PJMS- Volume 3 Number 1: January-June 2013

Case Report

Multinodular presentation of recurrent pleomorphic adenoma of submandibular salivary gland

Bothale Kalpana A[1], Mahore Sadhana D[2], Wilkinson Anne R[1]

[1]Associate Professor,
[2]Professor and Head
Department of Pathology,
NKP SIMS&RC, Digdoh Hills,
Hingna Road, Nagpur -40019.
kalpana_bothale@yahoo.co.in

Abstract :

A case of recurrent pleomorphic adenoma of salivary gland in an 18-year-old girl is reported herewith. The reported recurrence rate is less in submandibular gland. This case is reported for four recurrences within a short duration of five years. Multiple tumour masses showing abundant myxoid stroma is an alarming feature for recurrence.

Keywords: Recurrent pleomorphic adenoma, Submandibular gland.

Introduction:

The submandibular gland is the second most common site of pleomorphic adenoma after the parotid gland. The recurrence rate of pleomorphic adenoma in submandibular gland is less as compared to parotid gland. Majority of the patients have only one recurrence. Multiple recurrences are rarely seen in pleomorphic adenoma of the submandibular gland.

Case History:

An eighteen -year- old female patient presented with recurrent right neck mass of 2 years duration without any associated constitutional symptoms. She had undergone surgical resection thrice for the similar neck mass earlier which was diagnosed as pleomorphic adenoma of submandibular gland. Previous surgeries were done outside. On clinical examination, firm nodular mass was palpated measuring 7cm x 3cm in size. The patient was subsequently admitted for surgical excision of the recurrent submandibular tumor mass. The tumor mass was excised and sent for histopathological examination.

Pathological findings:

Gross Examination: The submandibular tumor consisted of multiple discrete nodules, varying in size from 2 mm. to 1 cm. in diameter. They were unencapsulated, grayish-white with fine nodularity on the external surface. These nodules were embedded in adipose tissue. The whole tumour mass measured 6.8cm x 2.8cm x 2.0cm. The cut surface showed multinodular appearance. The nodules were well circumscribed and discrete. There were no areas of hemorrhage and necrosis (Figure 1).

Microscopic appearance: Sections revealed multiple, discrete, circumscribed, unencapsulated masses embedded in fibroadipose connective tissue. They were composed of epithelial, myoepithelial cell elements and abundant stroma (Figure 2). The epithelial and myoepithelial cells were arranged in sheets and clusters with scanty to moderate amount of cytoplasm and round to oval uniform nuclei. Overall cellularity was less (hypocellular). At places keratin- like material was seen. The stroma was abundant and mostly myxomatous or mucinous. Hyalinization was seen in few areas. Foci of microcalcification and multinucleate giant cells were also present. The histopathology report was given as recurrent pleomorphic adenoma (RPA) of right submandibular salivary

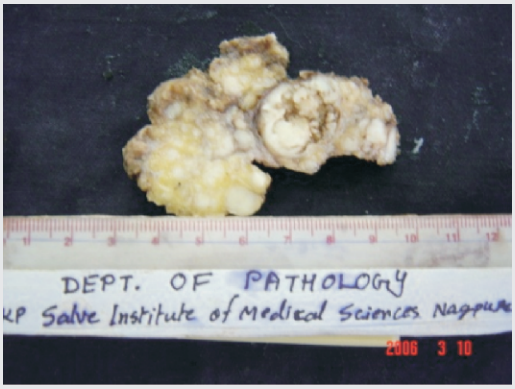

Figure 1: Cut surface of the tumour mass with multiple grayish white nodules surrounded by adipose tissue

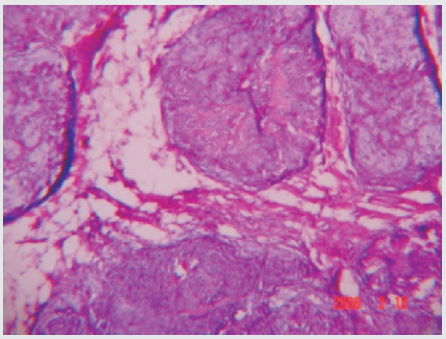

Figure 2: Photomicrograph showing multiple tumour masses composed of epithelial-myoepithelial cell elements and abundant myxoid stroma (H&E, X40)

Discussion:

Reports of the recurrence rate of pleomorphic adenoma (mixed tumor) of salivary gland vary greatly. Recurrence rates from 0 to 50% have been published for both major and minor salivary glands. Pleomorphic adenomas are slowly growing

neoplasms. To fully appreciate their potential for recurrence, a long term followup is necessary. Krolls SO and Boyers RC (1) studied 100 cases of mixed tumors of the salivary gland. Followup information was obtained on 89 patients. Out of these 89 patients, 74 mixed tumors were located in parotid, 11 in submandibular and 4 in minor salivary glands. 36 recurrent tumors in parotid, 2 in submandibular and 1 in minor salivary gland were noted. They reported one to five recurrences. Majority (53.9%) of the patients had only one recurrence. Only 5.1% had five recurrences. The shortest time interval between the initial surgical procedure and the first recurrence noted is less than a year and the longest is 21 years.

Yasumoto et al (2) reviewed clinical and imaging files of 15 patients with pathologically confirmed RPA of head and neck region (lacrimal gland, cheek, palate, parotid gland and submandibular gland). They noted that recurrence generally occur late with an average interval of 14.9 years between initial surgery and recurrence. The recurrence rate is less than 10% in submandibular gland versus 50% for the parotid gland, most likely because invariably the whole submandibular gland is excised for submandibular mixed tumour. High recurrence rate in parotid may be associated with inadequate initial surgical procedure (1). Naelm F et al (3) have studied 112 patients with pleomorphic adenoma of salivary gland (94 in parotid glands, 11 in submandibular and 7 in minor palatal salivary glands). Of the recurrent lesions 61% were hypocellular and 11% were hypercellular. They came to the conclusion that incomplete resection, hypocellularity and incomplete encapsulation are the major factors in recurrence. Koral K et al (4) analyzed medical records of the patients with RPA of the parotid gland. Before second surgery MRI has been performed in 15 patients. Multiple lesions are noted in 73% of patients. Their findings suggest that multiplicity of lesions is a reliable diagnostic indicator of recurrent disease. Clinical and morphological analysis of 21 cases of RPA of salivary gland has shown that recurring tumors are characterized by multinodular pattern. Myxoid component predominates in the tumour (5).

Adequate surgical resection yields an acceptable local control rate in patients with RPA of parotid gland. Postoperative radiation therapy may improve control in patients at high risk for another recurrence (6). Inadequate resection is predictor of local recurrence. Postoperative radiotherapy after inadequate resection appears to decrease the risk of recurrence (7,8). Laskawi R et al (9) studied surgical management of benign tumors of the submandibular glands. The first operation should extirpate the entire gland to minimize the risk of recurrence.

There is a risk of developing carcinoma ex pleomorphic adenoma in RPA. Most of these tumors will have malignant epithelial component but not malignant stromal component. Malignant transformation of these tumors takes up to 20 years or more (10). RPA occurs more often in younger patients and women. Surgical treatment should be individualized, taking into consideration the extent of the previous surgery and the type of recurrence (11). The management of RPA is a major challenge for the clinician. RPAs, particularly multinodular tumors, are prone to new recurrences especially when treatment of the initial tumor is performed according to currently accepted standards. MRI is considered to be the best tool to delineate the extension of the lesion, although it can miss microscopic nodules. Options for management include pure observation, not only for the elderly or infirm, but also in cases of small lesions until they begin to grow. When surgery is elected, it should be tailored to the single patient, because even if on one side a limited local excision is considered acceptable, on the other side a total or extended parotidectomy may be inadequate to control an RPA of parotid gland adjacent to the nerve. In these cases, facial nerve resection and reconstruction must also be considered and discussed with the patient in the preoperative counseling. Postoperative radiotherapy is an option, particularly for older patients for whom the risk of inducing other malignancies is considered to be low. Followup for patients treated for RPA should be done with regular ultrasonography for the lifetime of the patient and with magnetic resonance in selected cases. All patients should be informed about the possibility of the need for multiple treatment procedures.

References:

1) Krolls SO, Boyers RG. Mixed tumors of salivary glands-Long term followup. Cancer 1972; 30(1):276-281.
2) Yasumoto M, Sunaba K, Shibuya H, Kurabayashi T. Recurrent pleomorphic adenoma of the head and neck. Neuroradiology 1999; 41(4), 300-304.
3) Naelm F, Forsberg MI, Walsman J, Coulson WF. Mixed tumors of the salivary glands: Growth pattern and recurrence. Arch Pathol Lab Med 1976; 100(5):271-5.
4) Koral K, Sayre J, Bhuta S, Abemayor E, Lufkin R. Recurrent pleomorphic adenoma of the parotid gland in pediatric and adult patients: Value of multiple lesions as a diagnostic indicator. Am J Roentgenol 2003; 180(4):1171-4.
5) Laniukh SV, Kremenetskaia LE, Ssipkova TP. Recurrence of pleomorphic adenoma of the salivary glands. Stomatologiia (Mosk) 1990; 69(2):40-3.
6) Carew JF, Spiro RH, Singh B, Shah JP. Treatment of recurrent pleomorphic adenoma of the parotid gland. Otolaryngol Head Neck Surg 1999; 121(5):539-42.
7) Buchman C, Stringer SP, Mendenhall WM, Parsons JT, Jordan JR, Cassisl NJ. Pleomorphic adenoma: Effect of tumor spill and inadequate resection on tumor recurrence. Laryngoscope 1994; 104(10):1231-4.
8) Rowley H, Murphy M, Smyth D, O'Dwyer TP. Recurrent pleomorphic adenoma: Uninodular versus multinodular disease. Ir J Med Sci 2000; 169(3):201-3.
9) Laskawi R, Ellies M, Argiebe C, Schott A. Surgical management of benign tumors of the submandibular gland: A followup study. J Oral Maxillofac Surg 1995; 53(5):506-8.

10) Fowler MH, Fowler J, Ducatman B, Barnes L, Hunt JL. Malignant mixed tumors of the salivary gland: A study of loss of heterozygosity in tumour suppressor genes. Modern Pathology 2006; 19: 350-355.

11) Szwedowicz P, Osuch-Wójcikiewicz E, Bruzgielewicz A, Checiski P, Nyckowska J, Grotthuss M. Management of recurrent pleomorphic adenoma of the parotid gland. Otolaryngol Pol 2010; 64(7):50-4.