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Case Report

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Inverted duct papilloma of mid oesaphagus with unusual histological features

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Abstract

One of the unusual sites for inverted ductal papilloma is oesaphagus. The tumor has distinct histological features which differentiate it from other similar lesions. The case under discussion is extraordinary in many respects; not only because of unusual location but also for other features like malignant transformation; etc not reported even in sites where this is more common.

Keywords: Esophagus, Inverted duct papilloma, Carcinoma in situ changes.

INTRODUCTION

Inverted duct papilloma (IDP); one of the three papillomatous conditions; arising from minor salivary glands, is a rare clinical disorder; none of which is reported in major salivary glands (except for one exception). Most of them are noticed in upper aero digestive tract; particularly involving buccal mucosa and lips [1]. In 1981, Batsakis reported the first case under the title of epidermoid papillary adenoma. The more descriptive term for this condition was coined by White *et al* [2] and later Ellis reported the largest series of 18 cases. It was Stacey Mills [2] who succinctly described this tumor arising from duct like structure lined by basaloid cells with radial orientation and converging upon and opening into the central cystic cavity. This case report is unique in more than one sense because the site previously unreported and also because of unusual histological features, like koilocytic atypia, extensive metaplastic changes with dysplasia and noncontinguous carcinoma in situ type of transformation.

CASE REPORT

A 50 years old male reported for progressive dysphagia of 3 months duration. Endoscopic examination revealed in mid oesaphagus an ulcerative lesion of 1.5cm diameter. There was no exophytic lesion. The endoscopic biopsy was performed and sent for histological assessment. On low power view this tumour appeared as a cup shaped depression endophytically with close alignment with surface mucosa, and dipping down with circumscription. The central cavity; a classical features in IDP, was surrounded by radially oriented epithelial cells (Figure-1) with the luminal side being lined by metaplastic pseudo stratified columnar cells. The remnants of columnar mucinous cells are rarely encountered. Apart from extensive of koilocytic changes involving squamous lining (Figure-2) the neoplastic cells oriented around central cavity exhibit varying degrees of dysplastic alteration. Foci of carcinoma in situ were seen discontinuously in the lining mucosa (Figure-3). Because of the meagre biopsy material, elaborate immunochemical staining was not possible except for pan cytokeratin and P 63 immunostains which were positive in this case (Figure-4).

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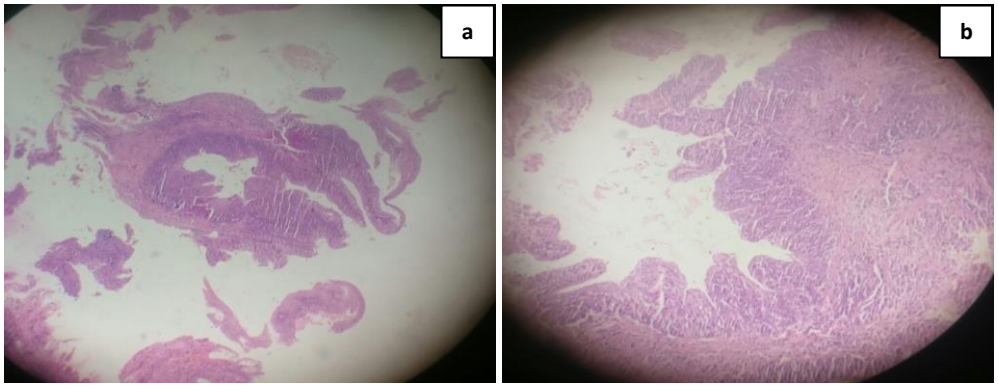

Figure 1: a and **b** Showing central crater surrounded by converging neoplastic cells (low and high power)

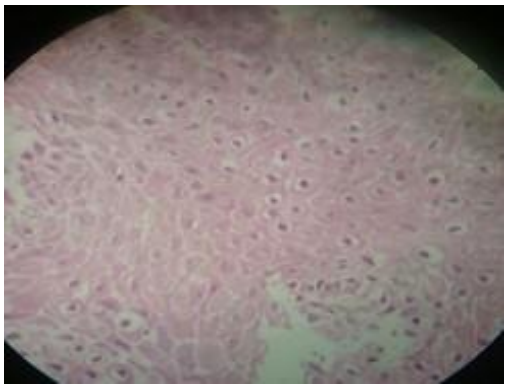

Figure 2: To show koliocytosis involving oesophageal mucosa

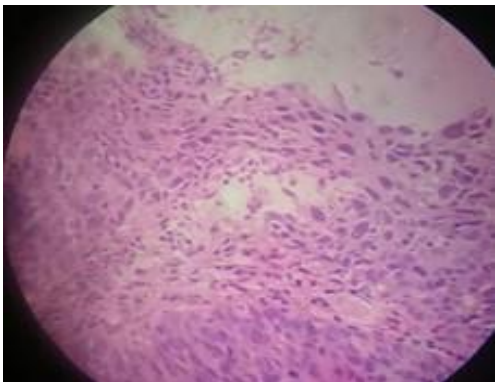

Figure 3: Carcinoma in situ arising from metaplastic cells lining luminal cells

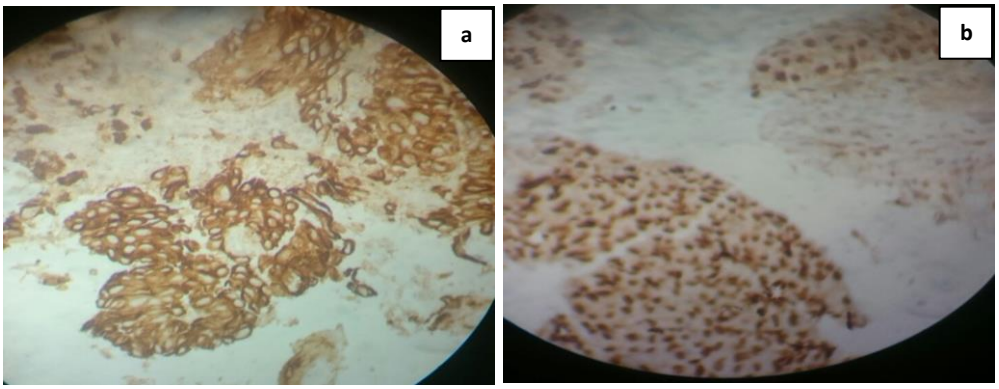

Figure 4: Staining reaction with pancytokeratin **(a)** and P 63 **(b)** immunostains

DISCUSSION

According to Glickman and Odze [3] tumours of minor salivary glands in oesaphagus are rare. Intraductal schenederian papilloma (ISP) closely resembles IDP but can be differentiated from it by its large size and non circumscribed growth pattern; growing in a haphazard manner without a central crater. The immunochemistry of luminal cells differs from these on the basal side as proved by Japanese authors [4] Ck 14 is predominantly expressed in basal cells while AM 5.2 and CK.7 stain luminal cells. In spite of the differing nature and the intensity of staining, there is a fundamental unity of both cells; since in both of them CK 19 is expressed strongly in all the tumor cells. The limited immunochemistry, done in our biopsy specimen shows positivity of same intensity in the lining of pseudostratified columnar cells throughout.

The case under discussion differed from all the reported cases in many respects: the extensive koilocytic changes affecting the lining squamous mucosa indicates substantial HPV infection. It is known from polymerase chain reaction, 33.3% of sinonasal polyps are due to HPV infection while in 9 cases of oral IDP tested for HPV DNA through hybridization techniques, HPV types 6 and 11 are identified in 23.3% of the 9 cases of IDP [5]. According to Kuhota *et al* [6] no cases of malignant transformation have been reported with IDP and hence many authors advocate local excision of the tumour as curative. The case under discussion proves malignancy is possible in IDP; if complicated by HPV infection and this is a reality we have to recognize when one is dealing with a case of this nature.

CONCLUSION

This case report shatters the belief that inverted duct papilloma's occur only in upper aero digestive tract and second misconception that it behaves in a benign manner. Since it is a treatable surgical condition, it is imperative that is unusual histopathology and its origin from excretory duct of minor salivary gland requires much better appreciation. The invagination of radially arranged epithelial cells opening into a central crater may require serial sectioning occasionally.

Conflict of Interest

The authors declare no conflict of interest.

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