



Unusual Presentation of Adenoid Cystic Carcinoma of Submandibular Salivary Gland

KEYWORDS

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ABSTRACT Adenoid cystic carcinoma is a malignant neoplasm most commonly originating in salivary glands of head and neck region. Its occurrence elsewhere is rare and extension to thyroid even rarer. We report a 60 year old female who came to general surgery outpatient department with chief complaints of swelling in the left upper and in front of the neck later on investigation she was diagnosed to have features of adenoid cystic carcinoma in fine needle aspirate of the thyroid swelling and subsequently confirmed by histopathology.

Introduction

Adenoid cystic carcinoma accounts for approximately 10 % of all salivary gland tumours. It is the most common malignant tumour of submandibular gland and minor salivary glands. The tumour extends well beyond the visible and palpable limits of the salivary gland region. This infiltrative capacity is the hallmark of this carcinoma. Pain and spreading along nerve sheath is often noted. While adenoid cystic carcinoma is not an uncommon tumour in the salivary glands, its occurrence elsewhere is rare. Extension into the thyroid gland is extremely rare.

Case Report

A 60 year old female came to general surgery outpatient department with a history of swelling in the upper part and in front of the neck for the past 3 years. Patient has no difficulty in swallowing and difficulty in breathing. No history of loss of weight and, no history of loss of appetite, no history of trauma, no history of fever, no history of loss of taste sensation, no history of increased pain while eating food. On examination, the patient was conscious and oriented, her vital signs were temperature was 98.6f, pulse rate was 78/min, blood pressure was 120/80mmhg, on examination the swelling was oval in shape, not moving up with deglutition, no other visible swellings were seen, surface was irregular, skin normal, no local rise of temperature, no tenderness, carotid pulsations felt, swelling was bimanually palpable, no loss of functions of facial nerve. Patient was admitted and all the base line investigations were done with USG & FNAC.

Ultrasonography :

An enlarged, irregular shape, irregular borders, of size 4x5x5cm blurred margins, and a hypo echoic, homogeneous with a solid nodule. Arising from submandibular gland ,with one cervical left lymph node of size 8mmx8mm . Thyroid gland normal, with no infiltration or engulfing of and vessels.

Features suggestive of : submandibular gland tumor ? carcinoma

Fine needle aspiration (FNA) of the submandibular gland region :

Mixture of small, uniform, basaloid cells with high N/C ratios and metachromatic stroma. Showing cribriform structures. The chromatin appears coarse but uniform without

identifiable nucleoli, and nuclear pleomorphism is minimal . The background is populated by scattered naked nuclei .

Features suggestive of : Adeno cystic carcinoma

And the patient was prepared for surgery, all the base line investigations were done and was within normal limits, wide excision of the submandibular gland with fat and lymphatic's were dissected in submandibular triangle , by preserving the lingual nerve , hypoglossal nerve, facial vein, facial artery and marginal mandibular branch of facial nerve. The specimen was sent for histopathology.

Microscopic features of the excised specimen

Multiple sections examined show tumor tissue along with normal salivary gland tissue. The tissue is composed malignant cells arranged in lobules separated by fibrocollagenous stroma. Cribriform architecture is seen in most of the lobules. The tumor cells are basaloid with scant cytoplasm and enlarged hyperchromatic disease basophilic nucleus. The tumor cells are invading the nerve bundles.

Impression: features suggestive of adenoid cystic carcinoma.

Post operatively patient was stable and got discharged on 6th post op day, patient was sent for radiotherapy in the view of peri neural invasion . Patient is under follow up for last 6 months having no local or systemic complaint till now.

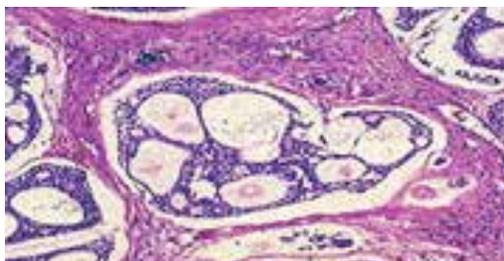
Discussion :

Adenoid cystic carcinoma is a slow growing but highly malignant neoplasm of the salivary gland, commonly affecting the minor salivary glands of head and neck region. Minor salivary glands are affected most by adenoid cystic carcinoma (40%), followed by the submandibular gland (18.2%) and the parotid gland (17.3%). The solid histological pattern is seen only in 21 percent of cases. Most individuals diagnosed with this disease are in their fourth decade of life, though a wide age range has been reported. The female and male ratio is approximately 3:2. Usual presentation is an asymptomatic slow growing mass, the absence of symptoms being responsible for late diagnosis. Here our patient was sixty years old female patient having a slow growing mass in left sided submandibular region without other symptoms. The three major histological pat-

terns of growth have been described: cribriform, tubular and solid. Combinations of the patterns are common. The prognosis of adenoid cystic carcinoma is greatly influenced by the pattern of growth, tubular pattern having the best prognosis while solid is known to have the worst prognosis. Adenoid cystic carcinoma has a relatively slow growth and infrequent lymph node metastasis but is well known for its perineural spread. In a study of 160 patients of adenoid cystic carcinoma by Fordice et al, disease specific survival was 89% at five years but only 40% at fifteen years. 5 Distant metastasis was the commonest cause of treatment failure. Lung is the most common site for distant metastasis, followed by liver. Spiro in his retrospective study of 196 patients followed up for 10 years reported some form of treatment failure in 68%, distant metastasis in 38%, and lung involvement either alone or in addition to other sites in 34%.¹² In cases of submandibular gland adenoid cystic carcinoma, surgical wide excision is very much possible and in T1 or T2 cases post operative radiotherapy may not be needed. Elective neck dissection is usually not done in N0 cases as lymph node metastasis is not common at all. Some authors prefer radiotherapy as a standard mode of treatment and especially in high risk cases.

Conclusion:

Adenoid cystic carcinoma (AdCC) is a very rare malignant tumour of salivary glands. The treatment should be aiming to eradicate the local disease and preventing distant metastases. A high index of suspicion is needed in diagnosing this disease early in order to have better prognosis and good quality of life



Figure(c): Histopathology slide

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Figure(A) : Pre operative picture



FIGURE(B) : Excision of submandibular gland