

A Rare Case of Pleomorphic Adenoma in the Left Submandibular Salivary Gland

J MANIGANDAN¹, A FAROOK², R NATARAJAN³

ABSTRACT

Pleomorphic adenomas, which are benign tumours of the salivary glands, are most commonly found in the parotid gland and comparatively infrequently seen in the submandibular gland. These benign mixed tumours, known as pleomorphic tumours, are composed of different types of myoepithelium, epithelium, and mesenchyme. Regardless of the histologic type, parotid gland neoplasms result in distinct swellings in front of and below the ear. With the exception of neglected malignant tumours, both benign and malignant lesions are normally palpable when they are first discovered. Malignancies are often discovered more quickly due to their rapid growth, even though benign tumours are known to exist for many months to years before gaining clinical attention. Ultimately, there are no reliable clinical criteria to distinguish between benign and malignant lesions. A 58-year-old female patient presented to the general surgery outpatient department with a six-month history of a firm, palpable, slow-growing, painless, freely mobile mass in the submandibular region that was not attached to the surrounding tissue, bone, or skin. There was no significant past history, and the patient was not on any medication. Axial and coronal views of the Computed Tomography (CT) scan showed a well-defined radiolucent mass in the medial aspect of the left submandibular gland. The patient underwent surgical exploration and excision of the mass under general anaesthesia, and the excised specimen was sent for histopathological examination. The entire gland was dissected freely and removed, with utmost care taken not to affect the nearby neurovascular structures. No enlarged lymph nodes were noted. The histopathological examination of the biopsy specimen revealed features of pleomorphic adenoma. Surgery is the treatment of choice for pleomorphic adenoma, and the tumour tissue must be completely removed because any residual tumour tissue has a high risk of recurrence or transformation into a malignant tumour. The patient was followed up for more than two years with no signs of recurrence.

Keywords: Mixed salivary Gland tumour, Recurrence, Surgery

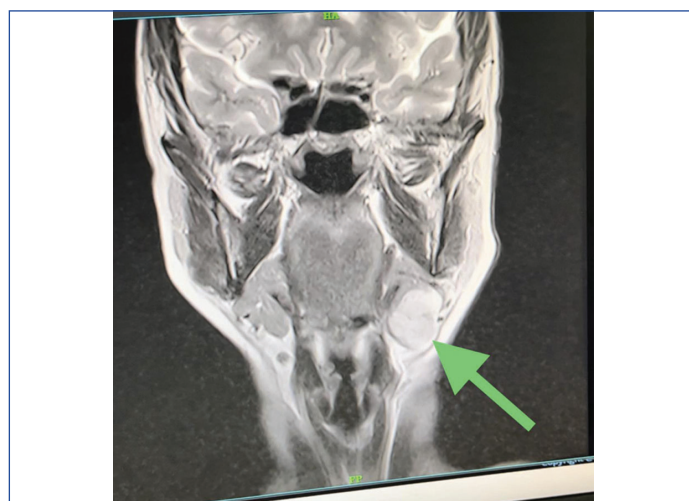
CASE REPORT

A 58-year-old female was referred to the general surgery outpatient department with a history of a firm palpable mass in the left submandibular region [Table/Fig-1]. The swelling was noticed six months ago and had an insidious onset, slow growth, and was painless in nature. There was no relevant personal or medical history. On palpation, the mass was firm and non-tender. The mass was oval in shape and measured 5×3 cm clinically. It was freely movable, with superior movement restricted by the left ramus of the mandible bone, and not attached to the surrounding tissue or skin. Bimanual palpation was negative, and there were no neuronal or functional disturbances in the adjacent tissues. No diminished salivation was found during an intraoral examination. A provisional diagnosis of a left submandibular gland tumour was made based on the examination and history. To determine the severity of the lesion, a CT scan was suggested to the patient.



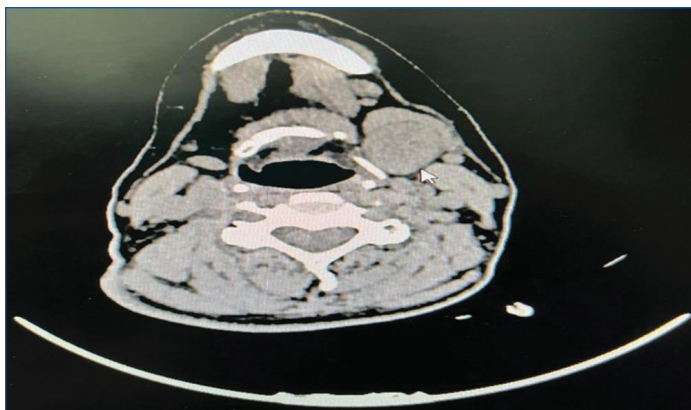
[Table/Fig-1]: Swelling in the submandibular region.

Axial and coronal views of the CT scan showed a well-defined radiolucent mass in the medial aspect of the left mandibular body, covering its entire extent and extending inferiorly into the submandibular region [Table/Fig-2,3].

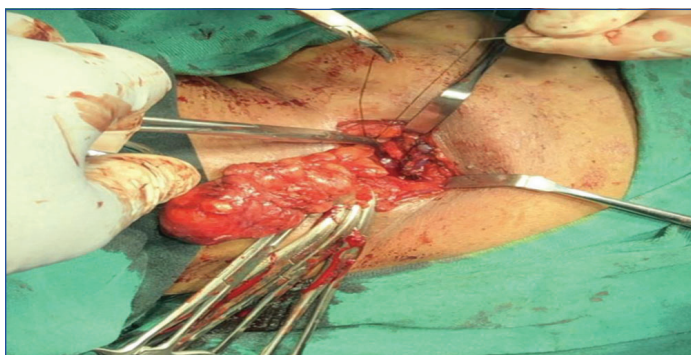


[Table/Fig-2]: Coronal MRI scan showing enlarged left submandibular salivary gland with well differentiated borders.

Under general anaesthesia, the patient underwent surgical exploration and excisional biopsy of the left submandibular gland mass. With a standard submandibular incision, the submandibular gland and the mass were carefully dissected, excised, and sent for histopathological examination [Table/Fig-4,5]. The intraoperative findings revealed that the superficial lobe of the left submandibular gland was diffusely enlarged, measuring 6×2×3 cm, without infiltrating the adjacent structures. The entire gland was dissected freely and removed. No enlarged lymph nodes were noted.



[Table/Fig-3]: Axial CT scan showing the enlarged left submandibular salivary gland.



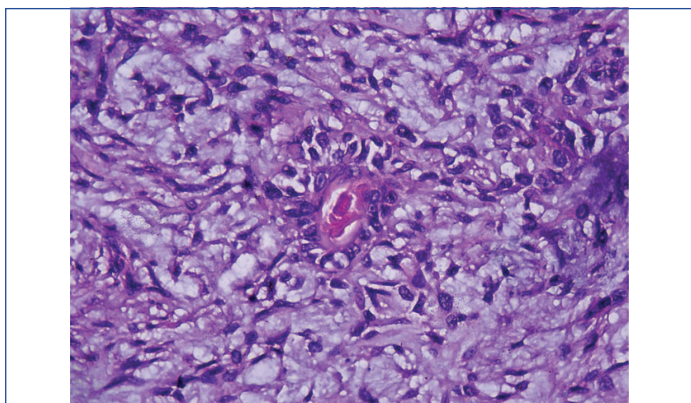
[Table/Fig-4]: Intraoperative photograph showing the careful dissection of submandibular salivary gland from adjacent vital structures.



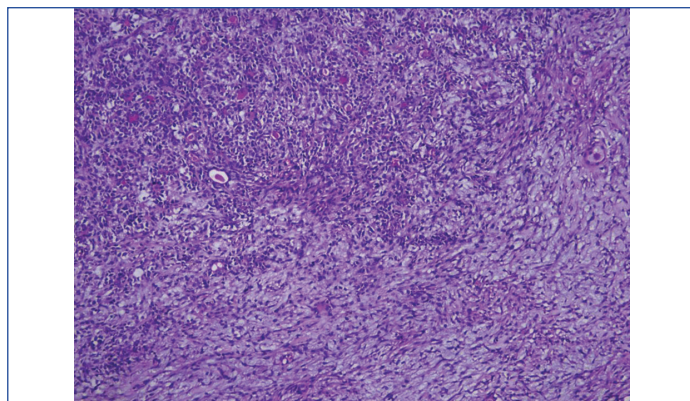
[Table/Fig-5]: Excised tumour- gross specimen (6x2x3 cm).

The primary closure was done in a layer-wise manner without attaching a drain. The histological examination confirmed pleomorphic adenoma as the histopathologic sections revealed darkly stained tumour cells lying in a predominantly mesenchyme-like background [Table/Fig-6-8].

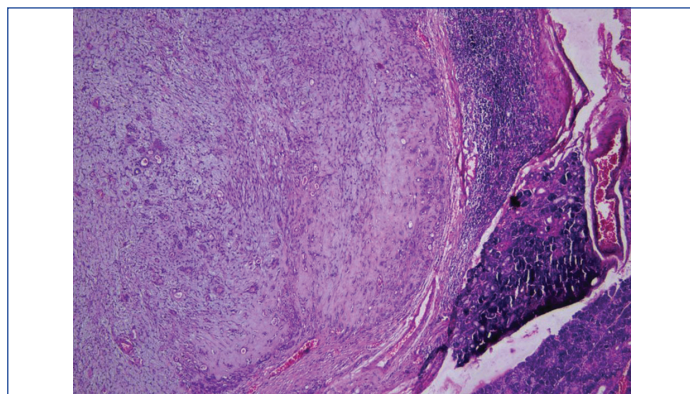
The patient was followed up regularly for almost two years with no complaints.



[Table/Fig-6]: Histopathological image of (400x) High power view showing bland cytomorphology of epithelial and myoepithelial cells.



[Table/Fig-7]: Photomicrograph (100x) Low power view showing epithelial cells in acini surrounded by abundant spindled myoepithelial cells in a background of chondromyxoid stroma.



[Table/Fig-8]: (40x) Scanner view showing the lesion (on the left) well demarcated from adjacent normal salivary gland (on the right), exhibiting all the three components chondromyxoid background, myoepithelial cells and few glandular epithelial cells.

Along with the tumour being removed under general anaesthesia during surgery, the submandibular gland was also removed. A 5 cm incision was made 2-4 cm below the inferior border of the jaw to cut through the platysma. The capsule of the gland and the soft tissue around it were preserved when removing the tumour. This method carries a risk to the marginal mandibular nerve, so the facial vein and artery were placed as close to the gland as possible and elevated superiorly after the transaction. The underside of the jaw was cut away to expose the gland and nearby soft tissue. The digastric muscle was then used to lift the inferior border of the gland. The lateral reflection of the gland reveals the mylohyoid muscle. Retraction of the free edge of the mylohyoid muscle allows visualisation of the lingual nerve, hypoglossal nerve, and Wharton's duct. The lingual nerve, which has its apex at the middle of the gland, supplies the gland with parasympathetic activity. The lingual nerve is located inferior to the hypoglossal nerve, which is situated below Wharton's duct and identified by an accompanying vein known as the ranine vein. Once all the tissues are identified, the gland's duct and branch of the lingual nerve are tied off and transected. Dissection allows for the separation and removal of the gland and the surrounding soft tissue. A rubber drain was inserted deep within the platysma, and the wound was then closed in stages. In cases with locally invasive tumours, the skin, tongue, mandible, lingual, hypoglossal, marginal mandibular nerve, and the floor of the mouth may all be removed. Since, this tumour is not invasive, the aforementioned parts were not removed.

DISCUSSION

Tumours of the salivary gland are rare, accounting for approximately 3% to 6% of all head and neck tumours [1]. The incidence of salivary gland tumours is around 2.5 to 3 cases per 100,000 per year in western countries such as the USA. Among these cases, about 65% to 80% arise from the parotid gland, 10% from the submandibular gland, and a very small number from minor

salivary glands, including the sublingual glands [2,3]. In contrast, between 70% and 90% of sublingual tumours and 40% to 50% of submandibular and small salivary gland tumours are malignant [4]. These tumours typically affect adults, with a slight female predominance. Warthin's tumour, however, is more common in males, likely due to a higher prevalence of smoking, a risk factor for this tumour [5]. Benign tumours are most commonly seen in the fifth to seventh decades of life, while malignant tumours tend to develop a little later [5].

Pleomorphic adenomas are benign tumours that exhibit both epithelial and mesenchymal differentiation, as they are composed of both ductal (epithelial) and myoepithelial cells. Due to their diverse histology, these tumours are sometimes referred to as mixed tumours. They account for approximately 60% of parotid tumours, are less common in submandibular glands, and are rare in minor salivary glands. Most cases occur in the superficial lobe of the parotid gland [2]. The relative incidence of pleomorphic adenoma is 50%, with the malignant variant mucoepidermoid carcinoma accounting for 15% [6].

Most pleomorphic adenomas present as rounded, well-demarcated masses that rarely exceed 6 cm in the largest dimension. Although they are encapsulated, in some locations (particularly the palate), the capsule is not fully developed, and expansile growth can lead to protrusions into the surrounding gland. In such cases, enucleation alone may result in recurrences. The main histologic feature is the significant heterogeneity described. The epithelial elements, resembling ductal cells or myoepithelial cells, are arranged in duct formations, acini, irregular tubules, strands, or sheets of cells. These elements are typically dispersed within a mesenchyme-like background of loose myxoid tissue containing islands of cartilage and, rarely, focal areas of bone [6].

Pleomorphic adenoma is an epithelial tumour with complex architecture, containing mucoid, myxoid, or chondroid tissue arranged in various patterns and embedded in a mucopolysaccharide stroma. It also consists of epithelial and myoepithelial elements. Pleomorphic adenoma is the most common benign salivary gland tumour, accounting for 90% of all salivary gland tumours [7]. While the parotid gland is the most common location for pleomorphic adenoma, it also frequently occurs in the submandibular gland [8]. It is the most common benign tumour of the submandibular gland. Differential diagnoses to consider include adenocarcinoma, mucoepidermoid carcinoma, lymphoma, and basal cell adenoma. CT scan or MRI are the recommended radiological tools for lesions originating from the main or minor salivary glands. Fine needle aspiration and ultrasonography-guided needle aspiration can be used as adjunctive diagnostic procedures, although they are not confirmatory. In cases of large lesions, an initial incisional biopsy may be performed. The preferred surgical technique is a direct submandibular incision, as it provides straightforward access [9].

In a 10-year study by Becerril-Ramírez Pb et al., a total of 22 cases of submandibular gland neoplasms were identified, of which 19 (86%) were benign and 3 (14%) were malignant. Pleomorphic adenoma was the most common benign tumour, accounting for 18 of the 19 cases. The female-to-male ratio of patients with pleomorphic adenoma was 3.5:1, with a mean age of incidence of 39.8 years [10].

In a study by Munir N and Bradley PJ spanning 16 years from 1988 to 2004, they evaluated a series of pleomorphic adenoma cases involving the submandibular gland. A total of 32 cases were treated during this period, with 22 (69%) being female, and the mean age of occurrence was 54 years. All patients had a clinically evident and palpable submandibular fossa mass, with 84% of cases being asymptomatic and 16% presenting with pain [11].

Rapidis AD et al., conducted a clinicopathological analysis of nine benign and 14 malignant submandibular gland tumours in 23

patients. They found that pleomorphic adenoma was the most common benign tumour, and its symptoms were relatively mild [12]. From 1990 to 2006, Adeyemo WL et al., conducted a study on cases of submandibular salivary gland cancers. They found that out of the 36 patients with submandibular gland tumours, 19 had malignant tumours, while 17 had benign tumours [13]. The most common tumour was pleomorphic adenoma (36.1%), followed by malignant lymphoma (11.1%), adenoid cystic carcinoma (11.1%), and anaplastic carcinoma (11.1%). The most common presentation was progressive painless swelling (80.6%), while painful masses (11.1%) and ulcerations (8.3%) were more indicative of malignancy.

De Oliveira FA et al., in Brazil reported that females are more frequently affected by salivary gland cancers, with a male-to-female ratio of 1:1.5. This ratio is 1:1.6 for benign tumours and 1:1.5 for malignant tumours. The average age of occurrence for benign tumours was 43 years, compared to 55 years for malignant tumours [14].

Alves FA et al., examined the immunohistochemical and clinicopathological characteristics of 60 cases of pleomorphic adenoma in Brazil. They found that the majority of cases (37/60; 62%) occurred in individuals between the third and fifth decades of life. Tumour sizes ranged from 1 to 10 cm. After three years of follow-up, only one patient had a local recurrence [15].

Patil P et al., reported a case of a 42-year-old male with a left submandibular swelling for eight months, which was diagnosed as pleomorphic adenoma. Total gland removal was performed, and the patient was followed for one year with no recurrence of the tumour [16].

Perumal CJ et al., reported a case of a 20-year-old female with an eight-year swelling in the left side of the jaw. The lesion was non-tender, multinodular, hard, and mobile, measuring 16x15x12 cm. An en bloc resection of the tumour was performed after an incisional biopsy, which showed features of myxochondroid and cellular pleomorphic adenoma [17].

Rai S et al., described a case of a 20-year-old male with a one-year history of left submandibular swelling, which was confirmed as pleomorphic adenoma through excisional biopsy under general anesthesia [18].

In a case report from 2019, a 38-year-old male presented with swelling of the right submandibular gland for two years, measuring 3x3 cm, with no nerve involvement or lymphadenopathy. Fine Needle Aspiration Cytology (FNAC) confirmed the diagnosis of pleomorphic adenoma. The gland with the tumour was excised without any complications during the postoperative period [19].

Abdel Khalek M et al., presented a rare case in 2019 of a giant submandibular pleomorphic adenoma measuring 34x26x20 cm. The 75-year-old woman had a tumour weight of 8.1 kg, involving the parapharyngeal and sublingual spaces and causing tracheal displacement. The tumour was completely excised, and the patient was discharged on the third postoperative day [20].

CONCLUSION(S)

Pleomorphic adenoma is the most common benign tumour that affects the submandibular gland. It typically occurs between the third and fifth decades of life and presents as a slow-growing, asymptomatic swelling. Although, the pleomorphic adenoma in the present case report is of small size, it is a rare case in our institution. Further research focusing specifically on malignancies that affect the submandibular gland is needed to enhance understanding of this condition.

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PLAGIARISM CHECKING METHODS: [Jain H et al.]

- Plagiarism X-checker: Jan 30, 2023
- Manual Googling: Jul 14, 2023
- iThenticate Software: Jul 20, 2023 (18%)

ETYMOLOGY: Author Origin**EMENDATIONS:** 7**AUTHOR DECLARATION:**

- Financial or Other Competing Interests: None
- Was informed consent obtained from the subjects involved in the study? Yes
- For any images presented appropriate consent has been obtained from the subjects. Yes

Date of Submission: **Jan 26, 2023**Date of Peer Review: **Apr 18, 2023**Date of Acceptance: **Jul 21, 2023**Date of Publishing: **May 01, 2024**