Case Report

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Ear, Nose and Throat Section

Salivary Duct Carcinoma with Comedo Necrosis in Level 1B Lymph Nodes: A Case Report

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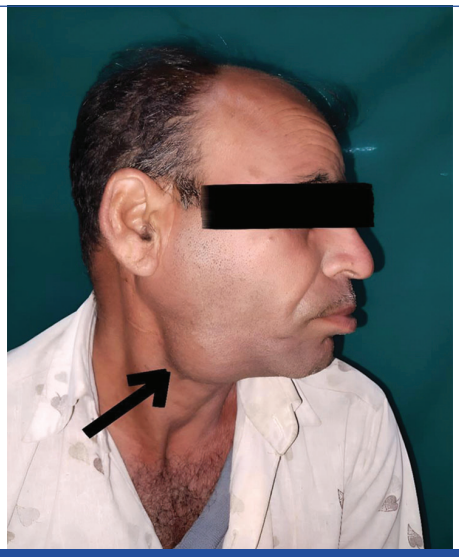
ABSTRACT

Salivary Duct Carcinoma (SDC) is a rare and highly aggressive cancer that originates in the ductal epithelium of the salivary glands, primarily affecting the parotid and submandibular glands. Cases involving minor salivary glands are uncommon. Due to its high grade, SDC requires aggressive treatment, including complete local excision and postoperative radiotherapy for optimal patient outcomes. The present report discusses a case involving a 50-year-old male with a solitary, firm, non tender swelling measuring approximately 4×4 cm in the right submandibular region, extending to the tonsillolingual junction. The right floor of the mouth displayed a fixed, congested mucosa attached to a palpable mass, causing oral bleeding. Histopathology revealed a cribriform pattern with comedo necrosis. Immunohistochemistry confirmed the diagnosis of SDC, with tumour cells showing positive results for cytokeratin-7, c-erb B2 and Mindbomb Homolog-1 (MIB-1). The mass was surgically excised, followed by postoperative radiotherapy. The patient was monitored for 21 months without any local recurrence or distant metastasis, indicating successful treatment. Complete surgical excision of the swelling, followed by postoperative radiotherapy, can lead to disease-free survival with early diagnosis and proper management.

Keywords: Immunohistochemistry, Lymph nodes, Radiotherapy

CASE REPORT

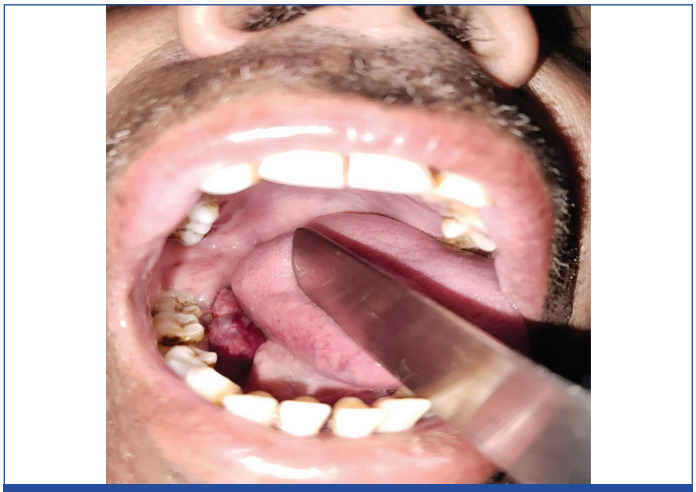
A 50-year-old male patient arrived at the Ear, Nose and Throat (ENT) Department with the chief complaint of progressive swelling in the right submandibular region, which had been developing for one year, along with oral bleeding for the past eight months. The patient reported no associated pain or neurological deficits [Table/Fig-1].



[Table/Fig-1]: Clinical picture of lateral view of patient.

A single, solitary, firm, non tender and mobile swelling measuring approximately 4×4 cm was observed in the right submandibular region, causing a bulge in the right floor of the mouth, extending to the tonsillolingual junction. The mucosa on the right-side of the floor of the mouth appeared fixed to the underlying palpable mass and was congested, which likely accounted for the oral bleeding [Table/Fig-2].

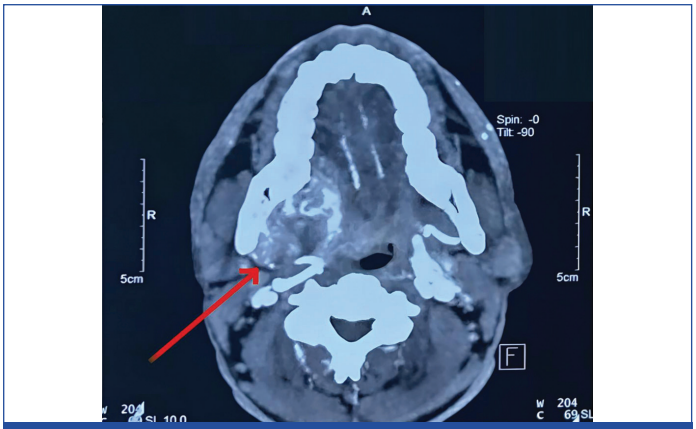
A well-defined hypoechoic solid lesion with internal vascularity was identified in the right submandibular region, measuring approximately 60×29×52 mm. This lesion caused anterolateral displacement of the right Internal Carotid Artery (ICA) and External Carotid Artery (ECA), while also compressing the right Internal Jugular Vein (IJV), suggestive of a right carotid space schwannoma or neurofibroma.



[Table/Fig-2]: Intraoral examination of mouth showing bulge over right-side floor of mouth.

A Computed Tomography (CT) scan of the neck with angiography showed a well-defined, heterogeneously enhancing, hypoechoic soft-tissue mass with internal vascularity in the right submandibular region, measuring approximately 5.3×4.3×8.1 cm. The mass exhibited multiple tortuous vessels and some cystic areas. Anteromedially, it displaced the tongue to the contralateral side while maintaining a fat plane between them. Posteromedially, it caused narrowing of the oropharyngeal lumen. Laterally, the mass reached the skin and subcutaneous tissue without eroding the right mandible. Posteriorly, it approached the right sternocleidomastoid muscle and carotid space, mildly compressing the lumen of the right IJV, yet preserving the fat plane. Superiorly, it extended to the right masticator space, again with a maintained fat plane adjacent to the right pterygoid muscle. These findings suggest a benign vascular lesion of the right submandibular gland, likely a haemangioma. Additionally, multiple subcentimetric lymph nodes were noted in bilateral cervical levels II, III and IV [Table/Fig-3].

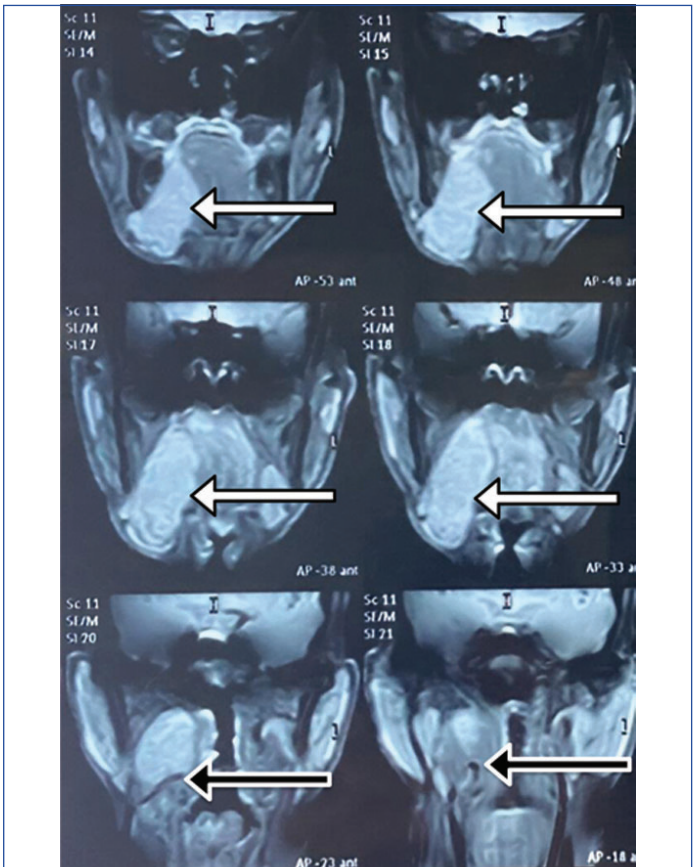
An MRI of the head and neck showed an approximately 70×41×70 mm Anteroposterior×Transverse×Craniocaudal (AP×TR×CC) sized, well-defined lobulated lesion with a smooth margin involving the right



[Table/Fig-3]: CT Neck with angiography shows a heterogeneously enhancing, hypoechoic soft-tissue mass in the right submandibular region (red arrow).

submandibular and parapharyngeal spaces. The lesion appeared isointense on T1 and heterogeneously hyperintense on T2/Short Tau Inversion Recovery (STIR) images (as compared to muscle). The lesion did not show diffusion restriction and exhibited heterogeneous post-contrast enhancement. The lesion caused compression and inferior displacement of the right submandibular gland posterolaterally. The carotid vessels appeared posterior to the lesion.

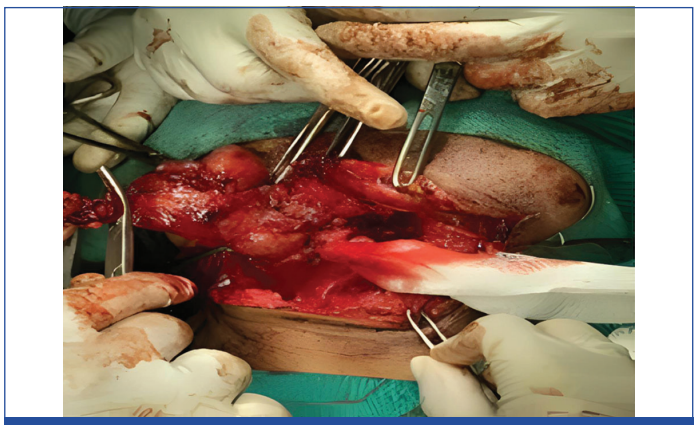
Medially, the lesion exerts a mass effect in the form of left lateral displacement of the tongue and floor of the mouth. It abuts and displaces the styloglossus, hyoglossus, stylohyoid and posterior belly of the digastric muscles, with a preserved fat plane between them. Posteromedially, the lesion reaches up to and involves the parapharyngeal space, pushing the oropharynx towards the left. Posterolaterally, the lesion abuts the deep lobe of the parotid gland. Posteroinferiorly, the lesion extends up to the platysma and abuts it. Inferomedially, the lesion reaches up to the hyoid bone. The findings were suggestive of a benign neoplasm—likely a schwannoma [Table/Fig-4].



[Table/Fig-4]: MRI head and neck shows serial sections of a well-defined lobulated lesion with a smooth margin. White arrow shows the lesion as heterogeneously hyperintense on T2/STIR images (as compared to muscle). Black arrow shows isointense on T1 images.

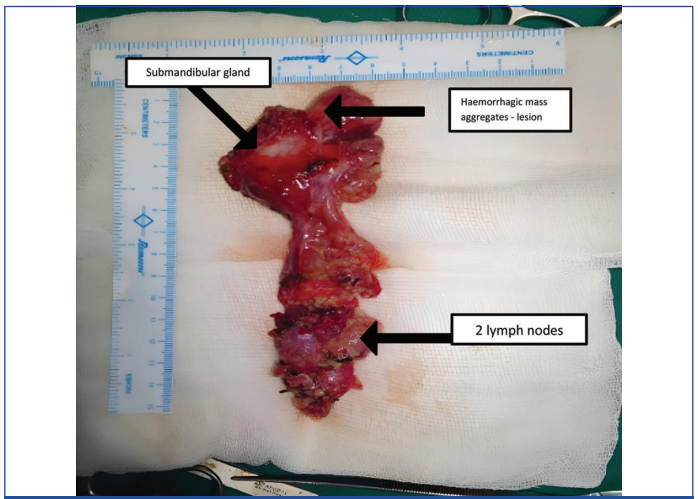
Fine Needle Aspiration Cytology (FNAC) was performed and the smears were highly cellular, displaying ductal epithelial cells arranged in sheets, clusters and microacini, along with solitary cells amidst a background of haemorrhage. The individual cells exhibited mild pleomorphism, granular chromatin and prominent nucleoli. Stripped nuclei of varying sizes were also present. These findings suggest a neoplastic aetiology with undetermined malignant potential.

The patient underwent general anaesthesia for the excision of a tumour in the right submandibular region. A skin crease incision was made in that area and after elevating the superior and inferior subplatysmal flaps, the mass was identified and dissected. Upon retracting the right mylohyoid muscle, the mass was found to extend into the floor of the mouth, medial to the ramus of the mandible, reaching the right masticator space. It was completely removed along with the right submandibular gland. Sutures were applied in two layers using 3-0 vicryl and a negative pressure Romovac drain (No. 10) was placed. There were no postoperative complications such as pain, bleeding, haematoma, or facial paresis [Table/Fig-5].



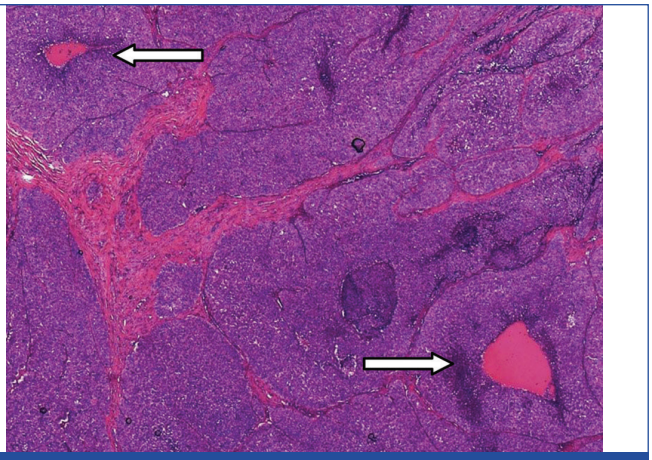
[Table/Fig-5]: Surgical excision of right submandibular lesion with gland.

Multiple brownish-white soft-tissue masses aggregated to approximately 9.5×9×3 cm, while the salivary gland measured 3.5×2.8×1 cm. Some areas showed haemorrhage and cystic changes. Additionally, two lymph nodes were present, with the largest measuring 0.6 cm in diameter [Table/Fig-6].



[Table/Fig-6]: Gross examination of specimen of right submandibular lesion with gland.

The study section reveals glands arranged in a back-to-back pattern with minimal intervening stroma. Individual cells display nuclear pleomorphism, a high nuclear-to-cytoplasmic ratio and hyperchromatism, with occasional mitotic figures. Focal areas exhibit comedo necrosis in the right level 1b lymph nodes, probably basal-like in histological pattern; however, no malignancy was evident in the adjacent right submandibular gland. The diagnosis was adenocarcinoma with comedo necrosis in the level 1b lymph nodes [Table/Fig-7].



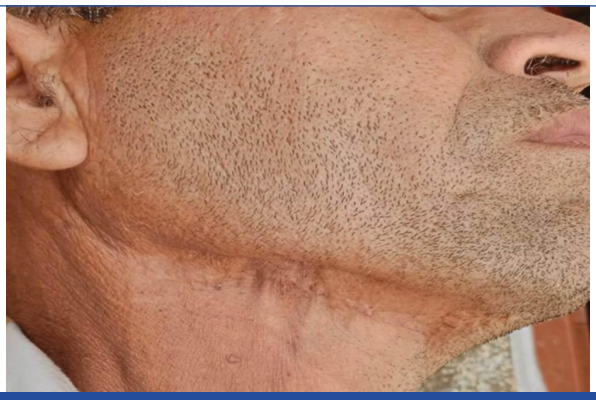
[Table/Fig-7]: Histopathological examination of submandibular lesion (H&E, 10x), white arrow shows focal areas of comedo necrotic lesion with pink necrotic center, surrounded by hypercromatic (purple) tumour cells.

The section shows a tumour primarily arranged in a cribriform pattern with areas of comedo necrosis and no perineural invasion is identified. The surrounding normal salivary gland tissue appears unaffected and the lymph nodes show no metastatic deposits.

Immunohistochemical examination reveals that tumour cells express CK7 (Clone OV-TL 12/30, Dako) and c-erbB2 (Polyclonal Rabbit c-erbB2 Oncoprotein, Dako), but do not express CK20 (Clone Ks 20.8, Dako), CK5/6 (Clone D5/16 B4, Dako), or p63 (Clone DAK-p63, Dako). The MIB-1 labelling index (Ki-67 Antigen, Clone MIB-1, Dako) is 40%. These findings are suggestive of SDC in the submandibular gland.

Approximately 34 days after the excision of the tumour, 60 Gy of radiotherapy was administered over 30 cycles, lasting 50 days. Five cycles of radiotherapy were given over six weeks, with a dosage of 2 Gy per cycle.

The patient was followed-up for 21 months. The scar site was healthy, showing no complications and no signs of recurrence [Table/Fig-8].



[Table/Fig-8]: Postoperative 21 months with no signs of residual lesion or recurrence.

DISCUSSION

The SDC is a rare, aggressive, high-grade malignancy of the salivary glands, representing about 0.2% to 2% of all salivary gland tumours [1,2]. Salivary gland cancers are uncommon, constituting less than 1% of all malignancies and under 5% of head and neck cancers [3]. SDC is most frequently found in the parotid gland (78-83%), while the submandibular gland (12%) and minor salivary glands (10%) are less commonly involved [3].

The SDC primarily affects men over the age of 50 [2], typically occurring during their fifth to sixth decades of life [3], with a male-to-female ratio of approximately 2:1. Patients range from 23 to 80 years old, indicating a prevalence among elderly men [4].

When evaluating lesions in the cervical lymph nodes or submandibular gland via imaging, the location, structure and number of lesions are crucial factors. Imaging techniques, such as CT or Magnetic Resonance Imaging (MRI), are more effective than clinical examination for detecting nodal metastasis, making them standard aspects of head and neck cancer work-up [5]. Differential diagnosis may include sialolithiasis, calcifications associated with the tumour, peripheral calcifications from tuberculosis, or other malignant tumours such as squamous cell carcinoma and papillary thyroid carcinoma [6]. In the present case, the radiological differential diagnosis considered were haemangioma, schwannoma, or neurofibroma, given their similar imaging characteristics. This rare adenocarcinoma exhibits a cribriform pattern and comedo necrosis, akin to ductal carcinoma of the breast [2].

Histologically, SDC is characterised by clusters of large cuboidal cells with both intraductal and invasive components, atypical mitotic figures and arrangements that include cribriform, papillary and solid patterns, along with comedo necrosis [4]—similar to the histopathological findings in this patient. The grading of SDC includes four histologic patterns: sarcomatoid, mucin-rich, micropapillary and basal-like [3]. Differential diagnosis for adenocarcinoma in neck lymph nodes as per histopathology include metastases from the lungs, breast and gastrointestinal tract, while primary salivary gland malignancies should also be considered [7].

Common immunohistochemistry markers used to confirm comedo necrosis include High Molecular Weight Cytokeratin (HMWCK) and p63 [8]. Immunohistochemically, SDCs are positive for low and HMWCKs, along with markers such as Carcinoembryonic Antigen (CEA), LeuM1 and Epithelial Membrane Antigen (EMA). Notably, all SDCs exhibit strong nuclear reactivity for androgen receptors [9]. SDC cells also show focal positivity for the apocrine marker GCDFP-15 and Mitochondrial Antigen (MIA), while typically being negative for S-100 protein, myoepithelial markers and oestrogen and progesterone receptors [2]. The p63 marker, indicative of squamous cell carcinoma, was negative in this case, helping to exclude that diagnosis. Ultimately, the histopathological diagnosis of SDC was established in the present case through haematoxylin and eosin staining, corroborated by immunohistochemical results showing positivity for cytokeratin CK7, c-erbB2 and MIB-1, alongside negativity for CK20, CK5/6 and p63 [10].

For primary disease, wide local excision followed by adjuvant postoperative radiotherapy is advised, particularly in cases of perineural or microvascular invasion, high-grade tumours, close or positive resection margins and cervical lymph node metastases at diagnosis [6,10,11].

Nakaguru M et al., conducted a thorough histological review of 151 SDC cases and analysed the relationship between different histomorphologic parameters and clinical outcomes in order to create a histological risk stratification model that predicts the prognosis of SDC patients. Four histological features—poorly differentiated clusters, vascular invasion, mitoses and substantial nuclear pleomorphism—were used to categorise the risk, according to the study's findings [12].

A total of 14 cases of SDC with perineural invasion and an invasive micropapillary component (invasive micropapillary SDC) were reported by Nagao T et al., [13]. These cases had small cell clusters that resembled morulas and lacked fibrovascular cores [13].

The study by Williams L et al., determined that SDC exhibited a variety of morphologies, including micropapillary (n=6), sarcomatoid (n=3), mucinous (n=2) and basal-like (n=1) [14].

A multidisciplinary cancer care team should discuss each case to provide optimal treatment recommendations, which may include surgery, radiotherapy, chemotherapy, hormonal therapy, or palliative care as needed.

CONCLUSION(S)

Adenocarcinoma with comedo necrosis is a rare and often challenging condition to diagnose, relying on the patient's history,

physical examination and primarily on histopathological and immunohistochemical findings. Despite the aggressive nature of SDC, which is linked to metastasis and low survival rates, early diagnosis and proper management can significantly enhance survival outcomes.

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