Title: A case report on Adenoid cystic carcinoma of nasal cavity: Successfully treated by endoscopic surgical excision

Abstract:

Cancer in sinonasal cavity is an extremely uncommon condition. Most of the time, the symptoms resemble allergy symptoms, making it difficult to diagnose a malignancy. It is critical to distinguish between benign and malignant nasal masses. A clinical correlation is required for this, as well as radiological and pathological linkage. Minor salivary gland tumours, particularly in the nose, are uncommon in the head and neck region. Adenoid cystic carcinoma is a relatively common malignant tumour that consists primarily of a minor salivary gland cancer in the sinonasal cavity. Only a few studies have been published, despite the fact that adenoid cystic carcinoma (ACC) is the second most prevalent cancer to develop in the sinonasal tract. The clinical manifestation and tumour stage will always influence the course of treatment. Due to the late presentation, it manifests in a stage where surgery and adjuvant therapy may be necessary for treatment. We are providing a case report of a male patient, age 55, who underwent successful endoscopic endonasal treatment one year back for a well localised (T1N0M0) left sided nasal tumour, an adenoid cystic carcinoma, at Apollo E.N.T. hospital, Jodhpur, Rajasthan, India.

Keywords: Sinonasal malignancy, adenoid cystic carcinoma,minor salivary gland, endoscopy, surgical excision

Introduction:

Initially referred to as a "cylindroma" by Bill Roth in 1859, adenoid cystic carcinoma (ACC) is an uncommon form of malignant epithelial tumour [(1)]. It is typical in the minor salivary glands surrounding the mouth, rare in parotids, and uncommon in the paranasal sinuses and nose [(2,3)]. Three to five percent of head and neck cancers are adenoid cystic carcinomas (ACC). It is distinguished by a moderate growth rate, a low likelihood of lymphatic dissemination, and frequent lung metastases [(4)]. In contrast to metastatic disease, which has survival rates of 43.3% and 55.4%, localised ACC that starts in the major salivary glands and small salivary glands of the oral cavity has OS rates of 93.9% and 92.4%, respectively [(5)]. ACC of the paranasal sinuses and skull base, however, is a disease with unique clinical consequences [(6)]. These tumours are frequently discovered after they have already spread, and adequate oncological resection is less possible because to the tumours' proximity to important tissues (such as the dura, brain, orbit, and central nerves) [(7)]. Perineural spread, which has an incidence of more than 50%, is another feature of paranasal sinus ACC [(6)]. It has been challenging to establish the characteristics and treatment choices for the many histologies that are reported in this region due to the rarity of sinonasal malignancies. However, endoscopic surgical excision is the preferred course of treatment for early stages like T1 . Adjuvant radiation therapy is only used in cases of positive margins or advanced stage and is not the primary method of treatment for ACC [(8-10)].

Case report:

A 56-year-old male patient arrived at our Apollo E.N.T. hospital complaining of a left sided nasal blockage for the past 12 months. Over time, he became aware of a pinkish tumour in the left side of the nasal cavity. He had a history of a headache on the left side and watering discharge from left eye. He also had the nasal quality of his voice. He denied any addiction. No trauma, nasal haemorrhage, anosmia, or hyposmia were in his past. No prior history of cheek fullness existed. His sight was unimpaired. On anterior rhinoscopy (figure1A), the inferior turbinate was enlarged and the nasal mass was seen medial to the inferior turbinate. The intraoral and neck examinations were normal. On diagnostic nasal endoscopy (figure 1B), the septum was being pushed toward the right side by a reddish mass that filled the whole left side of the nasal cavity. A contrast-enhanced computed tomography was indicated for the patient (Figures 2 and 3), which suggested a left-sided nasal mass occupying the left nasal cavity. The osteomeatal complex was being obstructed, and the septum was migrating to the right side. The ethmoid and maxillary sinus planes were preserved. Following clinical and radiographic evaluations, this tumour was staged as T1N0M0. A biopsy revealed that the patient had adenoid cystic carcinoma. The patient was scheduled for endoscopic removal of the nasal tumour. The patient had been informed about the nature of the illness and its potential for recurrence. Following agreement, the patient underwent an endoscopic endonasal approach for the removal of nasal mass. During surgery, an endoscopic medial maxillectomy was performed. The nasolacrimal duct was severed. The nasal tumour was completely removed, and the tissue sample was sent for histopathological analysis. Patient after surgery moved to recovery and continued receiving intravenous antibiotics for two days. It was identified as an adenoid cystic carcinoma in the histology report without perineural invasion. Since the tumour was precisely localised in the nasal cavity and margins were free of tumour, we didn't recommend radiotherapy in this case. Patient had no recurrence throughout his one-year follow-up and was doing well.

Discussion:

The sinonasal tract is home to a number of malignancies with salivary gland origins, such as adenocarcinoma, adenoid cystic carcinoma and mucoepidermoid carcinoma. [11] Adenoid cystic carcinoma (ACC) is the most common minor salivary gland tumour in the sinonasal tract, after squamous cell carcinoma, accounting for 10% to 25% of all ACC in the head and neck. [12-14] Nasal blockage, facial pain, epistaxis, nasal discharge, and loss of smell are among the common presenting symptoms of sinonasal ACC; same symptoms are often seen in patients with sinusitis and inflammatory nasal diseases. Delays in diagnosis and treatment may result from this. The nasal cavity and maxillary sinus are where these tumours most frequently occur. [18] Patients with nasal cavity tumours had the highest overall and disease-specific survival rates, whereas those with sphenoid tumours had the lowest rates. [15] Overall, 5-year survival rates for patients with sinonasal ACC from 50% to 86% have been reported. [18] Additionally, it has been noted that ACC of the paranasal sinuses has a lower metastatic rate than ACC of the primary salivary glands. Surgery is the mainstay of ACC treatment; adjuvant radiation therapy is only used if the tumour has positive margins or is

advanced in stage.[15-17] For early incidence, thorough removal of the main tumour with enough tumor-free margin is advised. A well-localized nasal tumour was endoscopically removed from the patient in the current case report (T1N0M0). In contrast to squamous cell carcinomas of the head and neck, ACC rarely develops cervical lymph node metastases. However, it is believed that the occurrence of lymph node metastases is a poor predictor of distant metastasis and survival.[19] The limited lymphatic distribution in the sinonasal tract and surrounding structures may be the cause of the sinonasal ACC's lower nodal metastasis rate than other ACCs of the head and neck. Patient in the current case study had no prior history of cervical lymphadenopathy.

Conclusion:

Paranasal sinus tumours are uncommon in comparison to other head and neck sites, particularly the major salivary glands. Sinonasal ACC is a rare tumour that usually manifests as late-stage disease. TNM stage, histological subtype, treatment modalities, and local control all affect 5-year overall survival. The key to success in the treatment of sinonasal ACC is early diagnosis. When the tumour is sufficiently localised, surgery is the preferred method of treatment. Adjuvant radiation may or may not be required, depending on the margin situation.

Consent:

The study was published with the written consent of the patient.

Ethical Approval

The procedure performed in this case report was in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

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Conflict of Interest:

The author (s) declares no potential conflicts of interest with respect to the research, authorship, and/or publication of this paper.

References:

1. Li Y., Hao D., Song X., Zhang C. Computed tomography and magnetic resonance imaging of adenoid cystic carcinoma in the maxillary sinus: a retrospective study with radiologic-histopathologic correlations. Oral Surg. Oral Med. Oral Pathol. Oral Radiol. 2020

2. Kumar V.P., Rao P.N., Kumar G.A. Adenoid cystic carcinoma of nasal cavity-a case report. Indian J. Otolaryngol. Head Neck Surg. 2003;**55**(1):43–46.
3. Lahjaouj M, Berrada O, Rayhane A, Abada RA, Rouadi S, Roubal M, Mahtar M. Advanced adenoid cystic carcinoma of maxillary sinus: Rare case report and review of literature. International Journal of Surgery Case Reports. 2021 Mar 1;80:105622.
4. Bhayani MK, YenerM, El-Naggar A, et al. Prognosis and risk factors for early-stage adenoid cystic carcinoma of the major salivary glands. Cancer 2012;118(11):2872–2878.
5. Ellington CL, GoodmanM, Kono SA, et al. Adenoid cystic carcinoma of the head and neck: Incidence and survival trends based on 1973-2007 surveillance, epidemiology, and end results data. Cancer 2012;118(18):4444–4451.
6. Gil Z, Carlson DL, Gupta A, et al. Patterns and incidence of neural invasion in patients with cancers of the paranasal sinuses. Arch Otolaryngol Head Neck Surg 2009;135(2):173–179.
7. Patel SG, Singh B, Polluri A, et al. Craniofacial surgery for malignant skull base tumors: report of an international collaborative study. Cancer 2003;98(6):1179–1187.
8. Lupinetti AD, Roberts DB, Williams MD, et al. Sinonasal adenoid cystic carcinoma: the M. D. Anderson Cancer Center experience. Cancer 2007;110(12):2726–2731.
9. Pommier P, Liebsch NJ, Deschler DG, et al. Proton beam radiation therapy for skull base adenoid cystic carcinoma. Arch Otolaryngol Head Neck Surg 2006;132(11):1242–1249.
10. Pitman KT, Prokopakis EP, Aydogan B, et al. The role of skull base surgery for the treatment of adenoid cystic carcinoma of the sinonasal tract. Head Neck 1999;21(5):402–407.
11. Harbo G, Grau C, Bundgaard T, et al Cancer of the nasal cavity and paranasal sinuses: clinico-pathological study of 277 patients. Acta Oncol 1997; 36:45–50.
12. Goepfert H, Luna MA, Lingberg RD, White AK. Malignant salivary gland tumors of the paranasal sinuses and nasal cavity. Arch Otolaryngol 1983; 109:662–668.
13. Bhattacharyya N. Factors affecting survival in maxillary sinus cancer. J Oral Maxillofac Surg 2003; 61:1016–1021.
14. Jones AS, Hamilton JW, Rowley H, et al. Adenoid cystic carcinoma of the head and neck. Clin Otolaryngol 1997;22: 434–443.
15. Lupinetti AD, Roberts DB, Williams MD, et al. Sinonasal adenoidcystic carcinoma: the M. D. Anderson Cancer Center experience. Cancer 2007;110(12):2726–2731.
16. Pommier P, Liebsch NJ, Deschler DG, et al. Proton beam radiation therapy for skull base adenoid cystic carcinoma. Arch Otolaryngol Head Neck Surg 2006;132(11):1242–1249.
17. Pitman KT, Prokopakis EP, Aydogan B, et al. The role of skull base surgery for the treatment of adenoid cystic carcinoma of the sinonasal tract. Head Neck 1999;21(5):402–407.
18. Rhee CS, Won TB, Lee CH, et al. Adenoid cystic carcinoma of the sinonasal tract: treatment results. Laryngoscope. 2006; 116:982–986.
19. Spiro RH. Distant metastasis in adenoid cystic carcinoma of salivary origin. Am J Surg 1997;174:495–498.

Figure 1.A Anterior rhinoscopy examination of left nasal cavity showing reddish nasal mass presented medial to inferior turbinate.

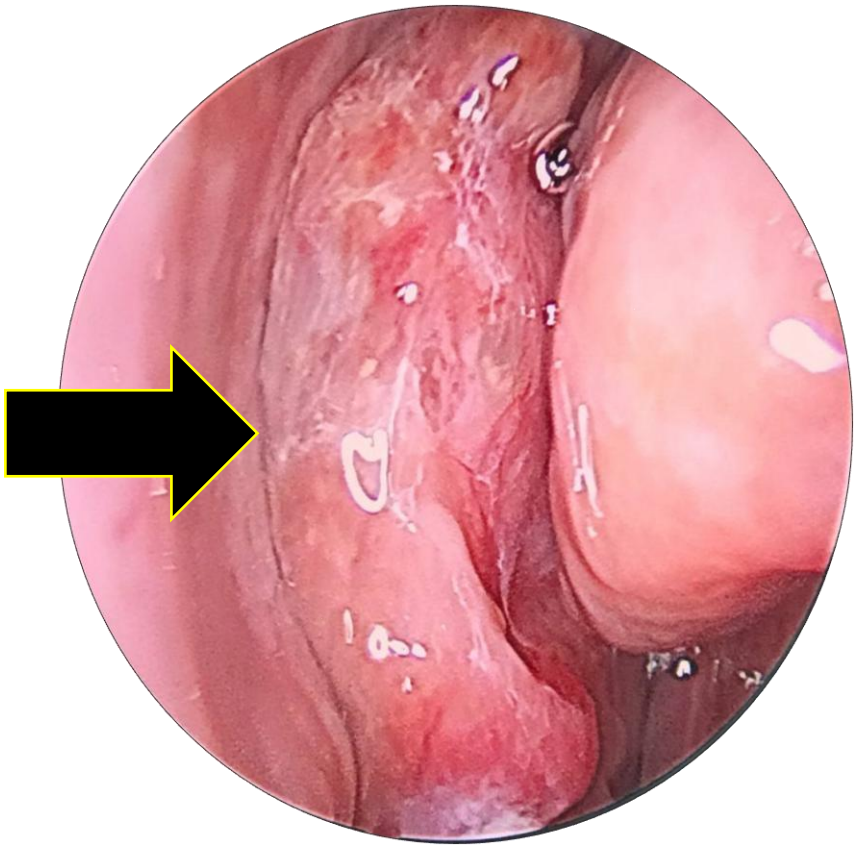


Figure 1B: Diagnostic nasal endoscopy shows nasal mass (arrow) occupying the left nasal cavity, lateral to septum and medial to inferior turbinate.

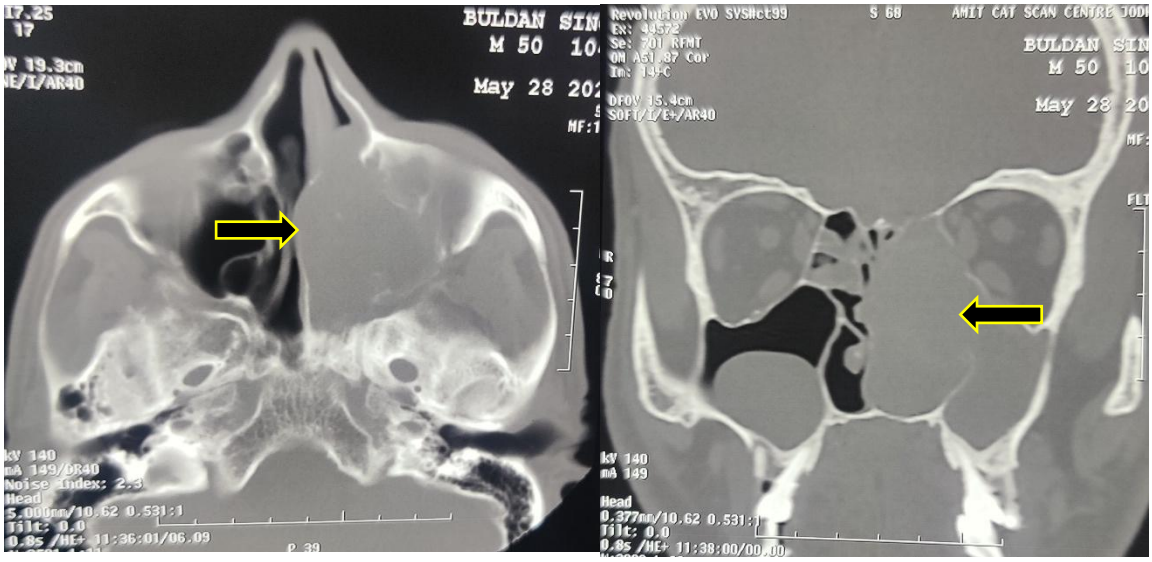


Figure 2: Axial computed tomography scan (Bone window) showing mass occupying the left nasal cavity obliterating the osteomeatal complex without any bone erosion

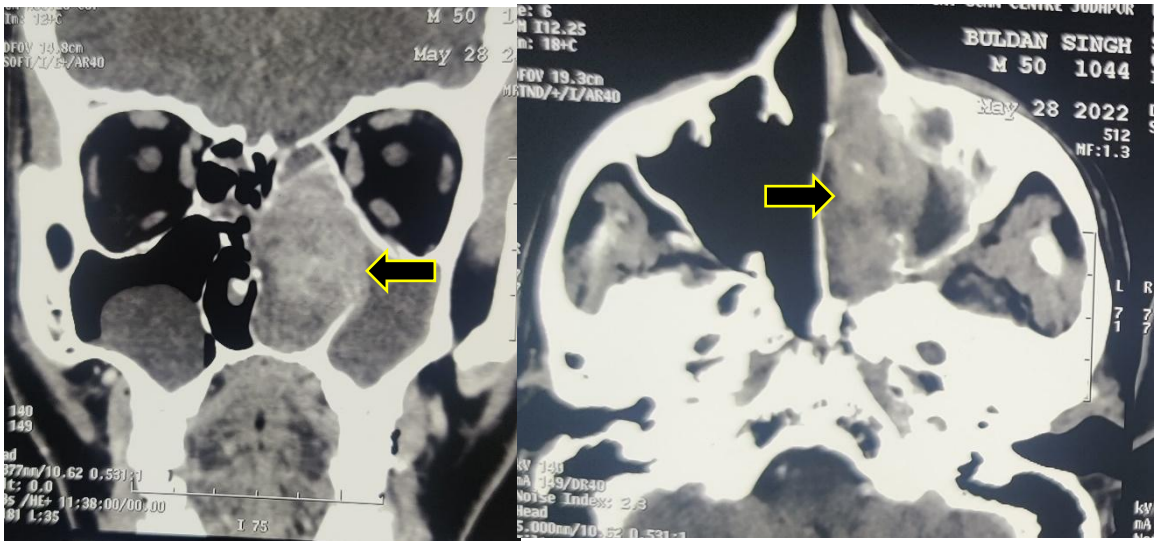


Figure 3: Axial + coronal computed tomography scan (soft tissue window) shows heterogeneously enhanced soft tissue mass occupying left side nasal cavity with retained section in left maxillary sinus.

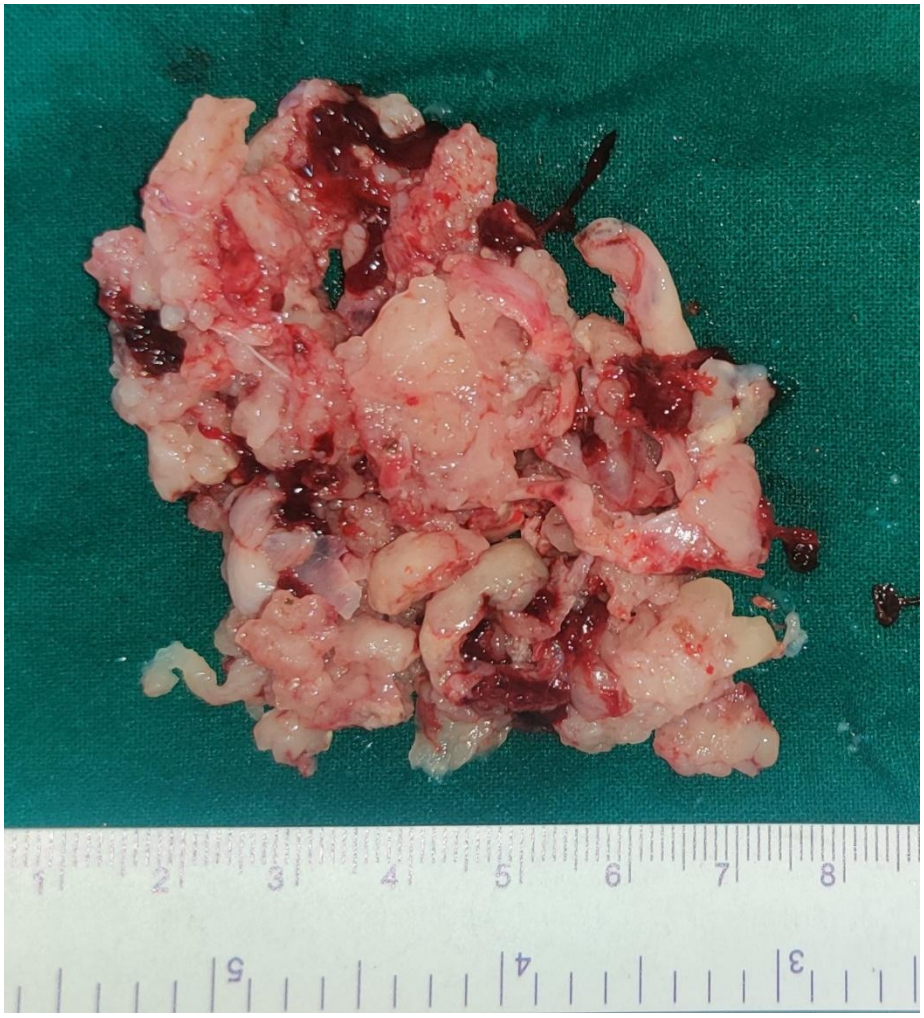
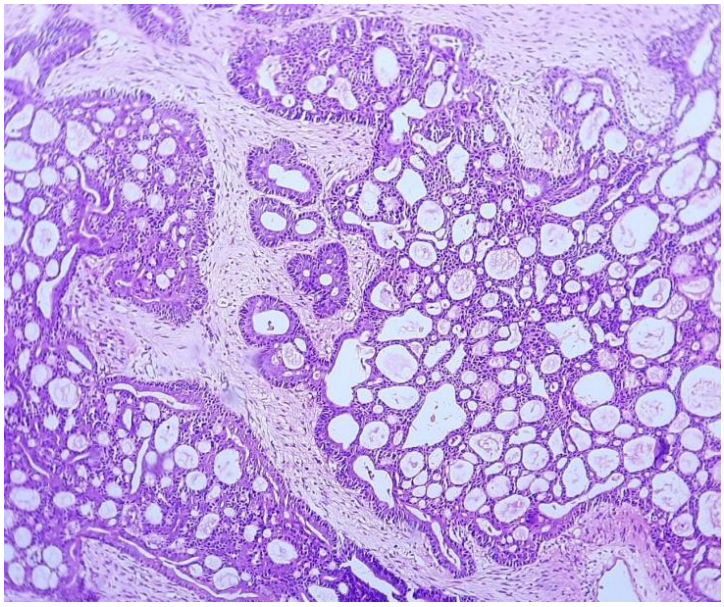
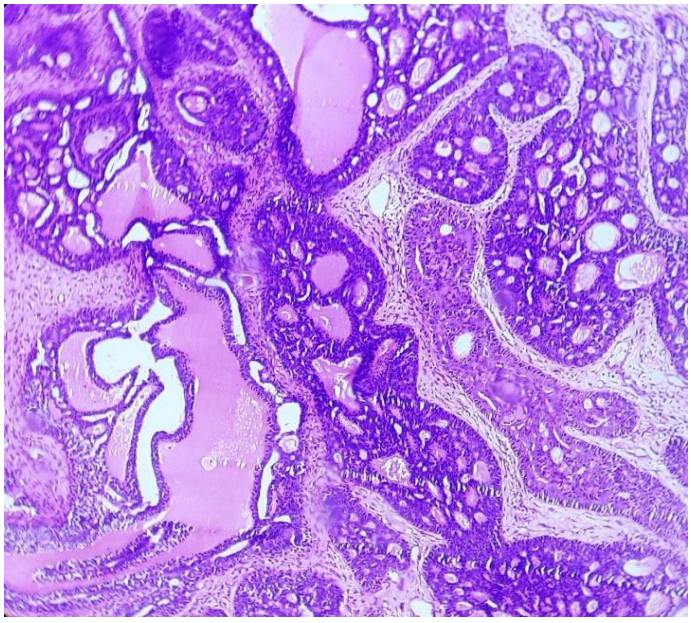


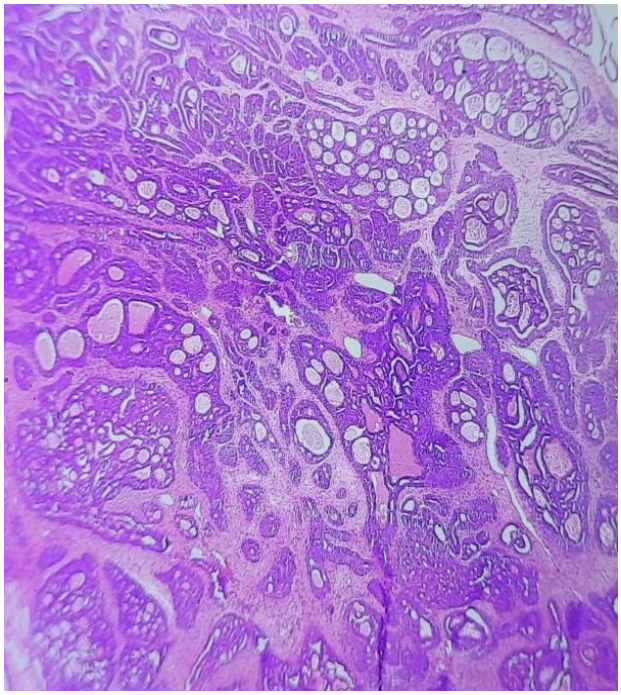
Figure 4: Specimen comprises of several grey brown soft tissue pieces together measuring 8 x8 x0.7cm.



(A)



(B)



(C)

Figure 5 (A,B,C): Histopathology slide comprises salivary gland tissue. The neoplasm has largely cribriform pattern with focal tubular areas without perineural invasion. Cribriform pattern areas show mainly myoepithelial cells with myxoid and hyaline globules. No anaplasia seen.