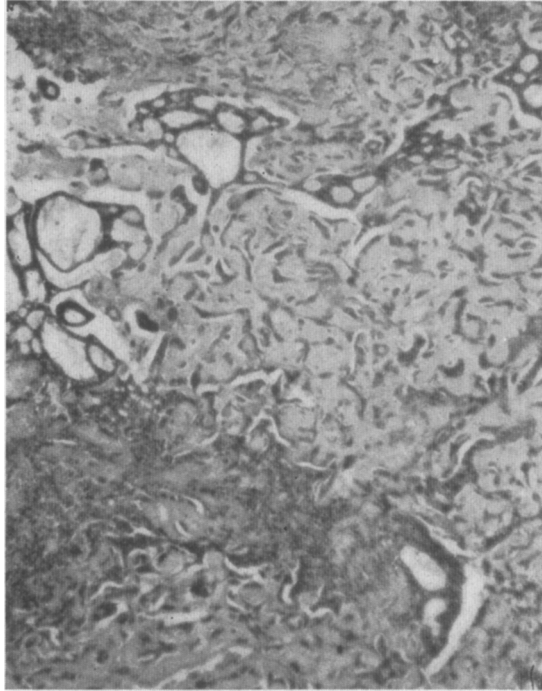


Fig. 5A.

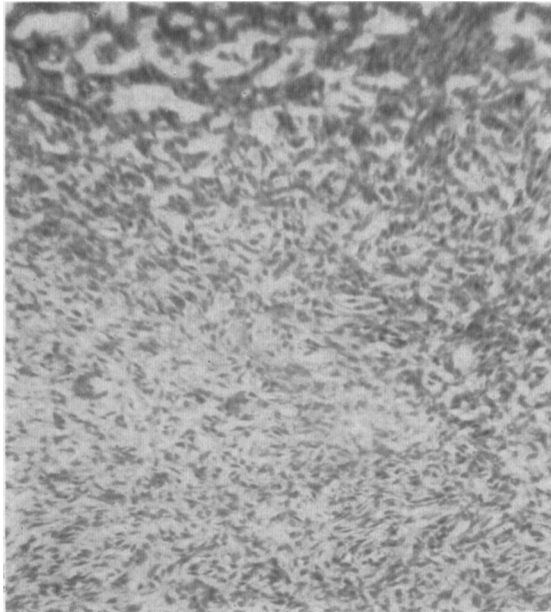


Fig. 5B.

This was a rare tumor found characteristically in the submaxillary and parotid area. Warthin¹ reported two cases in 1929. There were two out of several thousand tissues examined, of which over 700 were mixed salivary gland tumors, and over 500 were branchial cysts of the cervical region. In 1929 there were almost no reports in the literature. At present there are several reports, but they are not numerous.

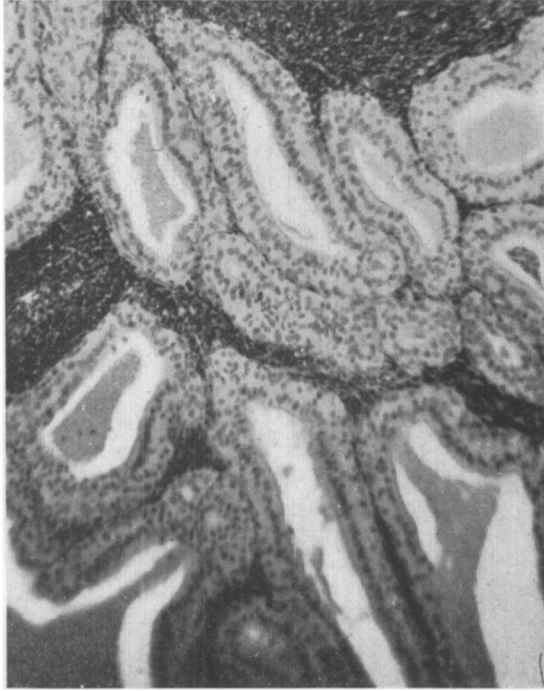


Fig. 6.

Warthin felt that these tumors were related to the branchial cleft cysts which are produced by heterotopia of pharyngeal mucosa. These, he suggested, were representative of heterotopia and dystopia of the eustachian tube mucosa to the parotid region. The term papillary cystadenoma lymphomatosum is used to describe this rare teratoid tumor.

CASE 7.—Tularemia.

“Enlarged submaxillary node.” No other history available.

The lymph node was enlarged as the result of a hyperplastic lymphadenitis. In a few areas there were granulomatous masses of varying size, as shown in Figs. 7A and 7B. The granulation tissue was composed of young fibroblasts and many large mononuclear cells. There was evidence of only slight angioblastic proliferation. The central portions of the granulomatous masses showed focal necrosis and suppuration. There was a marked accumulation of epithelioid cells bordering the focal area. No epithelioid giant cells were present. These changes present the picture of tularemia which is an unusual finding in the lymph nodes of this area.

CASE 8.—Nevus and neurofibroma.

A white man, aged 35 years, came to the clinic because of a dark purplish-brown, verrucous lesion present on the lower lip. He stated the lesion had been present since birth and had increased slowly in size. Recently, he had the impression it had been enlarging. The history and physical examination were essentially negative except for the presence of the lesion on the lip. This lesion began at the angle of the mouth on the right side and involved about two-thirds of the lower lip. It involved the full thickness of the lip and



Fig. 7A.



Fig. 7B.

extended down to the mucobuccal fold and up on the gingiva to the free margin. On the skin surface, it involved about one-half the depth of the chin. The lesion was of a soft consistency and a deep purplish-brown color. On the gingival mucosa below the central incisors were two dark brown to black spots, 2 mm. in diameter.

Biopsy of the lip and gingival tissue revealed the character of the neoplastic process. Beneath a hyperplastic mucosa there were individual and small groups of large polyhedral cells with a light, slightly blue-staining cytoplasm. The nuclei were light staining because of the finely divided chromatin. In a few areas, these cells had an associated pigment characteristic of melanin. They appeared as typical nevus cells. These nevus cells were in a delicate connective tissue stroma. Deeper in the tissue these nevus cells blended into a tissue composed of delicate hyperchromatic spindle cells which appeared to be of nerve origin. They produced areas characteristic of the neurofibroma. Fig. 8 shows both types of cells.

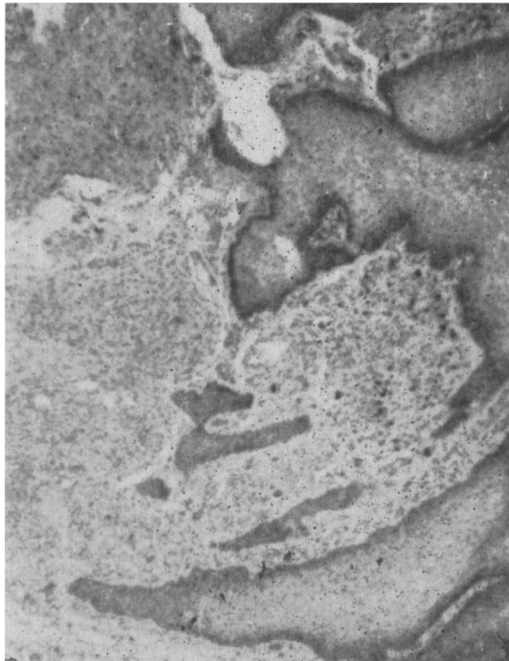


Fig. 8.

This case is of interest because of the frequent association between the nevus and the neurofibroma. It is also of interest because of involvement of the gingival mucosa, which is a rare finding.

The entire lesion was removed surgically. Recovery was uneventful and in four years the patient had shown no evidence of recurrence.

Because of the frequency with which cases are misdiagnosed from clinical findings alone, all cases should have a tissue examination. With information obtained from tissue sections, a more rational treatment can be outlined. From these few cases it can be seen that every case, regardless of its simplicity, may give us additional clinical and pathologic criteria upon which to base a diagnosis.

REFERENCE

1. Warthin, Aldred Scott: Papillary Cystadenoma Lymphomatosum, *J. Cancer Research* 13: 116, 1929.
2. Martin, H., and Ehrlich, H. E.: Papillary Cystadenoma Lymphomatosum (Warthin's Tumor) of Parotid Salivary Gland, *Surg., Gynec. & Obst.* 79: 611, 1944.