

**Salivary Heterotopia Of The Parathyroid Gland:
A Report of Two Cases and Review of the Literature**

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ABSTRACT

Two cases of periparathyroid salivary gland heterotopia are described. A review of the records of the Department of Pathology, Long Island Jewish Medical Center, over a four-year period, identified 759 surgical specimens containing parathyroid gland tissue. Of these, two (0.26%) contained foci of ectopic salivary gland tissue. Both cases were associated with cyst formation. To date, nine additional cases of heterotopic salivary gland tissue associated with the parathyroid gland have been described in the literature.

KEYWORDS

salivary gland, parathyroid, ectopia, heterotopia, hamartoma, choristoma

INTRODUCTION

Numerous malformations and minor developmental anomalies occur in the head and neck region. Different terms have been used to describe these lesions. A hamartoma is defined as a disorganized or excessive growth of normal tissue in an area where that tissue is commonly found. A choristoma is a disorganized focus of normal or fully differentiated tissue located at a site in which that tissue is not normally located. Designation of such entities as hamartomas or choristomas is most appropriate when they present as a distinct mass or lesion. In the head and neck, where a large number of different tissue types are located in close proximity to each other, this distinction can be difficult to make. Instead, the terms ectopia or heterotopia are commonly used.

Heterotopic salivary gland tissue has been described in a number of different anatomic locations¹. The most common site is the posterior lobe of the pituitary gland, where its presence is so common that it can be regarded as normal¹. Other frequent sites include the periparotid lymph nodes², the middle ear³ and the lower neck⁴ (in association with benign cervical lymphoepithelial cysts). Less

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common locations include the upper neck, the lingual mandible⁵, external auditory canal, cerebellopontine angle, thyroid gland, mediastinum, prostate gland, vulva and rectum⁶.

Numerous examples of salivary gland tumors, both benign⁷ and malignant⁸, have been documented as developing within heterotopic salivary gland tissue.

To date, nine cases of heterotrophic salivary glands of the parathyroid gland have been described in the English literature⁹⁻¹⁰. We describe two additional cases.

MATERIALS AND METHODS

The records of the Department of Pathology, Long Island Jewish Medical Center, were searched over a four-year period (01/01/1998 to 01/01/2002). All cases in which the final pathology report contained a reference to "parathyroid" were identified.

RESULTS

Of a total of 111,414 surgical accessions, 759 specimens containing parathyroid tissue were identified. Of these 759 cases, two (0.26%) contained foci of ectopic salivary gland tissue.

Case 1: A 30-year-old male underwent surgical removal of the right and left superior and inferior parathyroid glands for treatment of hyperparathyroidism. Histologic examination of the specimen demonstrated a focus of ectopic serous salivary gland tissue adjacent to an enlarged hypercellular right inferior parathyroid gland (Figs. 1a, 1b). Focal mild fibrous thickening of the capsule was noted. A cystic lesion, lined by a single layer of epithelial cells and filled with a PAS positive, diastase resistant, mucicarmine negative amorphous material was also identified. Focal areas of calcification and benign thymic tissue were also identified.

Case 2: A 60-year-old female with hyperparathyroidism underwent surgical removal of a right superior parathyroid gland adenoma composed of chief and oxyphil type cells. An adjacent enlarged superior mediastinal gland measuring 0.6 x 0.5 x 0.3 cm was identified at the time of surgery. On histologic examination, focal areas of normal appearing serous salivary gland tissue (Figs. 2a, 2b) were noted along with parathyroid and remnants of benign thymic tissue. Nearby

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cystic areas, lined by one to three layers of flattened squamous epithelium and filled with a basophilic, homogeneous, colloid-like material, were also identified.

DISCUSSION

Two cases of heterotopic salivary glands of the parathyroid gland area that occurred in relation to both the superior and inferior parathyroid glands are presented. This represented 0.26% of all parathyroid gland-containing surgical specimens accessioned at our institution over a four-year period. In both cases, cyst formation was noted adjacent to the heterotopic salivary gland tissue.

Youngs and Scofield⁹, reviewing cases of ectopic salivary gland tissue in the files of the Armed Forces Institute of Pathology, identified 2 cases that were associated with the parathyroid gland capsule. No mention was made of whether these occurred in the superior or inferior parathyroid glands. In the two “salivary heterotopia-cyst units” described by Youngs and Scofield⁹, there was no evidence of sialoadenitis or fibrosis in the salivary parenchyma, suggesting that the cystic areas were not related to obstruction of the salivary gland secretions.

Carney¹⁰ reported on seven cases of periparathyroid salivary heterotopia associated with cysts. Five occurred in the inferior parathyroid gland, one in the superior parathyroid gland. The specific location was not identified in one case. There was no evidence of sialoadenitis or fibrosis. In addition, Carney reported seven cases of periparathyroid cysts without evidence of associated heterotopic salivary tissue. The cyst contents were primarily mixtures of acidophilic and basophilic components. The acidophilic content was negative for mucin and thyroglobulin and was hypothesized to represent the end product of cell degeneration. The basophilic component was histochemically consistent with mucin.

Salivary gland determination appears to be regulated by elements in the extracellular matrix, fetal growth factors and an interaction between oral epithelium and its adjacent mesenchyme¹¹. The origin of heterotopic salivary-gland type tissue is unknown, but is likely of a developmental nature. The superior and inferior parathyroid glands develop from the fourth and third branchial arches

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respectively¹². Specifically, depressions on the inner aspect of the pharyngeal wall, called pharyngeal pouches, separate each of the branchial arches internally. During embryonic development, the third and fourth pouches expand into separate dorsal and ventral components, and their connection with the pharynx is lost. The dorsal components of the third and fourth pouches thus give rise to the inferior and superior parathyroid glands respectively. Youngs and Scofield⁹ suggested that the common association of heterotopic salivary tissue with cysts, sinuses and occasionally cartilage suggests a relationship to the branchial apparatus. In their view, anomalous differentiation of pluripotential cells within the wall of a branchial sinus results in the formation of ectopic salivary tissue. A number of cases of salivary heterotopia have been noted in patients with developmental defects of the second and, less commonly, the first branchial arches. Baerjee et al¹³ described the case of a 10 month old boy with Pierre-Robin syndrome in whom the lingual and palatine tonsils were almost completely replaced by ectopic salivary gland tissue¹³. Buckmiller et al¹⁴ reviewed 26 cases of salivary choristomas of the middle ear and suggested that this is a component of a syndrome consisting of unilateral hearing loss, abnormalities of the incus and/or stapes, anomalies of the facial nerve and other developmental defects of the branchial arches, such as preauricular pits.

Conversely, it has been proposed¹⁵ that heterotopic salivary gland elements form in relation to ectodermal thickenings called epibranchial placodes. These develop in relation to the underlying ganglia of cranial nerves VII, IX and X. The ectodermal lining of the placodal duct would have the potential for salivary differentiation.

It appears most likely that heterotopic salivary gland expression occurs secondary to developmental anomalies of the branchial arches during embryogenesis that results in displacement of branchial arch elements from their normal location. In the presence of an ectopic inductive influence from the ectomesenchyme, heterotopic salivary gland-type tissue develops.

CONCLUSION

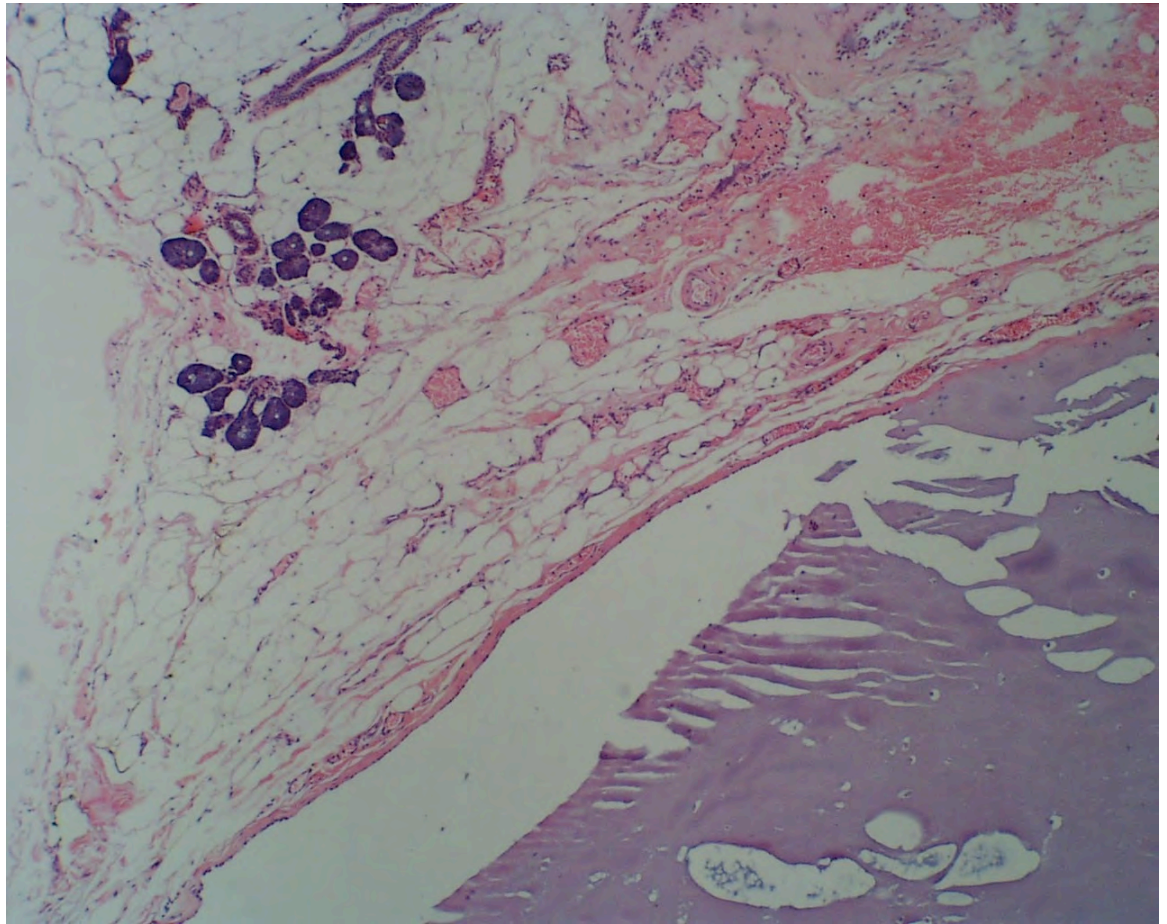
While heterotopic salivary gland tissue has been described in a number of different anatomic locations, its occurrence in relationship to the parathyroid glands is quite uncommon. We describe two cases of heterotopic salivary gland tissue of the parathyroid gland area. This represents only 0.26% of

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all parathyroid gland-containing surgical specimens accessioned at our institution over a four-year period.

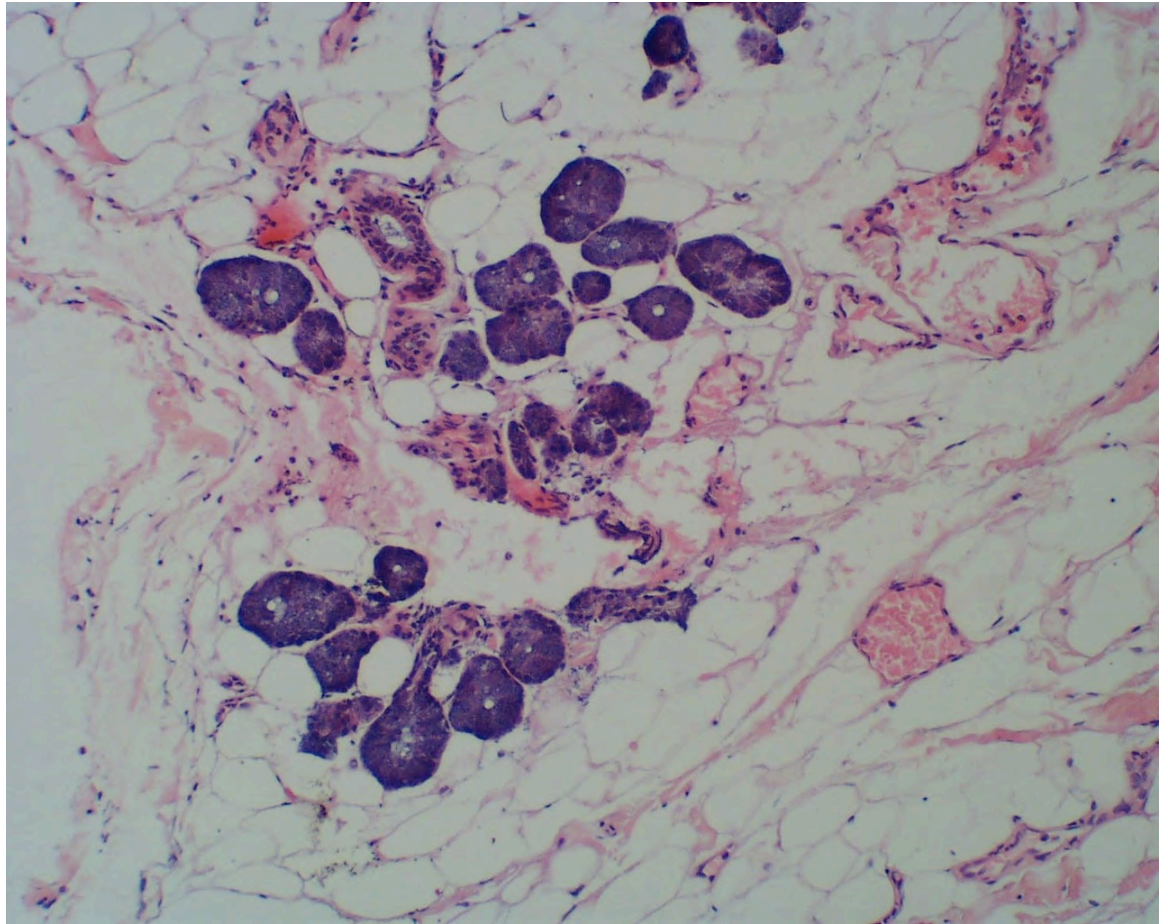
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Figure 1A. Low power view of ectopic salivary gland tissue adjacent to right inferior parathyroid gland. A cystic area is evident in the lower right (hematoxylin and eosin stain, original magnification 20x).



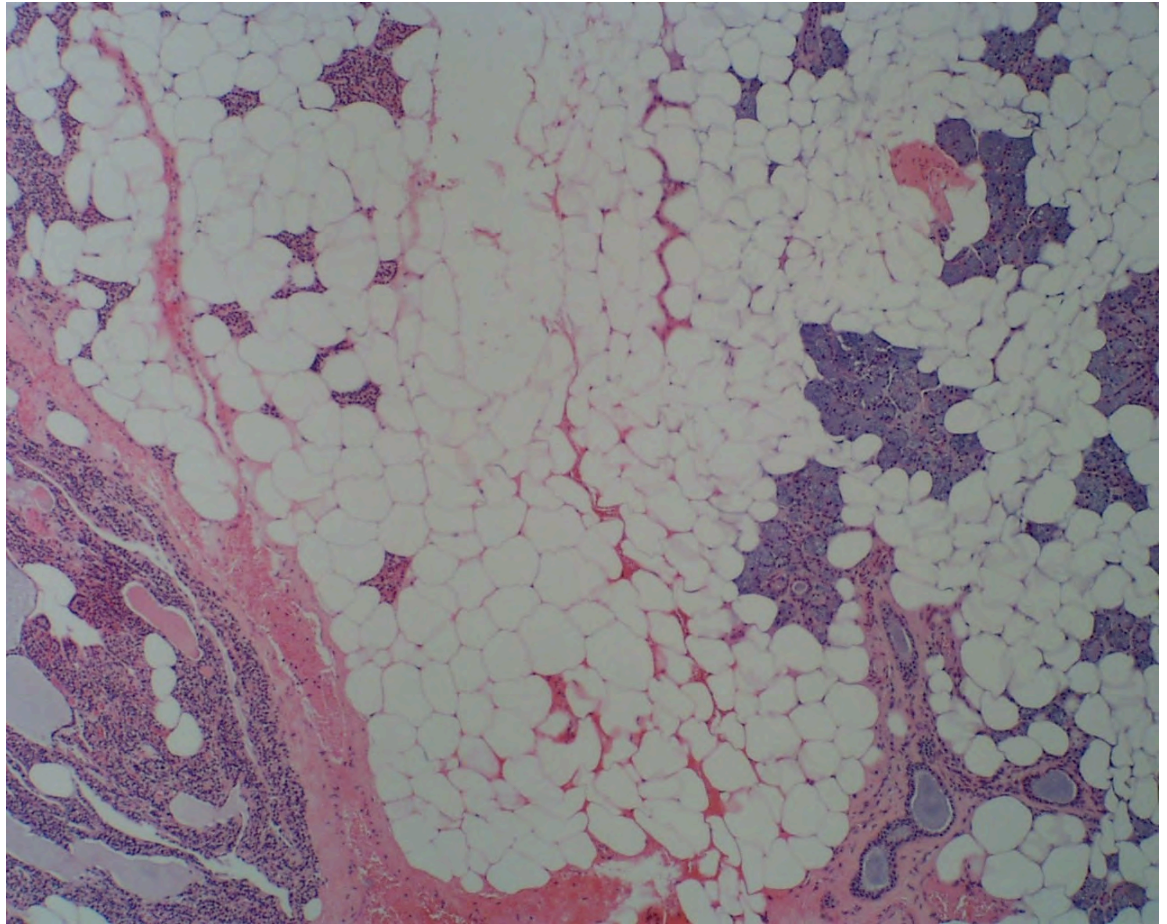
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Figure 1B: High power view of ectopic salivary acini and ducts adjacent to the right inferior parathyroid gland (hematoxylin and eosin stain, original magnification 40x).



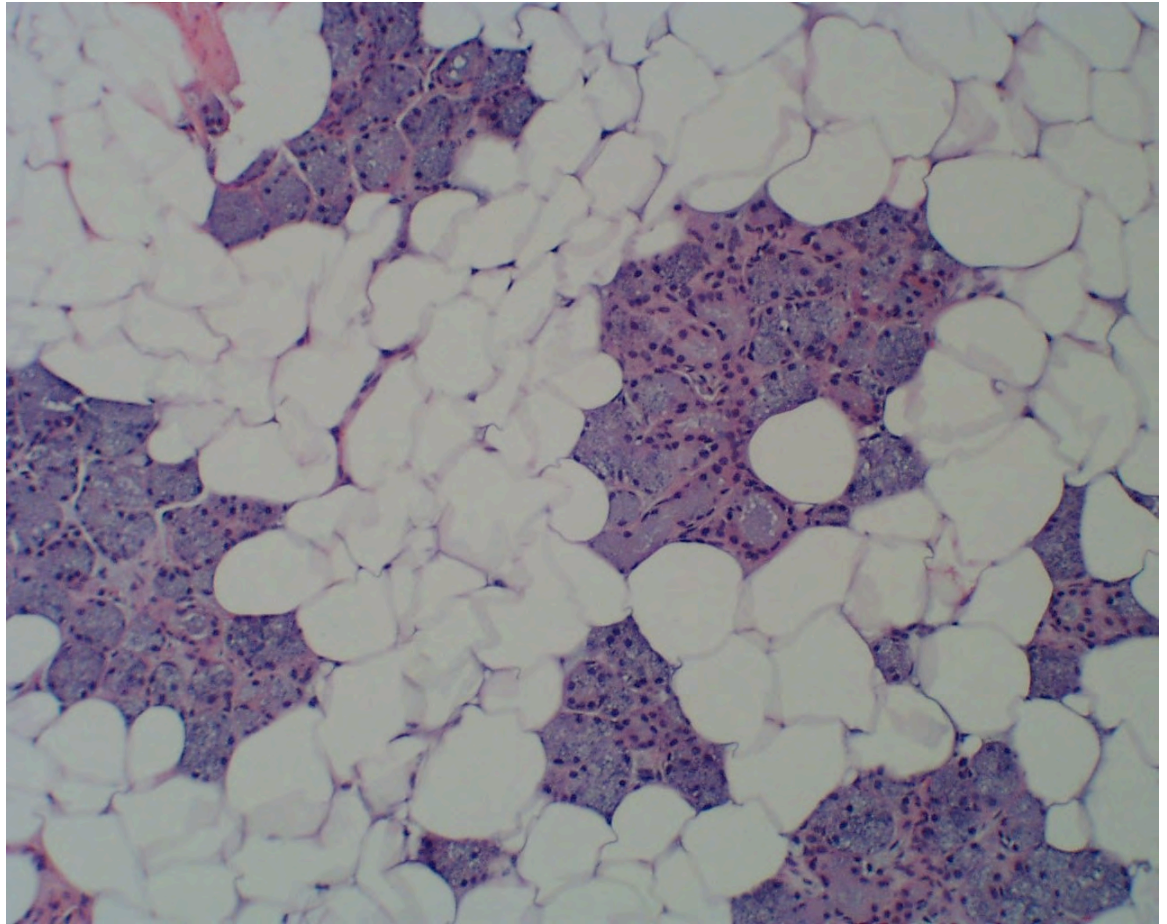
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Figure 2A: High power view of ectopic salivary gland tissue (right) adjacent to right superior parathyroid gland (hematoxylin and eosin stain, original magnification 40x).



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Figure 2B: Higher power view of ectopic salivary gland tissue adjacent to right superior parathyroid gland (hematoxylin and eosin stain, original magnification 80x).



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REFERENCES

- ¹ Warnock GR, Jensen JL, Kratochvil FJ. Developmental diseases. 1991: 10-25. In: Ellis GL, Auclair PL, Gnepp DR, eds. Surgical pathology of the salivary glands. Philadelphia, PA: WB Saunders Company.
- ² Luna MA, Tortoledo ME, Allen M. Salivary dermal analogue tumors arising in lymph nodes. Cancer 1987; 59: 1165-9.
- ³ Anderhuber W, Beham A, Walch C, Stammberger H. Choristoma of the middle ear. Euro Arch Otorhinolaryngol 1996; 253: 182-4.
- ⁴ Stingle WH, Priebe CR Jr. Ectopic salivary gland and sinus in the lower neck. Ann Oto Rhin Laryngol 1974; 83: 379-81.
- ⁵ Forrest WR. Ectopic salivary gland. Oral Surg Oral Med Oral Path 1974; 37: 653-4.
- ⁶ Denis J, Ganasinsia R, Puy-Montbrun T. Rare anorectal pathologic conditions. Current Opinion Gen Surg 1994: 103-7.
- ⁷ Ferlito A, Bertino G, Rinaldo A, Mannara GM, Devaney KO. A review of heterotopia and associated salivary gland neoplasms of the head and neck. J Laryng Otol 1999; 113: 299-303.
- ⁸ Curry B, Taylor CW, Fisher AW. Salivary gland heterotopia: a unique cerebellopontine angle tumor. Arch Path Lab Med 1982; 106: 35-8.
- ⁹ Youngs LA, Scofield HH. Heterotopic salivary gland tissue in the lower neck. Arch Pathol Lab Med 1967; 83: 550-6.
- ¹⁰ Carney JA. Salivary heterotopia, cysts, and the parathyroid gland: Branchial pouch derivatives and remnants. Am J Surg Pathol 2000; 24: 837-45.
- ¹¹ Denny PC, Ball WD, Redman RS. Salivary glands: A paradigm for diversity of gland development. Crit Rev Oral Biol Med 1997; 8: 51-75.
- ¹² Ten Cate AR. Oral histology: development, structure, and function. Fifth edition. 1998; 13-42. St. Louis MO: C.V. Mosby Company.

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A Report of Two Cases and Review of the Literature

- ¹³ Banerjee AR, Soames JV, Birchall JP, Reid C, Bray RJ. Ectopic salivary gland tissue in the palatine and lingual tonsil. *Int J Ped Otorhinolaryngology* 1993; 27: 159-62.
- ¹⁴ Buckmiller LM, Brodie HA, Doyle KJ, Nemzek W. Choristoma of the middle ear: a component of a new syndrome? *Otology and Neurotology* 2001; 22: 363-8.
- ¹⁵ Soucy P. Congenital cervical salivary fistula. *Can J Surg* 1985; 28: 130-1.