GSC Advanced Research and Reviews

eISSN: 2582-4597 CODEN (USA): GARRC2

Cross Ref DOI: 10.30574/gscarr

Journal homepage: https://gsconlinepress.com/journals/gscarr/

(CASE REPORT)

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Pleomorphic adenoma of the parotid with lymph node extension: A case report

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GSC Advanced Research and Reviews, 2025, 23(03), 067-073

Publication history: Received on 29 April 2025; revised on 08 June 2025; accepted on 10 June 2025

Article DOI: https://doi.org/10.30574/gscarr.2025.23.3.0157

Abstract

Pleomorphic adenoma (PA), the most common benign tumor of the salivary glands, typically arises from the parotid gland and is generally slow-growing, painless, and non-invasive. However, rare cases may exhibit aggressive behavior, including local recurrence and spread to regional lymph nodes. This case report presents a 53-year-old woman with a recurrent pleomorphic adenoma of the left parotid gland showing lymph node extension. The patient initially presented in 2021 with a painless mass in the left parotid region and underwent a total parotidectomy with modified radical neck dissection. Histopathological analysis confirmed pleomorphic adenoma with no malignant features. One year later, the mass recurred with associated multiple suspicious lymphadenopathies on imaging, prompting further evaluation. CT scan revealed a heterogeneous solid mass infiltrating adjacent muscles, with bilateral cervical lymph node enlargement. A second total parotidectomy was performed in 2024. Histopathology again confirmed pleomorphic adenoma without malignancy. Postoperative recovery was uneventful, with House-Brackmann grade III facial nerve weakness. This case underscores the potential for PA to recur and spread to regional lymph nodes even in the absence of malignant transformation, possibly due to incomplete excision or tumor seeding during prior interventions such as fine-needle aspiration biopsy (FNAB). Long-term follow-up is essential due to the risk of recurrence and potential progression to carcinoma ex-pleomorphic adenoma. This report highlights the importance of wide excision with clear margins and vigilance for lymphatic spread, even in benign histology, particularly in recurrent cases.

Keywords: Pleomorphic adenoma; Parotid gland; Lymph node extension; Recurrence; parotidectomy; FNAB; Benign salivary tumor

1. Introduction

Salivary glands are divided into major and minor salivary glands. The major salivary glands consist of a pair of parotid glands, submandibular glands, and sublingual glands, while the minor salivary glands are scattered throughout the oral cavity in the hundreds.[1-3] The primary function of the salivary glands is the production of saliva for lubrication and protection, buffering and cleansing food, maintaining dental integrity, antibacterial activity, taste, and digestion.[1]

Salivary gland tumors represent a diverse group of benign and malignant neoplasms with varying behavior. The incidence of salivary gland tumors accounts for 3%–6% of all head and neck tumors, with little variation over the past few decades. Approximately 70% of salivary gland tumors originate from the parotid gland. Among parotid gland tumors, about 75% to 80% are benign.[4–5] Pleomorphic adenoma (benign mixed tumor) accounts for 75% of parotid gland tumors, both benign and malignant, in adults. Adult women in their third to fifth decades are most commonly affected by pleomorphic adenoma.[6]

The only known effective treatment to date is total tumor excision. To minimize recurrence and the risk of malignant transformation, tumor integrity must be preserved during surgery, and the tumor must be completely removed.

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Although benign, this epithelial tumor has a tendency to recur and may undergo malignant transformation if incompletely excised, thereby increasing patient morbidity.[7] Pleomorphic adenoma can develop into benign but metastatic subtypes, most commonly spreading to bone, lungs, and lymph nodes.[4] Rarely, pleomorphic adenoma may metastasize while remaining histologically benign. These subtypes include metastatic pleomorphic adenoma, malignant mixed tumors such as carcinoma ex pleomorphic adenoma, and carcinosarcoma.[8-10]

2. Case report

A 53-year-old woman from Jembrana, Negara, Bali presented to the outpatient clinic at Prof. I.G.N.G Ngoerah Hospital with a complaint of a lump on her left cheek, at the same location as a previous episode two years ago. She first noticed the lump about 1.5 years ago (since mid-2022). The lump had gradually increased in size, but she reported no pain in the lump, no pain in the left ear, and no history of ear discharge. The patient denied hearing loss, headache, toothache, or facial numbness/tingling. She did not experience fever or pain during eating. Facial movement was preserved and symmetrical on both sides, both eyes could close fully, and her lips were symmetrical. There was no double vision or nosebleeds.

Three years ago, the patient had a similar complaint involving a lump on the left cheek, accompanied by enlarged lymph nodes (LNs) in the neck. A computed tomography (CT) scan at that time showed a heterogeneous solid mass in the left parotid gland, extending to the left parapharyngeal space and left pharyngeal mucosal space, encasing the left external carotid artery. Multiple lymphadenopathies were found in levels Ia/b, IIa/b, III, and V bilaterally. The patient underwent total left parotidectomy and left modified radical neck dissection (MRND) on October 4, 2021. Histopathological examination of the left parotid tissue revealed a pleomorphic adenoma with focal areas of hypercellularity and atypia, while histopathology of the lymph nodes showed no metastasis of malignant cells in any of the four sampled nodes.

The patient denied smoking and radiation exposure. She rarely consumes preserved foods such as salted fish or grilled foods. There was no history of lumps elsewhere, no history of head and neck malignancies, or other systemic illnesses. She also denied any family history of cancer.

On physical examination, her general condition was good, and she was alert (GCS E4V5M6), with a blood pressure of 130/70 mmHg, regular heart rate of 88 bpm, respiratory rate of 20 breaths/minute, and axillary temperature of 37.0°C. ENT local status examination revealed a firm, nodular mass in the left parotid region, fixed to the skin and underlying structures, measuring 6 cm x 8 cm, non-tender, with no skin discoloration **(Figure 1)**. Examination of the left ear and nose was unremarkable. Throat examination showed no abnormalities. There was no cervical lymphadenopathy, and facial nerve function was intact.

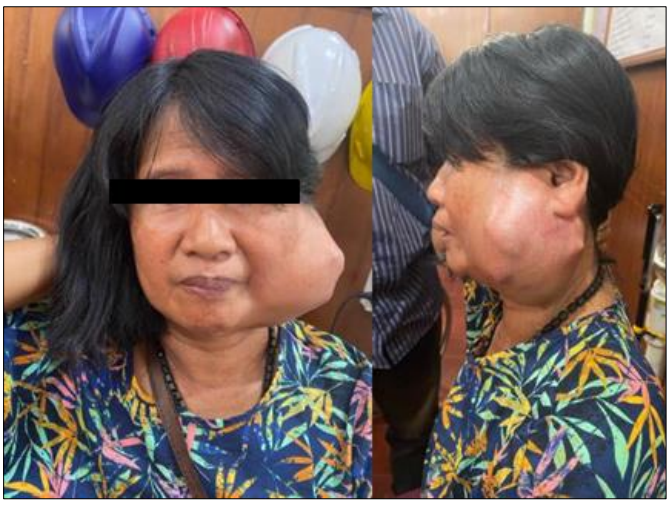

Figure 1 A lump was observed in the left parotid region, including the preauricular and retroauricular areas

In 2022, the patient underwent a CT scan due to a complaint of an enlarging lump on the left cheek. Based on the axial and coronal slices of the Midface and Neck CT scan with 2 mm thickness and contrast enhancement, a heterogeneous solid mass with well-defined margins and lobulated edges was observed in the left parotid region (left parotid space), suspected of infiltrating the left sternocleidomastoid muscle, suggesting a recurrent mass. Multiple suspicious

lymphadenopathies were identified in levels Ib, IIa/b, III, Va bilaterally, and Vb on the left. The patient was advised to undergo repeat surgery but did not return for follow- up.

A year later, the patient returned to Prof. I.G.N.G Ngoerah Hospital with a complaint of a lump that had reportedly grown larger. She was diagnosed with recurrent pleomorphic adenoma of the left parotid gland. The patient was given an explanation regarding the planned diagnostic procedures and surgery. A repeat Midface and Neck CT scan with axial, coronal, and sagittal slices of 2 mm thickness and contrast revealed a persistent heterogeneous solid mass with well-defined margins and lobulated edges in the left parotid region (left parotid space), now suspected of infiltrating both the left sternocleidomastoid and left masseter muscles, consistent with a recurrent mass with increasing size. Multiple suspicious lymphadenopathies were again identified in levels Ia/b, IIa/b, III, and Va/b bilaterally **(Figure 2)**.

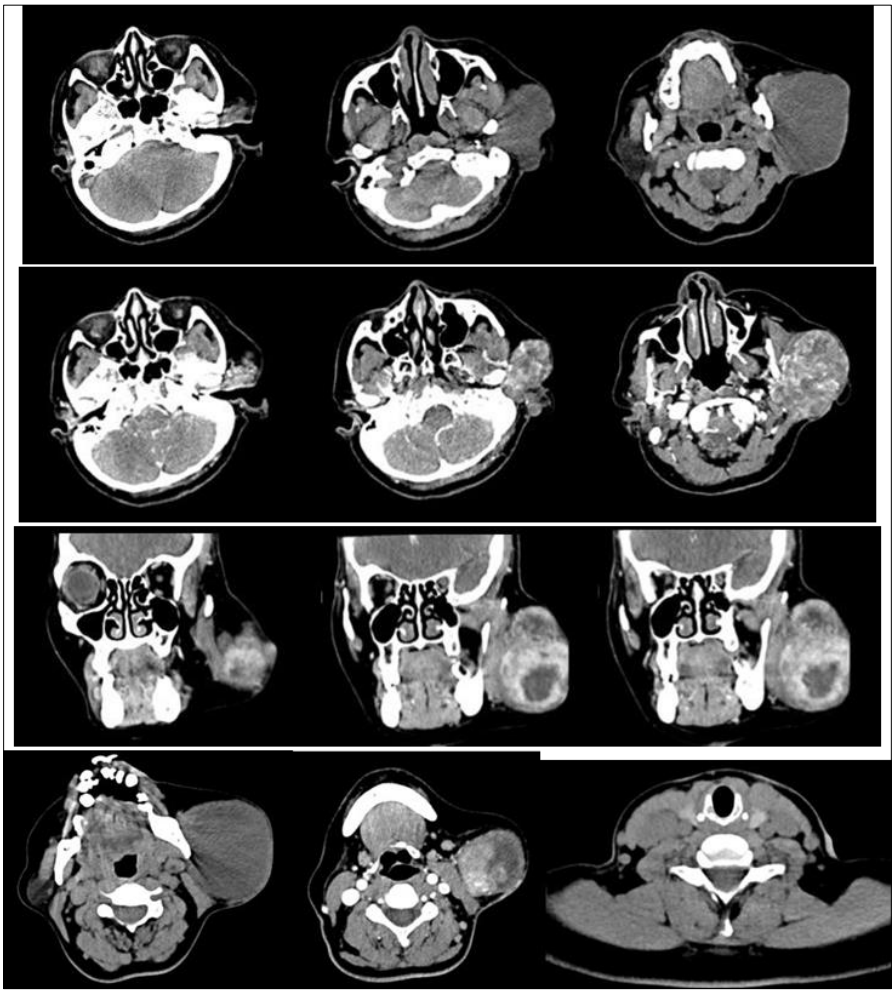

Figure 2 Axial, coronal, and sagittal slices of the contrast-enhanced midface CT scan show a tumor mass and bilateral lymph node enlargement

On November 15, 2023, laboratory tests, a posteroanterior (PA) chest X-ray, and a fine needle aspiration biopsy (FNAB) were performed. The laboratory and chest X-ray results were within normal limits. Cytomorphology from the FNAB was suggestive of a pleomorphic adenoma.

On January 17, 2024, the patient underwent a total left parotidectomy. This surgical procedure involved wide excision. During the operation, the main branches of the left facial nerve exiting from the stylomastoid foramen were identified; however, it was difficult to completely preserve the facial nerve branches. Intraoperatively, a firm superficial and deep multilobulated mass measuring 15 x 10 x 8 cm was found in the left parotid region **(Figure 3)**.

Postoperative follow-up: the patient was admitted to a general ward. A drain was placed in the left parotid region with drainage output of 15 cc on first postoperative day, and the patient was in good condition. The patient was discharged on the second postoperative day after drain removal and scheduled for outpatient follow-up. Wound care was

performed during follow-up visits on postoperative days 7 and 10 **(Figure 4)**. The surgical wound appeared clean and dry, with no signs of infection. Facial nerve examination revealed left facial nerve weakness, graded House-Brackmann III at the extratemporal level.

Histopathological examination of the tissue specimen showed a well-circumscribed tumor mass, encapsulated by fibrous connective tissue. The tumor mass consisted of a proliferation of neoplastic cells comprising three components: epithelial, myoepithelial, and stromal. The epithelial and myoepithelial components appeared hypercellular, predominantly arranged in cord-like patterns with some tubular patterns, embedded within a fibromyxoid stroma. The cells showed cuboidal morphology with eosinophilic cytoplasm. The myoepithelial cells appeared round to spindle-shaped, some with clear nuclei and eosinophilic cytoplasm, partially lining the outer layer of the tubular structures. The stromal component was myxoid mesenchymal material. The connective tissue stroma between tumor masses appeared hyalinized. No malignant cells were seen in the specimen. The histomorphology was consistent with pleomorphic adenoma.

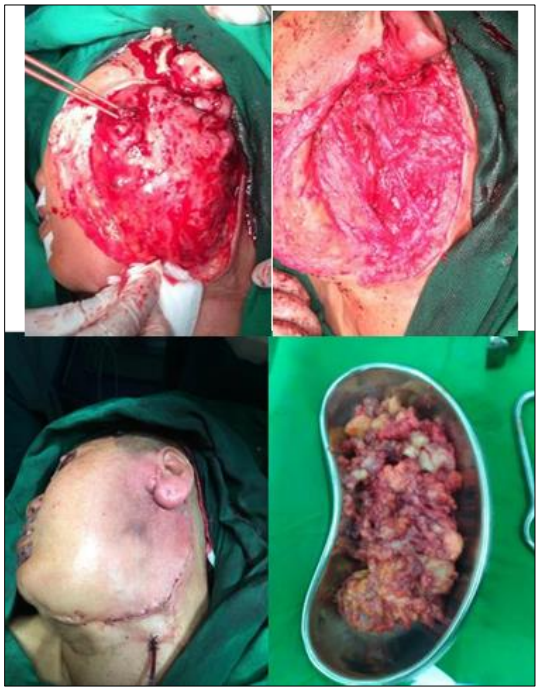

Figure 3 Tumor mass in the left parotid region (A); after the mass was excised (B); after the skin was sutured (C); and the separated mass measuring 15 x 10 x 8 cm (D)

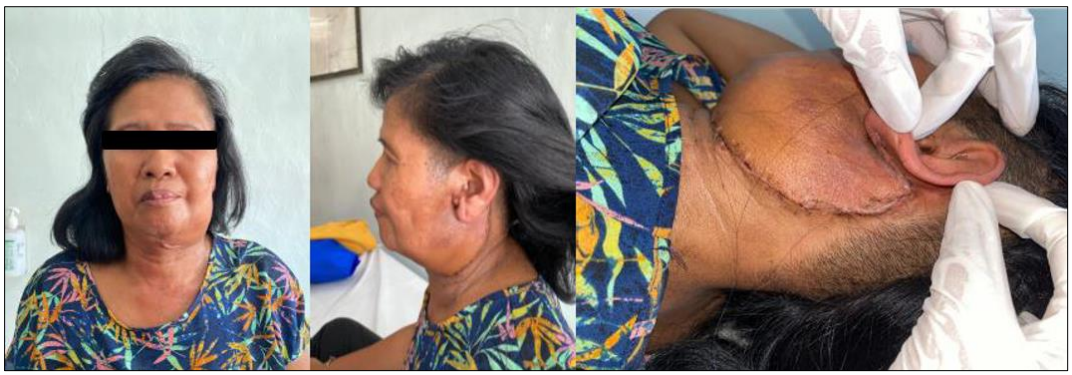

Figure 4 Well-maintained wound, 10 days post-operation

3. Discussion

Pleomorphic adenoma accounts for 65% of all salivary gland tumors, most commonly found in the parotid gland, followed by the submandibular gland and minor salivary glands. In the parotid gland, the mass is usually located in the superficial lobe and presents as swelling at the mandibular ramus in front of the ear. A small portion of lesions grow medially between the ascending ramus of the mandible and the stylomandibular ligament, appearing in the lateral pharyngeal wall or soft palate. The mass presents as an irregular nodular lesion with a firm consistency. It is generally asymptomatic, with no pain or facial nerve involvement.

In this case, a firm, lobulated mass fixed to the skin and base was found, measuring 6 cm x 8 cm, extending from the mandibular ramus in front of the ear to the postero-inferior aspect of the ramus (below the ear). There was no tenderness or skin discoloration. No facial nerve dysfunction was observed prior to surgery.

Eveson et al. reviewed 2,410 cases of pleomorphic adenoma and found that the disease typically presents between the 4th and 6th decades of life, with a mean age of 43 to 46 years. The patient was 53 years old, which fits the epidemiological profile of pleomorphic adenoma, typically occurring between ages 40–60, with the primary mass appearing at around age 50. Pleomorphic adenoma clearly shows a female predominance, with a male-to-female ratio of 8:13.

No history of radiation exposure was reported in this patient, which, while still suspected as a possible etiology of pleomorphic adenoma, remains unproven to date.

Oh et al. describe pleomorphic adenoma as a benign mixed tumor with mesenchymal and epithelial components. These are single cells that differentiate into epithelial or myoepithelial cells not merely a simultaneous proliferation of carcinogenic epithelial and myoepithelial cells. The tumor has three components: epithelial, myoepithelial, and mesenchymal. Diagnosis is established by identifying all three components.

The mesenchymal part shows variable epithelial patterns in a loose fibrous stroma, which may be myxoid, chondroid, mucoid, fibroid, or osteoid in nature. Myoepithelial cells are polygonal with pale eosinophilic cytoplasm.

In our case, the tumor mass consisted of a proliferation of neoplastic cells with the three characteristic components epithelial, myoepithelial, and stromal. The cells were cuboidal with eosinophilic cytoplasm. The myoepithelial cells ranged from round to spindle-shaped, some with clear nuclei and eosinophilic cytoplasm, and in part lining the outermost parts of tubular structures. The stromal mesenchyme was myxoid, with hyalinized fibrous stroma between the tumor masses.

In the initial 2021 presentation, a mass was found in the left parotid region with lymph node enlargement at levels Ia/b, IIa/b, III, and V bilaterally. We suspected this lymphadenopathy represented spread from the parotid pleomorphic adenoma, consistent with a publication by Ko et al., which reported level II lymph node involvement in patients without any prior invasive procedures or surgery in the head and neck area. One year after left parotidectomy and left MRND, there was recurrent enlargement in the left parotid region and lymphadenopathy at levels Ib, IIa/b, III, Va bilaterally, and Vb on the left. Contrast-enhanced CT revealed a mass in the left parotid region suspected of infiltrating the left sternocleidomastoid muscle, with multiple suspicious lymphadenopathies at the aforementioned levels. Pleomorphic adenoma can evolve into a benign but spreading subtype called metastasizing pleomorphic adenoma, or into a malignant mixed tumor known as carcinoma ex pleomorphic adenoma or carcinosarcoma. These tumors most commonly spread to the bone, lungs, and lymph nodes.

Most pleomorphic adenoma dissemination cases occur postoperatively due to incomplete excision, tumor spillage at the surgical site, or tumor seeding implantation of tumor cells via hematologic or lymphatic routes. Tumor spread has also been suspected after FNAB or core biopsy. In our case, lymph node involvement was already observed at the first tumor presentation three years ago, consistent with the case reported by Ko et al., showing lymph node metastases prior to any invasive procedure. The spread pattern in our case aligns with systematic reviews showing that pleomorphic adenoma most frequently spreads to bone, lungs, and lymph nodes.

The patient underwent left parotidectomy and MRND in 2021, with histology confirming pleomorphic adenoma. In 2022, one year post-surgery, the tumor enlarged again, and FNAB of the left parotid indicated recurrent pleomorphic adenoma. Fikova et al. reported slightly lower values (sensitivity 88.83% and specificity 96.23%) but emphasized that FNAB remains a reliable diagnostic method for pleomorphic adenoma.

Fonseca et al. reported a similar case in a 29-year-old woman who underwent two parotidectomies 15 and 8 years prior yet experienced recurrent growth in the parotid region. Mechanisms of pleomorphic adenoma recurrence include detached pseudopodia, residual satellite nodules, incomplete capsular excision, or accidental opening of fragile hypocellular subtypes, all of which make recurrence difficult to prevent. Preserving the tumor capsule during surgery is particularly challenging, especially in extended or infiltrative tumors.

The main treatment for recurrence or extensive pleomorphic adenoma remains surgical resection, as long as the case is operable. Other therapeutic options are still being studied, particularly for recurrent or widespread disease. Radiotherapy may be considered in unresectable cases. There are few reports on long-term prognosis, but multiple or distant spread has been identified as an important predictive factor for poor outcomes

4. Conclusion

Salivary gland tumors represent a diverse group of benign and malignant neoplasms, accounting for 3%–6% of all head and neck tumors. Approximately 70% of salivary gland tumors originate from the parotid gland. Pleomorphic adenoma (benign mixed tumor) causes 86% of salivary gland tumors in adults. Adult women in their third to fifth decades are most commonly affected by pleomorphic adenoma. Fine-needle aspiration biopsy (FNAB) is a reliable method for diagnosing pleomorphic adenoma. Complete surgical excision of the tumor is the definitive treatment.

Most cases of pleomorphic adenoma spread occur in the context of tumor recurrence, typically after previous procedures due to incomplete excision, possible tumor spillage at the surgical site, or tumor seeding, which refers to implantation of tumor cells through hematologic or lymphatic pathways. Even after surgical removal, long-term follow-up is necessary to monitor for recurrence.

Compliance with ethical standards

Disclosure of conflict of interest

No conflict of interest to be disclosed.

Statement of informed consent

Informed consent was obtained from the patient.

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