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Polymorphous adenocarcinoma mimicking pleomorphic adenoma: a diagnostic dilemma

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Abstract:

Polymorphous adenocarcinoma (PAC) is a rare salivary gland tumor, usually seen in minor salivary glands. It primarily affects adult females. Herein, we report an unusual case of polymorphous adenocarcinoma in a 72 year old male who presented with the non specific symptoms of swelling and mild pain below right lobe of ear similar to the pleomorphic adenoma. On Fine Needle Aspiration Cytology (FNAC), an impression of pleomorphic adenoma was given. Contrast enhanced computed tomography (CECT) of the neck was done, showed a relatively defined heterogenous enhancing soft tissue attenuated mass lesions suggesting likely neoplastic in nature, arising from parotid gland. Histopathological examination and immunohistochemistry (IHC) confirmed the diagnosis of polymorphous adenocarcinoma. Patient was conservatively managed and wide local excision was performed. Polymorphous adenocarcinoma should be included in the differential diagnosis of pleomorphic adenoma to avoid recurrence of the lesion.

Keywords: Polymorphous adenocarcinoma, Minor salivary gland, Mlignant tumors,Pleomorphic adenoma, Immunohistochemistry.

Introduction

Polymorphous adenocarcinoma is a rarely occuring malignant tumor of the salivary glands and almost exclusively involve minor salivary glands. It is the second most common primary malignancy occuring in the minor salivary glands after mucoepidermoid carcinoma, constituting 9 to 26.4% of all salivary

malignancy.[1] Most commonly it affects hard palate and soft palate minor salivary glands, followed by buccal mucosa, lip, tongue and major salivary glands.[2] It shows an indolent behaviour which often manifests as an asymptomatic slow growing mass in the oral cavity but can be painful and ulcerative.[3] It predominantly affects females than males with 2:1 ratio in the age range from 30-70 years.[4] Here, we present a case report of Polymorphous adenocarcinoma occuring below right lobe of ear in a 72 year old malewith a clinical diagnosis of pleomorphic adenoma.

Case Report

A 72 year old male patient presented to the ENT OPD with the history of swelling and mild pain in the right side of neck since 4 years. Initially swelling was small in size and remain static for 3 year and 6 months, but the size of swelling was suddenly increased in last 6 months. On examination, ulcerated swelling measuring 10X7 cm was present below the right lobe of ear which bleeds on touch. The swelling was firm in consistency, well circumscribed and tender. Vitals were stable and his past history was unremarkable.

Laboratory investigations showed hemoglobin 13 gm/dl. Other test findings such as total leukocyte count, differential leukocyte count and platelet count were within normal limit. FNAC was performed and showed an impression of pleomorphic adenoma.

Patient was referred to the radiology department with a probable diagnosis of pleomorphic adenoma. CECT of the neck was done which displayed a relatively defined heterogenous enhancing soft tissue attenuated mass lesions seen arising from right side parotid gland, suggesting likely neoplastic in nature.

A wedge biopsy was done and tissue sample was sent to the pathology department for histopathological examination and immunohistochemical study. Hematoxylin and Eosin (H&E) stained section showed an unencapsulated tumor with variable growth pattern such as cords, nests, tubules and papillaroid arrangement (Figure 1). Tumor cells showed mild nuclear pleomorphism and hyperchromasia alongwith evidence of infiltration (Figure 2). Mitotic count was low 1-2 per high power field. Areas of necrosis and congested blood vessels also seen. No perineural or lymphovascular invasion seen.

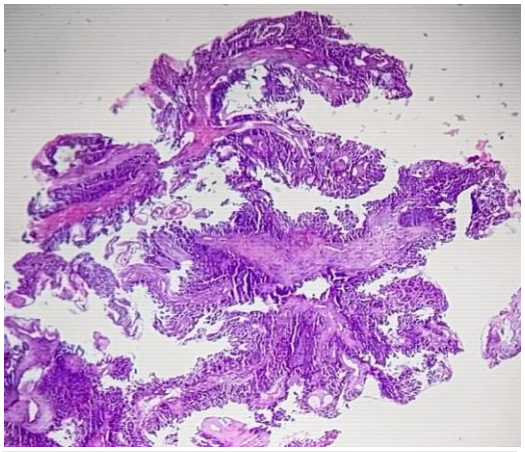

Fig. 1: Photomicrograph showing polymorphous growth pattern such as cords, nests, tubules and papillaroid arrangement.

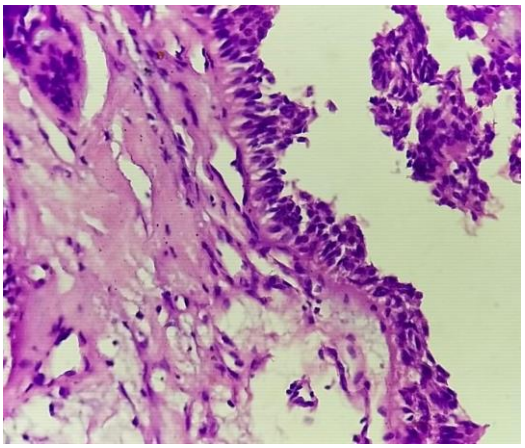

Fig. 2: Photomicrograph showing mild nuclear pleomorphism and hyperchromasia alongwith evidence of infiltration and few mitotic figures.

Additional immunohistochemistry studies revealed positive S-100 staining in tumor cells and patchy positivity of glial fibrillary acidic protein (GFAP) and p^{63} seen in tumor cells. A diagnosis of Polymorphous adenocarcinoma was made based on these pathological findings.

Patient was managed conservatively and complete surgical excision of the tumor was performed. The postoperative period was uneventful and got discharged after 5 days. On follow up, no recurrence was observed till date.

Discussion

Polymorphous adenocarcinoma is a rare diagnosis in a patient presenting as pleomorphic adenoma with non-specific symptoms. It is an uncommon but malignant subset of neoplasm occuring almost exclusively in the minor salivary gland.[5] PACs are though rare in major salivary glands, but they show

similar morphology those arising in minor salivary glands.[6] In this report, we discussed a case of PAC in a 72 year old elderly male patient who presented with ulcerated swelling and mild pain below right lobe of ear similar to the presenting features of pleomorphic adenoma. It usually manifests as an asymptomatic slow growing mass which may or may not be associated with pain, ulceration and bleeding. PAC patients are usually adults with the peak incidence in 6th to 7th decade.[5] It usually affects females than males with a male to female ratio of ranging from 1:1.02-1:2.[1]

FNAC and CECT imaging in our case was of not much helpful as the findings were suggesstive of pleomorphic adenoma and likely neoplastic respectively. Previously reported cases also revealed that polymorphous adenocarcinoma is difficult to distinguish from pleomorphic adenoma because they often show a very similar clinical features. Macroscopically, PAC shows firm, well-circumscribed and nonencapsulated mass alongwith areas of ulceration, necrosis and hemorrhage over the surface epithelium.[7]

Microscopically, PAC shows unencapsulated tumor with highly variable growth pattern in different areas of the neoplasm, with evidence of infiltration. Areas of necrosis and haemorrhage also seen.[8] Pleomorphic adenoma is a non-infiltrative tumor with dual cell differentiation.[6] Sometimes, they also create a dilemma in the histological analysis despite their characteristic morphology and only diagnosed after immunohistochemical analysis. So, immunohistochemical study is a useful tool in diagnosing PAC. Positive staining for GFAP in a mesenchymal like cell population adjacent to epithelial nests is commonly observed in Pleomorphic adenoma but not in PAC.[9] PAC stains diffusely with S-100 protien and epithelial membrane antigen.[10] In our case, immunohistochemistry studies showed positive S-100 staining in tumor cells and patchy positivity of GFAP and p^{63} seen in tumor cells, suggesting the final diagnosis of PAC.

Polymorphous adenocarcinoma is a low grade indolent neoplasm with good prognosis which rarely associated with invasion and metastasis. Complete surgical excision with clear margins is the most common recommended treatment for these slow-growing tumors. In case of recurrence, radical excision is required.[7]

Conclusion

Polymorphous adenocarcinoma is a rare malignant tumor of salivary gland which can clinically and morphologically mimic benign neoplasm. Therefore, one should be aware of this rare entity due its malignant potential. A thorough immunohistochemicalworkup is required to diagnose this entity.

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