



Monomorphic adenoma of base of tongue- a rare case report & review of literature

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General Note



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ABSTRACT

Monomorphic adenoma is a rare benign salivary gland tumour of epithelial origin representing <2% of all tumours in general population and more than 80% of them arise in major salivary glands, mostly in the parotid gland and rarely in upper lip or buccal

mucosa. Monomorphic adenomas are defined as epithelial benign tumours of the salivary glands, which are not pleomorphic adenomas. Due to prognostic implications, differential diagnosis (adenocarcinoma, adenoid cystic carcinoma, basaloid squamous cell carcinoma, mucocele, sebaceous cyst, lipoma, nasolabial cyst, etc.) has to be considered before confirming the diagnosis. A rare case of a monomorphic adenoma of the minor salivary glands is situated at the base of the tongue in a 49-year-old male is reported. A review of the literature of minor salivary gland tumors and their location in the oral cavity has been discussed. Use of coblation technique made the surgery easier with minimal blood loss and minimal post-operative morbidity.

Keywords: Base of tongue, minor salivary gland, monomorphic adenoma, coblation

Abbreviations: SGT (Salivary Gland tumor)

1. INTRODUCTION

Salivary gland tumours (SGTs) usually manifest as an enlargement of the salivary glands. The classic presentation of a benign SGT is a painless, slow-growing mass on the face (parotid gland), angle of the jaw (parotid tail, submandibular gland), or neck (submandibular gland) or a swelling at the floor of the mouth (sublingual glands). Salivary gland tumours (SGTs) constitutes <1% of all tumours, and 3-5% of all head and neck tumours. Minor salivary gland tumours are infrequent and constitute 10-15% of all salivary gland neoplasms.^[1] They are located in the palate (50%), lips (15%), buccal mucosa (12%), and tongue (5%), floor of the mouth (5%) and rarely in pharynx, larynx & trachea.^{[2], [3]} Monomorphic adenoma is an uncommon benign epithelial tumour of the salivary gland which originates most commonly from one of the major salivary glands. It also occurs in minor salivary glands of the oral cavity.^[4] An overall ratio of approximately 1:6 for the benign/malignant lingual salivary gland tumours has been reported. In the region of tongue, the most common benign tumour is the pleomorphic adenoma where as adenoid cystic carcinoma is the most common malignant tumour.^[5] The palate is the most common site in the minor salivary glands.^[11] Because of the multiple and varied sites of minor salivary glands, the presentation of benign tumours in these sites may be less specific. For example, bleeding and airway compromise may be the first signs of minor SGTs of the nasal septum, whereas tumours of the base of tongue may present with dysphagia and a globus sensation.^[5] We report a rare case of monomorphic adenoma of the tongue base presenting with dysphagia.

2. CASE REPORT

A 45-year-old male presented to our ENT outpatient department with chief complaints of dysphagia and globus sensation for six months. There was no history of fever, throat pain or oral bleeding during deglutition. The patient had an addiction of tobacco smoking with 20 cigarettes per day for past 20 years. Past and family history was uneventful. General and systemic examination was normal. Cervical lymph nodes were not palpable. Indirect laryngoscopy revealed a solitary sessile round shaped mass with a smooth surface of size 4 × 3 cm approximately which was present over the base of the tongue on left side. The mass was firm on palpation, incompressible and non-tender. 70 degree rigid endoscopic examination was done to confirm the above findings. Laryngeal inlet and pyriform fossa was normal. The mass was occupying the left half of vallecula. Right side vallecula was free (Fig.1). There were no visible pulsations or signs of inflammation seen over the surface of the mass.



Figure 1 Base of tongue swelling

Routine investigations and Ultrasonography (USG) scan neck were normal. Fine needle aspiration cytology (FNAC) was done and it was reported as a benign salivary gland lesion. CT scan (coronal & axial cuts) was done which revealed a contrast-enhancing mass of size $5 \times 3 \times 4$ cm approximately of heterogeneous density over the base of the tongue and a reduction in Anterior-posterior diameter of hypopharyngeal airway at the level of base of tongue (Fig. 2). There was no extension to the larynx and level I or level II cervical lymph nodes.

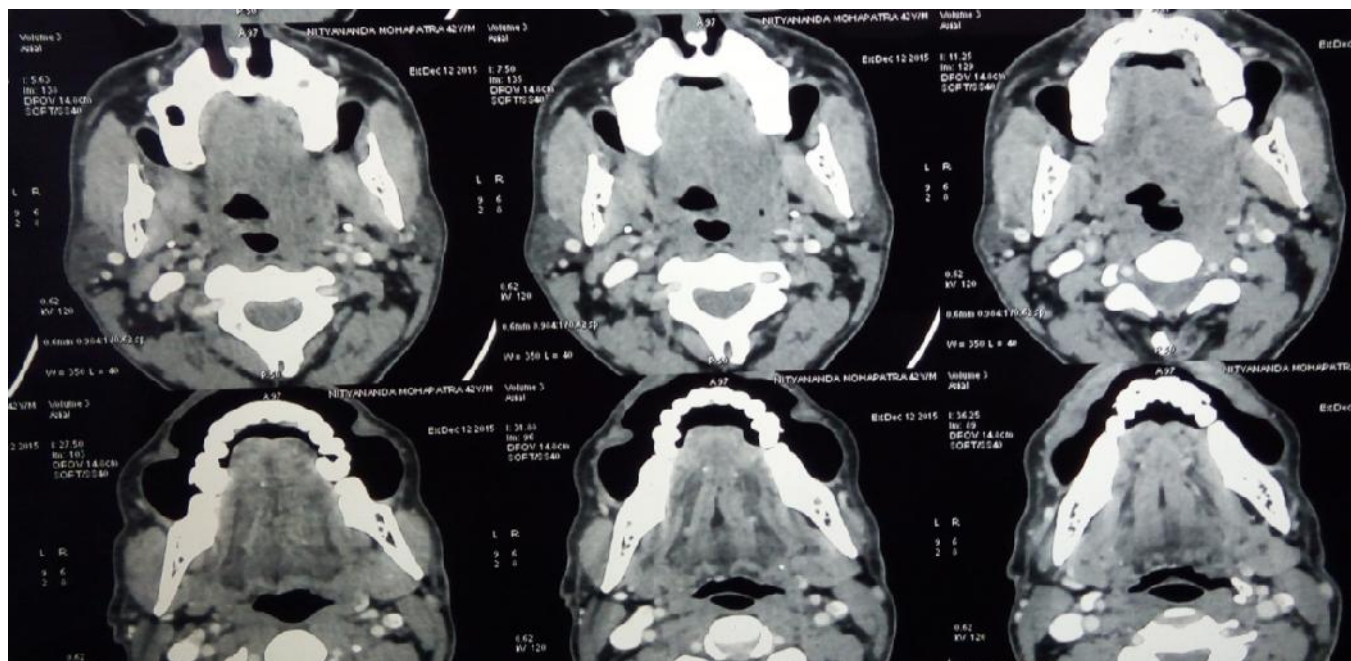


Figure 2 CT scan (Axial view) showing the extent of the tumour and narrowing of Anterior-posterior diameter

Under general anaesthesia, the patient was intubated transnasally under flexible bronchoscopic (FOB) guidance successfully. After giving adequate airway protection by soaked ribbon gauge, complete excision of the mass was done via intraoral approach with 70 degree rigid endoscopic guidance by coblation technique (using Evac 70 wand) with minimal bleeding (Fig. 3 & 4).

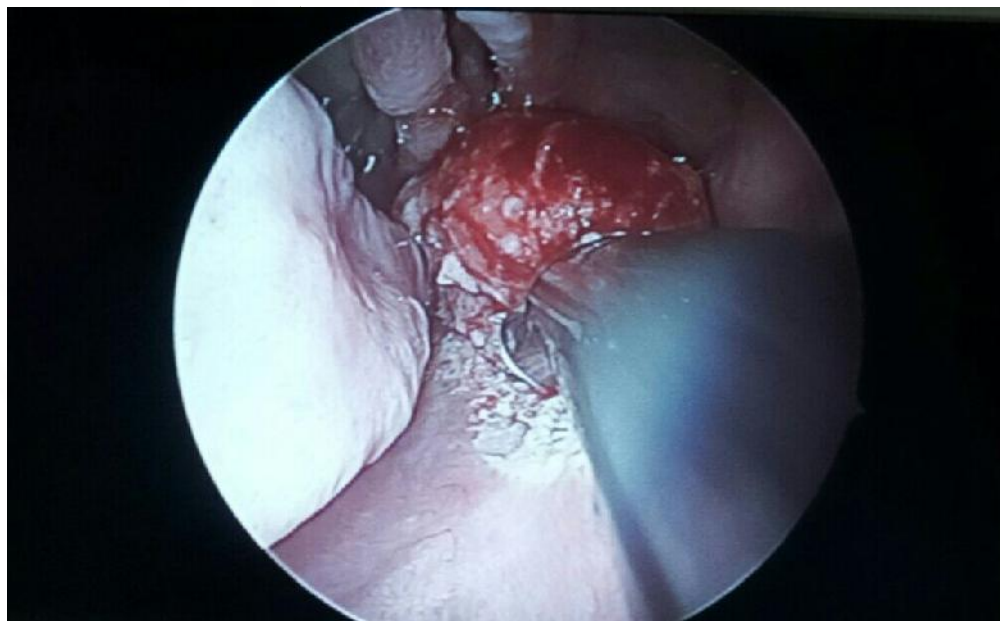


Figure 3 Excision of the base of tongue mass using coblation technique

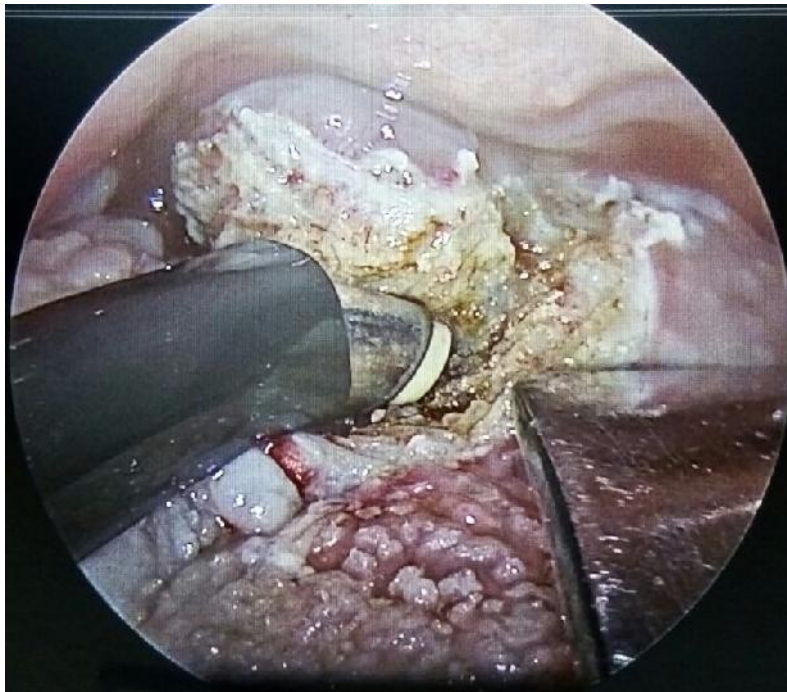


Figure 4 Intraoperative coblation

The excised mass was a well circumscribed one and was sent for biopsy. The excised area was raw & there was no bleeding as such, and was left raw to allow healing by secondary intention. Post-operative period was uneventful (Fig 5). Patient was extubated without any difficulty and Ryles tube was placed intra-operatively for feeding. Hence, tracheostomy was avoided. However the patient was kept overnight in semi-ICU under keen observation. Patient was shifted from semi-ICU to ward with all vitals within normal limits. Ryles tube feeding was started and input-output chart was maintained. Oral feeding was then started on 5th post-operative day after ryles tube removal. Oral rinsing with antiseptic gargle was done regularly. Histopathological examination of the tissue showed uniform basaloid cells arranged in a trabecular pattern suggestive of monomorphic and ductal differentiation and confirmed the diagnosis of monomorphic adenoma with clear margins (Fig 6). The postoperative course was uneventful. The patient has been asymptomatic and no signs of recurrence were present during the six months follow-up period.



Figure 5 post-operative view

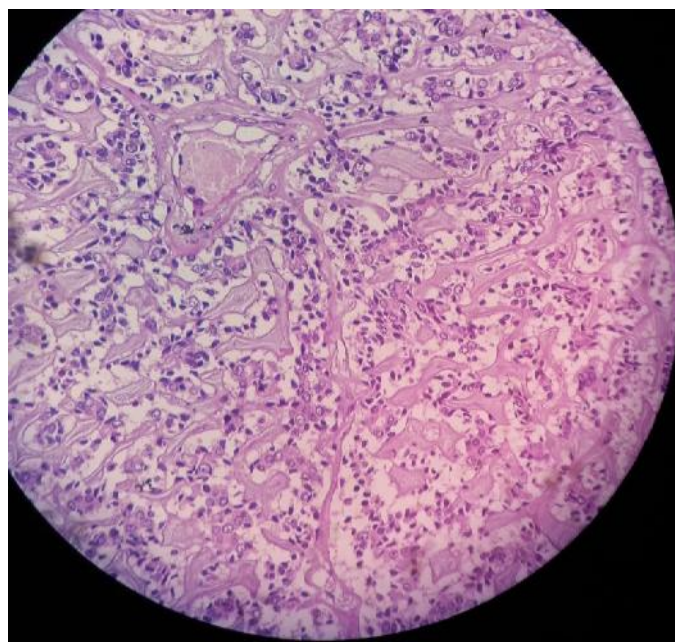


Figure 6 Histopathological study (H x E, 280) showing basaloid cells in trabecular pattern

3. DISCUSSION

Tumours of the salivary glands are classified based on their cytological, architectural, and biological characteristics. The World Health Organization (WHO) classification of 1992 groups classifies both benign and malignant tumours of salivary glands into epithelial and nonepithelial categories. Benign epithelial tumours include pleomorphic adenoma, Warthin tumour, monomorphic adenoma, intraductal papilloma (IDP), oncocytoma, and sebaceous origin neoplasms. Benign nonepithelial tumours (mesenchymal origin) include hemangioma, angioma, lymphangioma (cystic hygroma), lipoma, and neural sheath tumours. An additional category of tumour-like lesions includes necrotizing sialometaplasia, benign lymphoepithelial lesions, cystic lymphoid hyperplasia (in persons with AIDS), and salivary gland cysts. ^[7] Monomorphic adenoma is a rare benign tumour of minor salivary glands.

Classifications of salivary gland monomorphic adenomas use a variety of principles, including the type of cell, cellular arrangement, and embryologic development, and as a result, could represent a heterogeneous group of tumours. There is a considerable confusion in various literatures concerning the terms monomorphic adenoma, basal cell adenoma, and canalicul adenoma. It is recommended that (1) the term “monomorphic adenoma” is to be used for all benign epithelial salivary gland tumours that are not pleomorphic adenomas, (2) that “basal cell adenoma” is to be used to identify a specific entity that is one component of the monomorphic adenoma group and exhibits a number of histologic subtypes, and (3) that “canalicul adenoma” be used to describe another entity, distinct from basal cell adenomas which also belongs to the monomorphic adenoma group with larger, paler cells in central areas and composed purely of tubular cells and is more common in oral cavity. The basal cell adenoma was first named by *Kleinsasser & Klein* in 1967. ^[8] Histologically four characteristic patterns of monomorphic adenoma have been described: Solid, Trabecular, Tubular and Membranous. ^[4] The most common is the solid type, followed by the trabecular and trabecular-tubular types, the membranous and the rare tubular types. All have a fibrous stroma and lack a myxochondroid matrix as in pleomorphic adenoma. The trabecular-tubular variant, however, exhibits subtle stromal spindled cellularity that has been shown to be of myoepithelial origin, underscoring the relationship to pleomorphic adenoma. ^[9, 10, 11] According to Batsakis and Brannon ^[16] histological classification of monomorphic adenoma is as follows:

1. Tumours of terminal duct origin: Basal cell adenoma, canalicul adenoma.
2. Tumours of terminal or striated duct origin: Sebaceous adenoma, sebaceous lymph adenoma.
3. Tumours of striated duct origin: Oncocytoma, papillary cystadenoma lymphomatosum.
4. Tumours of excretory duct origin: Sialadenoma papilloform or inverted ductal papilloma.

Monomorphic adenomas are benign salivary gland tumours that have a predilection for development in the upper lip, palate and parotid gland but rarely may involve base of tongue as in the present case. According to study reviewed by Chaudhary *et al.* ^[15], the base of the tongue was the site of origin in only 1% of all intraoral minor salivary gland tumours. Typically, patients are between fifth and seventh (mean age 60 years), but an age range of 29 to 87 years has been reported in the literature. ^[1] Males are affected more commonly than females. Clinically monomorphic adenomas present as a painless mass, usually superficial within the glandular body, and a brownish appearance is usually observed. It is a slow growing encapsulated tumor, firm in consistency, mobile painless mass usually of size less than 4cm in major diameter. ^[12] Most patients are not aware of the tumours until they develop difficulty in swallowing. They are detected incidentally on routine physical examinations. Monomorphic adenoma may resemble clinically as a mucocele of the oral mucosa. In general, the latter usually appears in the lower lip of young people, whereas the former usually appears in the upper lip / tongue of the elderly. ^[13] Histopathologically they may resemble ameloblastoma-like pattern. ^[14] The cells of origin are myoepithelial and intercalated duct cells. The minor salivary glands consist of 600-1000 small independent glands found across the oral cavity, palatine tonsils, pharynx, and larynx. ^[10] The salivary gland neoplasms are rare and represent a variable group of benign and malignant tumours with different behavioural characteristics. The concept of salivary gland tumours was described by BERTRANDI in 1802. ^[10] The etiological factors for the salivary gland neoplasms is not well-defined, but some association has been found with radiation therapy, ^[10]Smoking, ^[12]Epstein-Barr virus infection, ^[13]Genetic predisposition, ^[10]Sunlight and Vitamin A deficiency, Chemotherapy.

The diagnosis is made on the basis of histopathological study. Incisional biopsy gives a better chance of diagnosis than fine needle aspiration. Differential diagnosis includes tuberculoma, neurilemmoma, amyloid tumour, mucoepidermoid carcinoma, adenocarcinoma, adenoid cystic carcinoma, basaloid squamous cell carcinoma, mucocele and angioleiomyoma. ^[15] The different surgical approaches are transoral resection, midline transhyoid approach and lateral pharyngotomy. ^[18] Monomorphic adenomas of the minor salivary glands are best treated by wide surgical excision with a margin of normal tissue, the extent of which will vary depending on the tumour size, location, appearance and histology. In the present case, the tumour was resected transorally by coblation technique as the tumour was well defined and smaller in size.

Coblation is a plasma-mediated tissue ablation technique. It involves passing a bipolar radiofrequency current through isotonic saline to convert it into an ionized plasma layer. ^[18] This layer effectively disrupts intercellular molecular bonds in the tissues resulting in a vaporization effect. ^[19] Surface irrigation and suction are applied to prevent significant pooling of saline inside the oral cavity. ^[20] Coblation generates a substantially lower thermal effect compared to electrocautery and CO₂ Laser, estimated between 45°C - 85°C, with a subsequent presumption of diminished collateral thermal damage to surrounding tissues. ^[21] No point during the procedure of the coblative energy is introduced into the body as the process is focused only on the electrodes. Minimal tissue penetration and less surrounding tissue damage were proposed the advantages of the device. The handpiece or "wand" that creates the plasma field also aspirates the dissolved tissue. Ablation proceeds from a superficial to a deep plane, destroying the mucosal surface as the wand is applied directly to the tissue. CO₂ Lasers can also be used for removal of oral lesions like monomorphic adenoma. Although laser beams have an advantage of easy access to the anatomical site, inherent hemostatic properties, minimal postoperative pain but it has certain disadvantages like airway fire, risk of blindness, oral and facial burns, and risk of inhalation of plume of vaporised tissue and requires proper training to handle lasers. ^[14,19]

Post-surgery it has a favourable prognosis. Irradiation is reserved for recurrences and inoperable cases. Patients with nonresectable tumours should receive radiation therapy although salivary gland tumors, as a group, are not highly radiosensitive. Chemotherapy is never curative, and its use as an adjunctive has not yet been established. Recurrence is uncommon and may be due to partial excision or a multifocal origin of the tumour. Periodic follow-up pattern is important to look for any relapse or any malignant transformation. ^[22] In the present case report, the patient was followed up at regular intervals to assess the postoperative healing and recurrence.

5. CONCLUSION

A significant number of minor salivary gland tumours are asymptomatic and are picked up on routine dental or oral cavity examination. Monomorphic adenoma is an important topic in otolaryngology because of their rare occurrence, improper diagnosis, management, and unpredictable clinical course. The minor salivary glands were the second common site of involvement, engaged in 28.5% of all tumors. ^[14] Therefore, treatment should always aim at complete excision of the tumor; allow secondary healing of the lesion and with a long-term follow-up to detect recurrences in a prompt time.

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