Canalicular adenoma of the parotid gland: A case report

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Abstract
Canalicular adenomas are uncommon benign salivary gland tumors commonly affecting the minor salivary glands. Usually, 90-95% of these are reported to occur in the upper lip, followed by the buccal mucosa and palate. Here, we report a case that involved the major salivary gland as a solitary discrete mass in the lower pole of the parotid gland. The mass was surgically excised and which histopathologically revealed normal parotid tissue along an extremely cellular area enveloped by a fibrous connective tissue capsule. Cellular areas showed cells with hyperchromatic nuclei, basally placed nucleus arranged in the form of interlacing cords, small islands, all separated by thin delicate connective septa all these features characteristic of canalicular adenoma.

Introduction
Canalicular adenoma is an uncommon benign neoplasm that can manifest in minor salivary gland ductal tissue throughout the oral cavity and infrequently in the parotid gland.[1] They comprise 1% of salivary gland neoplasms.[2] Most patients are over 60 years of age. Men and women are equally affected.[3] Rarely canalicular adenomas are known to occur in the major salivary glands. Complete surgical removal with a narrow margin of apparently normal tissue is recommended in the literature with excellent prognosis.[4]

Case Report
A 53-year-old female patient reported with a complaint of swelling below the right ear lobe since 1 year. The swelling had progressed gradually and attained a size of 1 × 2 cm. She gave a history of dull pain, while touching the concerned site. There was no evidence of facial palsy. On extra-oral examination, there was no gross facial asymmetry. Skin overlying the swelling was normal in appearance. No visible pulsation was noted. On palpation, a solitary, diffuse swelling in the right parotid region measuring about 1 × 2 cm in size could be felt. There was no local rise in temperature. Skin was pinchable. No paresthesia or anesthesia was noted in the concerned site. Swelling was diffuse, and it was soft to firm in consistency; mobile in all directions and mildly tender. Based on the history and clinical examination a provisional diagnosis of Benign tumor of right parotid gland was made. Ultrasonographic report showed the presence of calculus within the parotid gland [Figure 1]. Sialography was done which showed normal ductal architecture [Figure 2]. Fine-needle aspiration cytology was done and was reported as non-specific and non-contributory. Superficial lobe parotidectomy was planned, and pre-anesthetic evaluation was done and the patient was considered medically fit to undergo the procedure. Surgery was performed under general anaesthesia with preservation of the facial nerve. The specimen was sent for histopathological examination. Gross soft tissue specimen obtained was measuring 2 × 3 inches which were brownish in color and firm in consistency. Histopathologically hematoxylin and eosin and per-iodic acid Schiff stains were used, and the sections revealed normal parotid salivary gland tissue, with an extremely cellular area enveloped by a fibrous connective tissue capsule. Cellular area showed cells with hyperchromatic basally placed nucleus arranged in the form of interlacing cords, small islands, all separated by thin delicate connective septa [Figures 3 and 4]. Areas of duct formation...
Canalicular adenoma of the parotid gland

Puttaswamy, et al.

Canalicular adenoma of the parotid gland Puttaswamy, et al.

lined by double layered cells enclosed by PAS positive material. Certain foci of the cellular area showed hyalinization and hard tissue formation. The cellular area was surrounded by normal appearing salivary gland tissue. The specimen showed sufficient marginal clearance. With these findings, it was diagnosed as canalicular adenoma of the superficial lobe of the parotid gland. The surgical site healed uneventfully. However, post-surgery facial nerve palsy was noted on the right side. Physiotherapy and transcutaneous electrical nerve stimulation (TENS) therapy was advised. After 1 month’s duration of TENS therapy for thrice a week, improvement was noticed in the transient facial palsy. In the 3-year follow-up until date patient has no recurrence of the tumor.

Discussion

Canalicular adenoma is a benign salivary gland tumor that was defined by World Health Organization classification in 1991.\(^5\) Canalicular adenomas represent 1% of all salivary gland neoplasms and 4% of minor salivary gland neoplasms. The term “canalicular adenoma” was first coined by McFarland in 1942.\(^6\) In the past, there was difficulty differentiating benign salivary gland tumors because the classification schemes failed to separate canalicular adenoma from other monomorphic adenomas. The cause of canalicular adenoma is unknown. Even less is known about its multicentric tendency and its characteristic localization to the upper lip. Some authors have suggested that the multifocal nature of this tumor is related to “field cancerization” caused by an unknown,\(^7,8\) whereas others have suggested the multifocal dysplasia of developmental origin.\(^9\) There is increasing evidence that canalicular adenoma is derived from ductal luminal cells.\(^1\) Until 2005 only five cases of canalicular adenoma involving the parotid gland have been reported.\(^10\) Mair and Stalsberg have speculated that these tumors may have a very slow growth with a long latency period or that they may arrest early in development while they are still microscopic in size and clinically in apparent.\(^9\)

It appears that some adenomas progress to become clinically evident, perhaps if sufficient time elapses.\(^11\) In the present case, the patient had noticed a swelling in the region of the tail of the right parotid gland and the swelling had gradually increased over a period of 1 year; even though the swelling was 1 × 2 cm in size it had not caused a significant change in the facial symmetry. The patient reported only with localized dull aching pain while touching the affected site.

Recurrences of canalicular adenomas after surgical removal are rare but may occur many years after surgery, even in sites

Figure 1: Ultrasound scan showing calculus

Figure 3: PAS stain 4X magnification

Figure 2: Filling phase of sialography shows normal ductal architecture

Figure 4: Hematoxylin and eosin stain 20X magnification
remote from the primary tumor as a result of incomplete excision of the tumor or due to the presence of microscopic foci with a multicentric growth pattern. In the present case with 3 years of followup until date, the lesion has not recurred. This can be attributed to the encapsulation of the tumor that was resected in-toto with sufficient marginal clearance and absence of microscopic foci of tumor cells in the vicinity of the lesion.

To the best of our knowledge, there are no reports on malignant transformation of canalicular adenomas.

**Conclusion**

Canalicular adenomas are benign very slow growing tumor masses. Very rarely the major salivary glands are involved, and the parotid glands are most likely to be involved. We have reported a case in which the superficial lobe of the parotid was affected. Sialography failed to depict the characteristic picture of the tumor of the gland. Superficial parotidectomy specimen on histopathological examination showed the presence of canalicular adenoma.

**References**
