



Case Report:

Polymorphous low grade adenocarcinoma-an unusual presentation

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Abstract:

Polymorphous low-grade adenocarcinoma (PLGA) is a neoplasm that occurs frequently in the mucosa of the soft and hard palates, in the buccal mucosa and in the upper lip and is very rare within the nasopharynx. We present a case of PLGA, which presented as a nasal polyp.

Key Words: Polymorphous low-grade adenocarcinoma, minor salivary gland neoplasm, nasal polyp

Introduction:

Polymorphous low grade adenocarcinomas (PLGA) are minor salivary gland neoplasms with a predilection for intraoral sites. Extraoral location of the tumor is rare and the nasal tumors are less than 1%.^[1] It generally involves the palate, but it has also been reported in the base of the tongue, upper lip, buccal mucosa, tonsil, and retromolar pad.^[2] So far, only 3 cases have been reported, where PLGA presented as a sinonasal mass.^[3,4] Histologically, PLGA resembles pleomorphic adenoma, benign mixed tumor and to an extent, to adenoid cystic carcinoma. We present a case of PLGA, which presented as a nasal polyp.

Case Report:

A male patient presented to the Dept. of ENT with a history of nasal obstruction and a single episode of epistaxis. There was no history of an upper respiratory tract infection or trauma. Nasopharyngoscopic examination revealed a polypoidal structure in left nasopharynx. No cervical lymph node enlargement was found. The FNAC of the polypoidal structure showed cellular smears. The cells had high N/C ratio, scanty cytoplasm, nuclei of variable chromatin density and inconspicuous to conspicuous nucleoli. Beaded fragments of hyaline stroma were present between cell clusters.[Fig. 1] A diagnosis of adenoid-cystic carcinoma was made. Polypectomy was done and sent for histopathological examination. HPE revealed mildly pleomorphic plump columnar cells arranged in tubular, cribriform, solid and fascicular patterns. No area revealed papillary configuration. Mitotic activity was low. Basement membrane material was found associated with the tumor cells. Peripherally the tumor cells revealed infiltrative pattern with foci of neurotropism[Fig 2], suggesting a diagnosis of Polymorphous low grade adenocarcinoma.

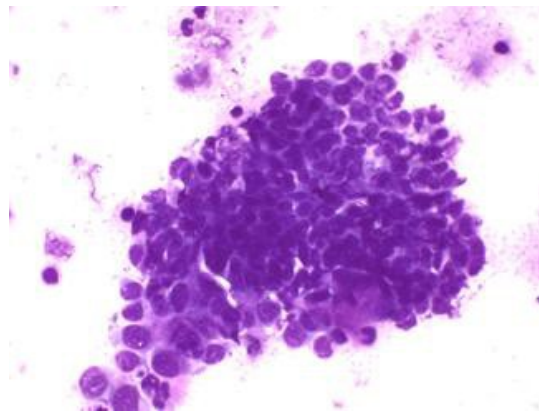


Figure-1: Microphotograph showing pleomorphic tumor cells with interspersed beaded fragments of hyaline stroma present between cell clusters.(x400,Giemsa)

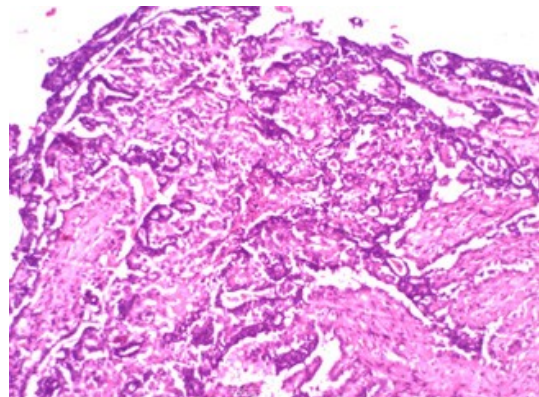


Figure-2: Microphotograph showing mildly pleomorphic plump columnar cells arranged in tubular, cribriform, solid and fascicular patterns along with basement membrane material.(x400,HPE)

Discussion:

Since the original description of these tumors, PLGA has been recognized as a distinct salivary gland tumor that has a predilection to occur in the minor salivary glands and is associated with slow growth and indolent biology. PLGA occurs over a wide age range but does not seem to occur in the first or second decades of life. There is a nearly 2:1 female to male ratio for the patients. Clinically, wide age range has been described (23 to 94 years). PLGA in women tends to occur at a slightly younger age (56.6 years) compared with male patients (59.7 years), but there is no statistically significant difference. The typical clinical presentation of PLGA is that of an asymptomatic mass located within the oral cavity. Rare intraosseous cases have been described. Nasal cavity and nasopharyngeal involvement in PLGA is rare (1% and 0.5%). To date only few cases have been reported in nasal cavity. Clinical symptomatology ranges in duration from a few days to 40 years, with an average length of symptoms of 27 months. Patients who present with pain, bleeding, or ulceration do not have more aggressive disease nor are they more prone to develop recurrences. Grossly, the tumor is usually unencapsulated, well circumscribed, lobular and firm. Microscopically, different architectural patterns can be seen in various areas. The most common patterns of growth included tubular, trabecular, solid, and cribriform patterns, with a focal papillary pattern identified less frequently. Myxoid change in the background may also be seen. In contrast to the architectural polymorphism, the nuclei are uniform and bland with absent or negligible mitoses.^[5]

The differential diagnoses of PLGA are adenoid cystic carcinoma and pleomorphic adenoma. Both adenoid cystic carcinoma and pleomorphic adenoma can have similar architectural patterns; however, the cells in ACC tend to be smaller with hyperchromatic nuclei and coarser chromatin. Mitoses are numerous. The infiltrative growth pattern and neurotropism are an important diagnostic clue favouring PLGA, when the differential includes pleomorphic adenoma. Although, the tumor has variable architectural patterns, the cells have minimal cellular pleomorphism. Local recurrences can occur. Rarely regional lymph node metastasis and distant (lung) metastasis have been documented. PLGAs chiefly involve the minor salivary glands. Only few cases involving the major salivary glands have been reported so far..

A definitive diagnosis of PLGA is established by detecting this characteristic histological feature of the resected tumor. The patient has been followed-up without evidence of recurrence and did not undergo radiation therapy. We suggest that radiation therapy may be reserved for patients with recurrence and regional metastasis.

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