CASE REPORT

Intramucosal nevus of buccal mucosa in a male child

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SUMMARY

Nevus (mole or birthmark) is a benign tumour of skin and mucosa characterised by the presence of melanin-producing, neuroectodermally derived cells, which can be light to dark brown, reddish brown, blue or flesh coloured. It varies in shape from oval to round. Oral melanotic nevi are uncommon oral lesions causing focal pigmentation. They were found only in 0.1% of population in a large survey. Nevi can be acquired over time or congenital. Acquired nevi are considered benign neoplasms whereas congenital nevi are hamartomas. They are located usually on the palate but less commonly on buccal mucosa, gingiva and lips. This article presents a case report of an intramucosal nevus of buccal mucosa in a 5-year-old boy with its surgical removal.

BACKGROUND

Nevi are benign proliferations of nevus cells either in the epithelium or in the subepithelial stroma. They are best categorised as hamartomas rather than true neoplasms. Nevi of the oral cavity are usually called mucosal melanocytic nevi or intramucosal nevi. In 1943, Ackermann and Field reported the first documented case of an oral nevus. Pigmented nevi are uncommon lesions in the oral cavity and were found only in 0.1% of patients in a large survey (King *et al*, 1967).[1]

Oral pigmentation may be exogenous or endogenous in origin. Exogenous pigmentation is commonly due to foreign-body implantation in the oral mucosa. Endogenous pigments include melanin, haemoglobin, haemosiderin and carotene. Pigmented lesions caused by increased melanin deposition may be brown, blue, grey or black; depending on the amount and location of melanin in the tissues.[2] They can be seen in persons of all ages and are usually less than 5 mm in diameter. When seen intraorally, they are most commonly observed on the hard palate. Clinically pigmented nevi are asymptomatic, flat or slightly elevated spot or plaque of brown or brown–black colour.[3]

The most common intraoral nevi are intramucosal (55%) and blue (32%) nevi, while junctional (5%) and compound (6%) nevi are relatively rare.[4] Clinically, most nevi present as small (<0.5 cm), raised (more than 80%), pigmented lesions (about 85%) that range from 0.1 to 0.5 cm in size, but 20% are flat, macular lesions. Occasional lesions lack pigmentation (about 15%) and may appear as excrescences of normal colour. In this paper, we have reported a case of intramucosal nevus of buccal mucosa which is rare with regard to the site of presentation, age and gender with its management.

CASE PRESENTATION

A 5-year-old boy reported to our hospital with the symptom of growth on the right side of buccal mucosa. History revealed presence of growth for 2 months only. The patient did not complain of pain or other secondary symptoms associated with the growth. Medical history and past dental history were not significant. Family history was not contributory. Intraoral examination revealed a well demarcated, firm, painless, slightly raised, oval, black papule measuring approximately 5 mm in diameter over the right buccal mucosa (figure 1). There was no other pigmented lesion on the oral mucosa.

INVESTIGATIONS

Routine blood examinations including haemoblobin%, total leucocyte count (TLC), differential leucocyte count (DLC), clotting time (CT) and bleeding time (BT), which are required before any surgical procedures are performed. They were all within normal limits.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis of oral pigmented nevi includes melanotic macule (focal benign melanosis), smoker's melanosis, physiological and ethnic pigmentation and amalgam tattoos. In case of more purplish or bluish lesions haemangiomas or other vascular anomalies should also be considered in differential diagnosis.

TREATMENT

Following routine haematological investigations excisional biopsy of the growth under local anaesthesia was planned. The growth was excised and

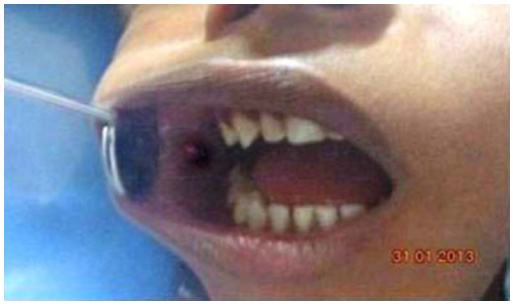

Figure 1 Intramucosal nevus on the right side of buccal mucosa.

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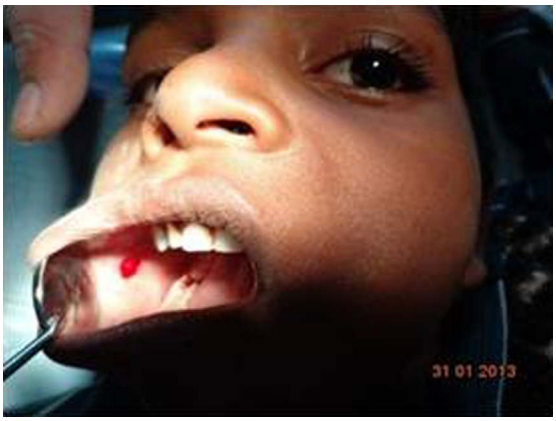

Figure 2 Area after excision.

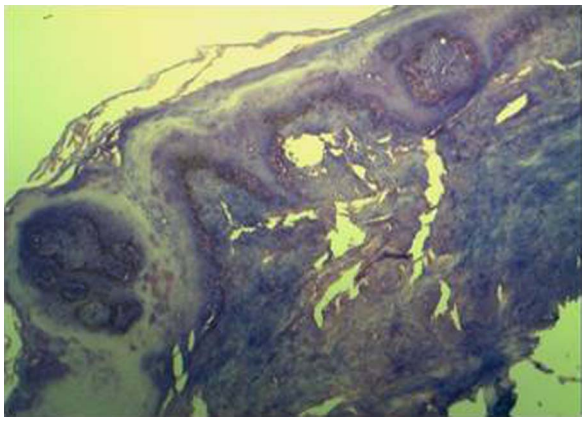

Figure 4 Histopathological picture of intramucosal nevus.

sent for histopathological examination (figures 2 and 3). The H&E sections on microscopic examination showed a layer of parakeratinised stratified squamous epithelium with areas of atrophy in between. The underlying connective tissue stroma contained collections of nevus cells in the form of islands and sheets. The cells were epitheloid with uniform nuclei, less cytoplasm and variable amount of melanin pigmentation. Adjacent to the nevus nests, the connective tissue was collagenous with few capillaries (figure 4). The clinical and histopathological features were suggestive of intramucosal nevus.

DISCUSSION

Diagnosis of pigmented lesions of the oral cavity and perioral tissue is challenging. Definitive diagnosis usually requires histopathological evaluation. Oral melanotic nevi (OMN) are rare benign tumours of melanocytes. Epidemiological data regarding OMN are too scanty to predict its aetiology and pathogenesis.[5] Dark brown, black or bluish discolourations of the oral mucosa can be separated into three categories: vascular lesions, exogenous and endogenous pigmentations. The vascular lesions originate from traumatic purpuric submucosal haemorrhages characterised by the retention of blood, intracellular fluid and haemosiderin pigment in the tissues. These lesions do not blanch with diascopy (firmly pressing a microscope slide or a piece of clear plastic over a lesion). However, they tend to resolve and disappear with time. Haemangiomas can also present with similar clinical features, although they can be differentiated by their blanching characteristics when diascopy is applied.[6] Exogenous pigmentations include metallic tattoos, foreign bodies, occupational exposure and locally acting biological agents. All these aetiological agents were ruled out by case history of the patients and this case was diagnosed as endogenous pigmentation type.

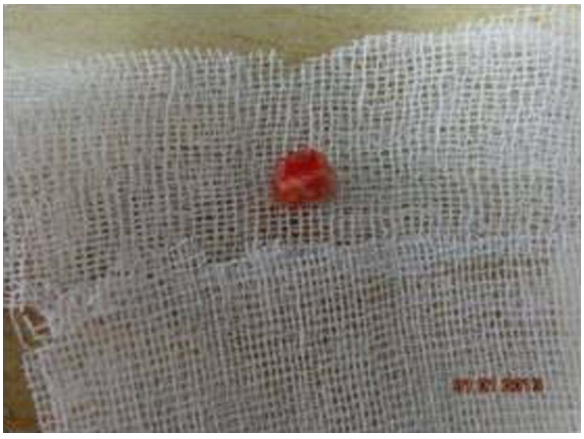

Figure 3 Excised mass.

Nevus cells are thought to originate from melanocytes and have hydropic swollen nucleoli that occupy a large portion of the nucleus. The histology confirms the presence of nevus cells in the connective tissue.[7] Histological variants of OMN correspond to melanocytic proliferations at various levels: (1) proliferation of benign neoplastic melanocytes along the epithelial–mesenchymal junction (junctional nevus); (2) migration of these cells into the mesenchymal compartment (compound nevi) and (3) loss of the junctional component of the nevus, so that all remaining nevomelanocytes are located within the subepithelial compartment (intramucosal nevi).[5] This patient presented with an intramucosal type of nevus at the buccal mucosa in whom histological appearance of the lesion confirmed diagnosis. Buchner *et al* did a clinicopathological study of 32 new cases and reviewed 75 cases from the literature and found that pigmented nevi are much less common in the oral cavity than on the skin. They also supported earlier reports that the most common type of nevus was intramucosal, followed in decreasing order by the common blue nevus, compound nevus and junctional nevus.[8] Buchner and Hansen in still another review and analysis of data on 191 cases of oral pigmented nevi from the literature and from two studies at the University of California found that 55% of the pigmented nevi were of intramucosal type, 32% of common blue nevi, 6% of compound nevus, 5% of junctional nevus and 2% were of combined types. Site wise, 41% of all nevi were found on the hard palate, 20% on the buccal mucosa, 12% on the vermillion border and 11.5% on the gingiva. They were rarely found on the soft palate, tongue and retromolar pad.[9] Our case was of intramucosal type at buccal mucosa, which is an uncommon site of occurrence.

These lesions remain asymptomatic and are twice as common in women as in men.[10] In our case the lesion was present in a male patient, which is a rare finding in male population. Patients in whom this lesion is found are usually between 20 and 40 years of age (mean age, 35 years).[10] In 2004, Buchner *et al*[11] analysed 773 cases of solitary pigmented melanocytic lesions in the oral mucosa and revealed that oral melanocytic nevi comprised 11.8% of the entire melanocytic group with mean age at diagnosis being 30.5 years and palate the most common site. In this case the lesion appeared at early age of 5 years that too in the buccal mucosa which further made the case rarer.

A report from Netherlands during 1980–2005 by Meleti *et al* revealed an annual incidence of excised OMN around 4.35 cases

per 10 million populations per year. According to them there was no concrete support for the idea that the presence of an oral melanocytic nevus indicates a risk of future development of oral malignant melanoma.[12] Though to date there are no reported cases of malignant transformation of intramucosal type of OMN, all OMN should be surgically excised as a prophylactic measure because of constant chronic irritation of the mucosa in nearly all intraoral sites occasioned by eating, tooth brushing, etc.

Learning points

- Pigmented lesions of the oral mucosa range from the common and harmless to the rare and deadly. A systematic and careful diagnostic approach should be used when confronting such lesions in the oral cavity.
- Early diagnosis of the pigmented lesions may improve the extremely poor prognosis associated with melanoma of the oral cavity.
- Instruct the patients to watch for pigmented lesions in the oral cavity, if detected they should immediately report to their physicians/oral surgeons.
- Surgical removal of suspected pigmented nevus should be considered because oral mucosa remains under continuous frictional, thermal and biological challenges.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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