PLEOMORPHIC ADENOMA OF THE PAROTID GLAND: A CASE REPORT

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Introduction

Pleomorphic adenoma, also known as benign mixed tumor, is the most common tumor of salivary gland origin, accounting for about 60-70% of all salivary gland tumors (Peel and Seethala, 2007; Downer, et al., 2011; Papadogeorgakis, et al., 2011). These tumors are most often found in the parotid gland (Oh and Eisele, 2006; Makeeff, et al., 2010), followed by the submandibular gland and the minor salivary glands (Oh and Eisele, 2006). It represents 70-80% of all tumors of the parotid gland. The lesion is usually solitary (Dubrulle and Souillard, 2006).

The typical presentation is that of a painless, slowly growing mass (Peel and Seethala, 2007). The gross appearance is smooth and lobular with a well-defined capsule (Oh and Eisele, 2006). Larger tumors often have a bosselated surface and may distend overlying skin and cause erosion of bone and remodeling deep to the tumor (Peel and Seethala, 2007). Microscopically, the tumor consists of epithelial and mesenchymal elements. The epithelial component forms a trabecular pattern with a mesenchymal stroma. The mesenchymal portion may be myxoid, chondroid, fibroid, or osteoid. The stroma varies from tumor to tumor and may have a combination of any of these tissue types within it. Histologically, pleomorphic adenomas show incomplete encapsulation with pseudopod extensions. Appropriate surgical therapy requires resection with an adequate margin of normal tissue surrounding the tumor (Oh and Eisele, 2006). While pleomorphic adenoma is a benign tumor, it has the capacity to recur and to undergo malignant transformation (Peel and Seethala, 2007).

This article presents a case of pleomorphic adenoma of the right parotid gland, with involvement of the homolateral neck region.
Case Presentation

A 20-year-old female was referred by her surgeon for diagnosis and treatment of "a lump in the right neck region" which was suspected as Nasopharyngeal Carcinoma (NPC). She was presented with a 5-year history of a slowly enlarging painless mass in her right preauricular and homolateral neck region. Her symptoms could not be explained by infection or trauma. The patient was a non-smoker and she didn’t have history of head and neck irradiation exposure. There was no other significant medical or surgical history or pertinent family history.

On clinical examination, the patient presented with single, well-defined, nontender mass of the right parotid and homolateral neck region (Fig. 1). Previous Fine Needle Aspiration Biopsy (FNAB) revealed metastatic carcinoma prone to NPC but nasoendoscopic examination did not show mass in the nasopharynx. No other anomalies of the head and neck were seen on clinical examinations. Facial nerve function was also normal. Preoperative assessment of the patient comprised of a neck Computed Tomography (CT) Scan and repeated FNAB. An enhanced axial CT scan showed inhomogeneous enlargement of the cervical lymph node in the right and homolateral neck regions. Radiologically, it revealed lymphoma or lymphadenopathy. But, there was no mass in the nasopharynx. Repeated FNAB was done selectively in the tumor, identifying it as pleomorphic adenoma. The patient was informed about the benign nature of the tumor and expressed her desire of preservation of the facial nerve.

Based on this diagnosis and the clinical characteristics and extension of the tumor, a submandibular incision was designed to gain access to the tumor. Surgical excision of the mass was performed (Fig. 2). After general anesthesia a horizontal incision of 5 cm allowed for exposure of the tumor mass. The lesion was encapsulated, and could be removed by blunt preparation. The wound was closed using non-resorbable sutures (Fig. 3). The surgical specimen was of a size of 8 x 5 x 3 cm (Fig. 4). Macroscopically, the tumor appeared to be encapsulated. When cut open, a solid tumor mass of a yellowish-pinkish color was obvious. The specimen was fixed in 95% ethanol, and sent to the pathology laboratory with the diagnosis of pleomorphic adenoma. No malignant transformation was identified in the specimen. Postoperative healing was uneventful with a satisfactory cosmetic outcome (Fig. 5). Facial nerve function revealed normal. Clinical follow-up until 4 weeks did not reveal any recurrence (Fig. 6).
**Discussion**

Pleomorphic adenoma is the most common tumor of salivary gland origin, accounting for about 60% of all salivary tumors in large series (Peel and Seethala, 2007). Up to 80% occur in...
the parotid gland (Peel and Seethala, 2007; Avecedo, et al., 2010). Bilateral occurrences of pleomorphic adenoma are rare. Pleomorphic adenomas can occur in any decade, but the mean age is 46 years with a slight female predilection. The typical presentation is that of a painless, slowly growing mass (Peel and Seethala, 2007). In the present case, a 20-year-old female was referred for diagnosis and treatment of "a lump in the right neck region" which was suspected as NPC. The patient was presented with a 5-year history of a slowly enlarging painless mass in her right preauricular and homolateral neck region. Her previous Fine Needle Aspiration Biopsy (FNAB) revealed metastatic carcinoma prone to NPC but nasoendoscopic examination did not show mass in the nasopharynx.

Actually, NPCs are epithelia neoplasms (Chan, Teo, and Johnson, 2004). NPC is the most common primary malignant neoplasm of the nasopharynx (Sivanandan and Fee Jr., 2005). The commonest presentation is a neck mass that is found on examination to represent a painless, unilateral, cervical lymphadenopathy. In almost all cases, the superior cervical nodes are the first group to be affected followed by enlargement of the mid and lower cervical nodes (van Hasselt and Leung, 1999; Chan, Teo, and Johnson, 2004). In parts of Asia, NPC is endemic with incidence rates of 15-50/100,000. NPC is the third commonest form of malignancy amongst men. The median age at presentation is 40-50 years. The incidence rises after the age of 20 and decreases after 60 years (Chan, Teo, and Johnson, 2004).

The imaging appearance of the pleomorphic adenoma deserves special attention. Pleomorphic adenomas frequently show gradual enhancement (Dubrulle and Souillard, 2006; Curtin, 2007). Not truly lobulated, the margin of many pleomorphic adenomas is slightly undulating or shows several slight bulges (Curtin, 2007). Due to an enhanced axial CT scan of the present case, the cervical lymph node in the right and homolateral neck region showed to be inhomogeneously enlarge. The margin of the tumor was not noted. Radiologically, it revealed lymphoma or lymphadenopathy. A delayed scan frequently will show the delayed enhancement typical of this tumor and demonstrate the margin. This has actually become more of a problem with the most modern CT scan technology. Many scanning protocols attempt to emphasize visualization of the arteries and veins and so the scan is done as the contrast is being injected and at that time the tumor may not be visible against the background of the normal parotid gland. The delayed enhancement phenomenon does not tend to be a problem with Magnetic Resonance Imaging (MRI). The multiple sequences used with MRI will almost always be sufficient to
visualize the tumor. Also, several sequences are done after the injection of intravenous contrast so there would be time for gradual enhancement to occur. Consistent reliable visualization of lesions is the primary reason that most head and neck radiologists prefer MRI to CT for investigation of salivary gland tumors (Curtin, 2007).

The gross appearance of the tumor is smooth and lobular with a well-defined capsule (Oh and Eisele, 2006). They usually range from 2 to 5 cm in greatest dimension (Peel and Seethala, 2007). Needle biopsy may have its greatest utility in the diagnosis of a submandibular mass, where it can help to distinguish neoplastic from more common inflammatory changes, which may spare the patient unnecessary surgery. Aspiration cytology may also differentiate a reactive lymph node adjacent to a salivary gland from a tumor within the gland itself (Spiro and Spiro, 2003). Histologically, pleomorphic adenomas show incomplete encapsulation with pseudopod extensions. This case shows that the surgical specimen was of a size of 8 x 5 x 3 cm (Fig. 4). The solid tumor mass of a yellowish-pinkish color was obvious. Due to the histological findings in the primary tumor, the tumor appeared to be encapsulated. No malignant transformation was identified in the specimen.

While pleomorphic adenoma is a benign tumor, it has the capacity to recur. Recurrence rates range from 0.8-6.8% in large series with long-term follow-up (Peel and Seethala, 2007). In the present case, the lesion was encapsulated, and could completely be removed by blunt preparation. Surgical management is the only recognized therapeutic alternative (Makeieff, et al., 2010). The two main aspects of parotid surgery are recurrence of the tumor and damage of the facial nerve (Papadogeorgakis, et al., 2011). Appropriate surgical therapy requires resection with an adequate margin of normal tissue surrounding the tumor (Oh and Eisele, 2006). It is now widely accepted that pleomorphic adenoma standard treatment must include at least partial superficial parotidectomy allowing tumor removal with a generous cuff (minimum 2 cm margin, except when abuts the facial nerve) of surrounding parotid tissue via en bloc resection (Makeieff, et al., 2010). Enucleation or pseudocapsular violation may lead to tumor spillage and localized tumor recurrence.

The factors that are thought to predispose to recurrence are poor surgical technique, violation of integrity of the capsule, multicentric origin of the tumor, and benign tumors that extend beyond the zygomatic arch (Papadogeorgakis, et al., 2011). While complete excision is required to ensure a favorable outcome, preservation of anatomic structures is also a major
concern. In this case, postoperative facial nerve function revealed normal and wound healing was uneventful with a satisfactory cosmetic outcome (Fig. 5). Since treatment of the relapsed tumor is complicated with a high rate of secondary recurrence, this will result in a lifetime follow-up of the patient who has to be informed on the possibility of further multiple recurrences (Peel and Seethala, 2007).

Rarely, a histologically benign pleomorphic adenoma can metastasize and behave like a low-grade malignancy. There are no features that can predict this rare occurrence. However, many of these tumors metastasized after at least one initial local recurrence, suggesting the possibility that altered anatomy secondary to surgery gave access to vascular and lymphatic channels. Thackray and Lucas have estimated that left unresected, approximately 25% of pleomorphic adenomas would eventually undergo carcinomatous change. These metastases seem to show a particular affinity for lungs and bone especially the vertebral column. The average age at diagnosis is 50–60 years (Peel and Seethala, 2007). Misdiagnosis is common because the residual PA component may be small, and because various carcinoma subtypes may be present (Zhao et al., 2013). Eneroth and Zetterberg undertook a microspectrophotometric DNA analysis of pleomorphic adenomas and demonstrated a difference in the DNA content between morphologically benign pleomorphic adenomas of short duration and those of long duration. This finding supports the hypothesis that the risk of carcinomatous transformation in a pleomorphic adenoma increases with the age of the tumor (Peel and Seethala, 2007). Therefore, follow-up of the patient is needed.

**Conclusion**

Pleomorphic adenoma is the most common tumor of the parotid gland. Consistent reliable visualization of lesions is the primary reason that most head and neck radiologists prefer MRI to CT for investigation. Needle biopsy may have its greatest utility in the diagnosis of the tumor. The two main aspects of parotid surgery are recurrence of the tumor and damage of the facial nerve. While pleomorphic adenoma is a benign tumor, it has the capacity to recur and to undergo malignant transformation.
References


