

FOCAL EPITHELIAL HYPERPLASIA A NEWLY DISCOVERED DISEASE IN NORTH WEST FRONTIER PROVINCE OF PAKISTAN

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INTRODUCTION

The term focal epithelial hyperplasia was introduced in 1965 to describe certain multiple nodular elevations of the oral mucosa observed among American Indians in New Mexico, USA and Mato Grosso district in Brazil¹. Since then it has been reported from several other countries including Iraq, Israel and South Africa². We report two cases of this disorder from North Western Frontier Province of Pakistan.

CASE 1

An Afghan boy, aged 10 years, reported to the dermatology unit of Postgraduate Medical Institute, Lady Reading Hospital, Peshawar with complaints of asymptomatic multiple nodular lesions in the oral mucosa (Figure 1).



Figure 1. Typical oral lesions in focal epithelial hyperplasia.

He was referred from orodental department for evaluation of any systemic cause of his illness. The father had noticed a few nodular elevations on the lower lip of the child 5 years ago, which had progressively increased in size and number. New lesions started appearing on the upper lip, sides of the tongue, hard palate and other areas of the oral mucosa. The past and family history were not contributory. On clinical examination numerous rounded to polygonal fleshy elevations were present on labial mucosa, sides of the tongue, at the junction between hard and soft palate and hard palate itself. Their size ranged from 0.1 to 0.5 cm in diameter. They were soft to firm in consistency and nontender. Indirect laryngoscopy was normal. All the relevant laboratory investigations, i.e., complete blood picture, urinalysis, chest x-ray, thyroid function tests and scan were within normal limits. VDRL test was negative. A biopsy was taken which showed epithelial hyperplasia with parakeratosis, acanthosis, papillomatosis, elongation and anastomosis of rete ridges (Figure 2).

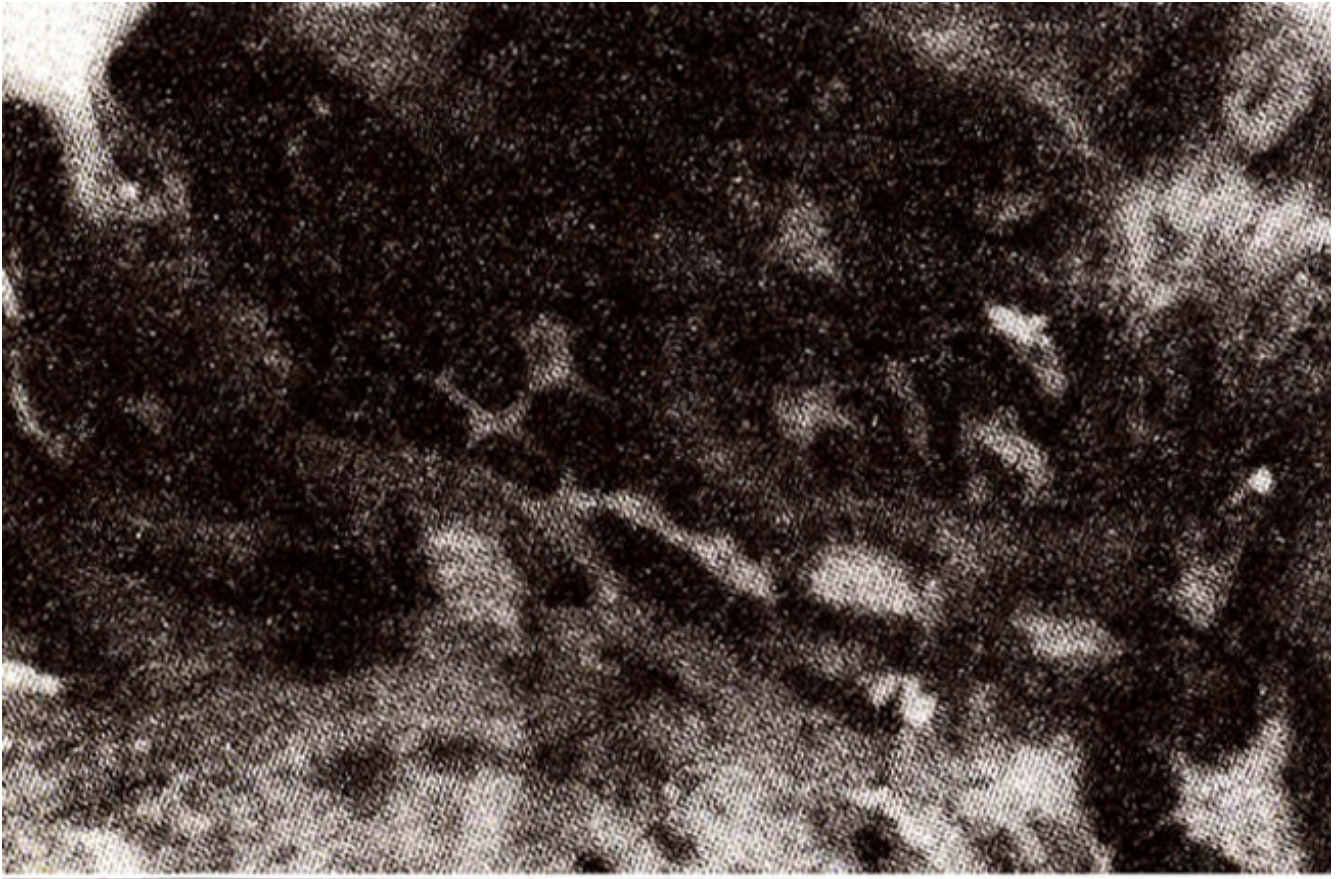


Figure 2. Marked acanthosis, elongation and fusion of rete ridges (1x40).

A diagnosis of focal epithelial hyperplasia was made. The lesions were treated with cryosurgery in four different sittings at two weeks intervals. The response to this modality was good and the lesions did not recur until 6 months of follow-up.

CASE 2

A ten year old female child from Chitral reported with asymptomatic papulonodular elevations on the upper and lower lips causing cosmetic concern. A few papular elevations on the lower lip of the child were noted 2 years before which subsequently increased in number and size and also started appearing on the upper lip. No family history of such problem was present. On clinical examination numerous soft to firm, white to flesh coloured papules were seen on the lower and upper labial mucosa. A few scattered lesions were also present on the tongue and other parts of the oral mucosa whose size ranged from 0.1 to 0.5cm in diameter. The lesions on the lower lip were somewhat whitish and slightly hyperkeratotic. They were less noticeable on stretching the oral mucosa. Indirect laryngoscopy did not reveal any such lesions in the larynx. All the relevant investigations were within normal limits.

Histology of an oral papule showed epithelial hyperplasia with marked acanthosis, papillomatosis, elongation and fusion of rete ridges. Some vacuolated cells were also present in the epidermis. A diagnosis of focal epithelial hyperplasia was made. The lesions on the lower lip were treated with cryosurgery but the patient was lost to follow-up.

DISCUSSION

The first complete description of focal epithelial hyperplasia was made by Archard, Heck and Stanley³ who, described the condition as nodular masses on the oral mucosa usually from 1 to 5 mm in diameter and often so numerous that they appear sprinkled over the mucosal surfaces. The nodules are soft and

elevated with surface like the adjoining oral mucosa or have a flat whitish surface¹. This condition predominates in children and young adults, 3 to 18 years of age. The most commonly reported site is the lower labial mucosa. The floor of the mouth and palatal mucosa are conspicuous for their absence of lesions⁴. Both of our cases fulfil these criteria except that our first case had involvement of the hard palate also. Familial occurrence of this disease has been described⁵ but no such association was found here. The focal epithelial hyperplasia occurs with an unusual racial and geographic distribution appearing in 30% of the Eskimos of Greenland and in 3% of the Indians of North, Central and South America. It rarely occurs in the rest of the world population⁶ which could be in part due to ignorance among clinicians about this newly described entity. It may be useful to carry out a survey to determine the prevalence of this condition in our population. There has been convincing recent evidence now that a type of human papilloma virus may be the major factor in the causation of this disease⁶⁻⁸. Papova virus group particles have been identified by electron microscopy⁸ and immunoperoxidase techniques⁶. More recent studies with DNA hybridization techniques suggest UPV 13^{9,10} and HPV 32¹⁰ to be associated with focal epithelial hyperplasia. A high risk HPV type 16 DNA¹¹ has also been found in one case of focal epithelial hyperplasia. The negative family history and the clinical course of the disease in both of our cases support this notion, however the possibility of genetic factors in its causation cannot be ruled out^{1,5,6}. The disease is said to regress spontaneously². This does not appear to be the case in our patients as both of them had a long history of slowly progressive disease. The possibility that the disease may have a different course in our patients cannot therefore, be ruled out. The main morphological changes described so far are epithelial hyperplasia, parakeratosis, acanthosis, elongation and anastomosis of rete ridges^{1,7,12}. Dyskeratosis, epithelial atypia and inclusion bodies have not been found⁹. Our histopathological findings correlate well with those described. However, basal liquefaction (an occasional finding⁹) was not seen in our cases. The treatment modalities described are electrocoagulation, excision and shaving^{4,6}. Cryosurgery has been described to be ineffective⁴. It appeared to be very effective in our first case. More studies, however, are needed to determine the effectiveness of this form of treatment.

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