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# Retroauricular Pleomorphic Adenoma Arising from Heterotopic Salivary Gland Tissue

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**Summary:** A 38-year-old woman is described who presented with a slowly growing mass on the posterior aspect of the left ear. Excision and histopathologic evaluation revealed a pleomorphic adenoma (PA) originating from heterotopic salivary gland tissue. Many authors have presented cases of PAs originating from ceruminous glands in the external auditory canal or of so-called chondroid syringoma originating from apocrine and eccrine sweat glands. This is the only case in the recent literature of a PA originating from a heterotopic rest of salivary gland tissue in the retroauricular region. The 3 main sources of PAs, their embryologic derivation, and treatment are described. (*Plast Reconstr Surg Glob Open* 2016;4:e1028; doi: 10.1097/GOX.0000000000001028; Published online 23 September 2016.)

## CASE REPORT

A 38-year-old woman presented with a slowly growing mass on the posterior aspect of her left ear. The mass had been present for more than 10 years, increasing in size from about 1 to 20 mm in diameter over this time span. Physical examination revealed a slightly mobile, nonpainful soft mass with no draining sinuses. The patient underwent excision of the mass and reconstruction of the area under suitable intravenous sedation and local field block. The lesion was excised circumferentially down through skin and the subcutaneous tissue and was found to be densely adherent to the posterior aspect of the auricular cartilage. It was then carefully dissected and sent to pathology for permanent section.

Grossly, the specimen was a 1.5- × 1.1- × 0.6-cm piece of tan-pink firm tissue with attached yellow lobulated adipose tissue.

The resultant wound could not be closed primarily, so it was elected to do a posteriorly based advancement flap. Wide undermining to deep subcutaneous tissue adjacent to the mastoid fascia was then undertaken. The flap was advanced and back-cut in relaxed skin tension lines. The

flap was then inset with interrupted 4-0 Monocryl (Ethicon Inc.; Somerville, N.J., USA) deep dermal sutures. Epidermal coaptation was achieved with Dermabond (Ethicon Inc.; Somerville, N.J., USA). The procedure was well tolerated and the subsequent healing uneventful.

Histopathologic analysis of the mass showed a benign tumor associated with salivary gland tissue, consistent with parotid gland. The tumor had epithelial and myoepithelial cell differentiation, including focal tubule formation, chondromyxoid matrix, and aggregates of spindled myoepithelial cells. These features were diagnostic of pleomorphic adenoma (PA), a benign salivary gland tumor, and are shown in Figures 1–3. Continued follow-up was recommended.

## DISCUSSION

Heterotopic tissue of salivary gland origin, located most commonly in the head and neck region, occurs more often than historically believed.<sup>1</sup> The frequently benign nature of these anomalies causes few clinical symptoms. However, heterotopic salivary gland tissue can rarely give rise to neoplastic processes. As with orthotopic major salivary glands, neoplastic processes originating in heterotopic rests are predominantly benign. PAs are the most common benign tumors of the salivary glands, making up nearly 65% of all salivary gland neoplasms.<sup>2</sup> However, Warthin tumors have been found to be more prevalent than PAs in heterotopic salivary tissue.<sup>3</sup> The case presented in this article is an example of a PA originating from a heterotopic rest of salivary gland located on the posterior aspect of the ear. The rare location and histologic origin of this tumor make this case noteworthy.

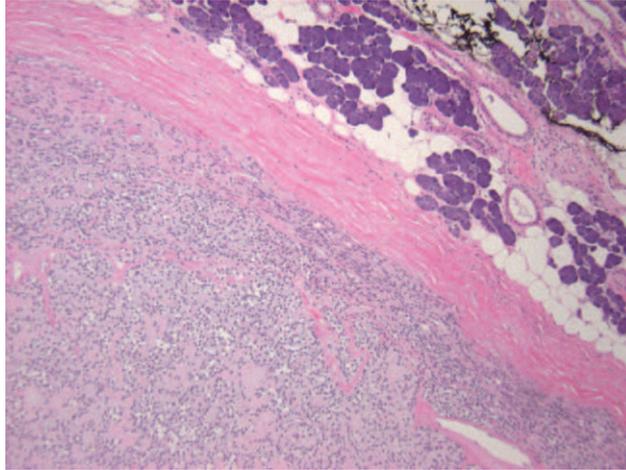
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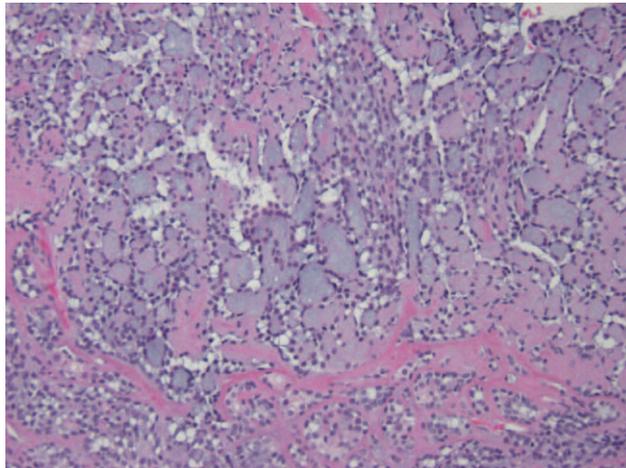
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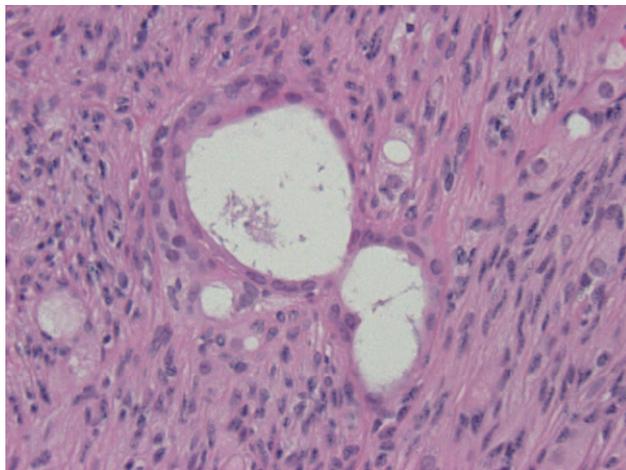
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**Fig. 1.** Biphasic tumor with stromal and epithelial components consistent with PA. Tumor arises from normal parotid gland tissue (upper right); H&E, 100x.



**Fig. 2.** Spindled to ovoid myoepithelial cells in a chondromyxoid and collagenous stroma; H&E, 200x.



**Fig. 3.** Cuboidal epithelial cells forming duct-like structures in a background of spindled myoepithelial cells. The dual cell population is characteristic of PA; H&E, 400x.

Heterotopic salivary gland tissue primarily arises in areas of the head and neck, including lymph nodes, the external auditory canal, hypophysis of the mandible, mastoid bone, middle ear, tongue, thyroid gland and in the upper and lower regions of the neck.<sup>1</sup> Three different mechanisms of the development of such tissue have been proposed. First is the development and persistence of abnormal vestigial tissues; second is the dislodgement of a part of the definitive organ during mass movement and embryologic development; and third is abnormal differentiation of the local tissue.<sup>1</sup>

The anatomically correct parotid gland originates as an epithelial bud at the angle of the mouth between the maxillary and mandibular prominences. A tube emerging in this region becomes the main parotid duct with its end proliferating into the glandular functional tissue. The parenchyma of the parotid gland is formed by splitting of the end of the parotid tube.<sup>4</sup>

PAs arise from 3 types of glandular tissue: salivary, ceruminous, and sweat glands. The classic presentation is a middle-aged woman with a benign neoplasm in the parotid gland. Less commonly, PAs are found in the other major or minor salivary glands. An even less frequent location is in extrasalivary tissue with myoepithelial cells, including apocrine, eccrine, ceruminous, and heterotopic salivary gland tissue. Thus, PAs have been previously described in the external auditory canal, skin, breast tissue, and vulva and accounts for half of lacrimal gland tumors.<sup>5</sup>

PAs originating in the external auditory canal are uniquely derived from ceruminous glands, the modified apocrine sweat gland of the ear canal.<sup>6</sup> Their histologic presentation is similar to that of PAs originating from salivary tissue as all are believed to be of myoepithelial origin.<sup>6</sup>

Chondroid syringoma is the term identifying PAs originating from apocrine or eccrine sweat glands. Chondroid syringomas, like PAs of the salivary and ceruminous glands, are benign. They are exceedingly rare with an incidence of less than 0.01% of primary skin tumors. They are most commonly localized to the head and neck region.<sup>7</sup> The histology of these tumors is similar to that of PAs originating from salivary tissue.

The case here presented is an example of a PA originating from heterotopic salivary gland tissue. This can be identified by the presence of histologically normal salivary glandular parenchyma surrounding the neoplasm, the location being separate from normal anatomical salivary tissue, the lack of ceruminous glands, and the absence of eccrine and apocrine sweat glands upon histologic evaluation.

Management of PAs for all 3 origins is wide excision with negative margins.<sup>2,7,8</sup> PAs can grow to extreme proportions if not excised. Malignant transformation occurs in 3% to 4% of tumors that are left untreated.<sup>5</sup> Recurrence occurs at a rate of up to 4% for PAs of salivary gland origin, with those most at risk for recurrence having irregular borders, increasing the likelihood of positive margins.<sup>5</sup> The average time range of recurrence is 7 to 10 years.<sup>5</sup>

## SUMMARY

We present a case of a 38-year-old woman with a retroauricular PA of heterotopic salivary gland origin, which to this date has rarely been reported. This case illustrates that PAs may present as nodular lesions of the skin in the head and neck region even if not proximal to the major salivary glands. Because of their ability to grow to extreme sizes and because of their malignant potential, recognition and treatment of PAs are critical.

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