Congenital lip pits without associated anomalies

Sir,

Pits of the lower lip (fistulas of lower lip, paramedian sinuses of lower lip, humps of lower lip, labial cysts) are a very rare congenital malformation, first described by Demarquay in 1845.[1] Lip pits usually appear as two humps on the vermillion border of the lower lip equidistant from the midline.[1-3] They can also occur on the upper lip in the midline or in the oral commissures. A single pit may be located on the lower lip, centrally or lateral to the midline. The occurrence of lip pits is associated with developmental defects in the paramedian portion of the vermilion of the lip.[4] Associated developmental defects are Van der Woude's syndrome, popliteal pterygium syndrome and oral-facial-digital syndrome.[2] A review of the literature in 2005 found only 10 previous reports of isolated lower lip pits.[3] Herein, we report a case of isolated lip pits without a family history of similar lesions.

An 8-year-old boy presented to us with an asymptomatic deformity in his lower lip which was present from birth. On examination, there were two shallow paramedian depressions on the vermilion border of the lower lip with no evidence of a sinus tract or fistula and also no associated cleft lip or palate [Figure 1]. The lower lip was soft and there was no discharge on pressure. There were no other anomalies of lip, face or palate. The birth and medical history were unremarkable. There was no

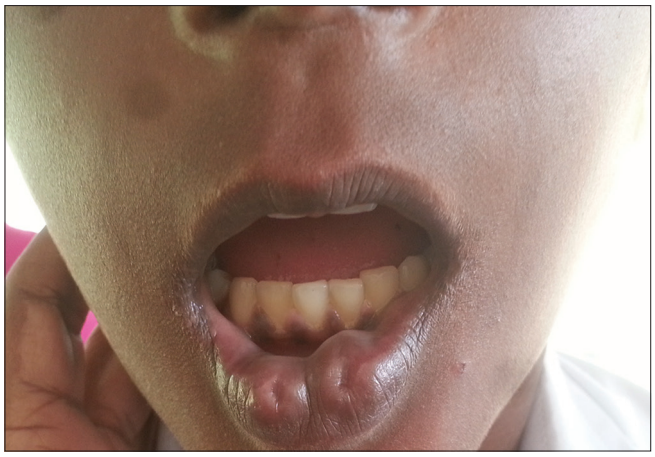

Figure 1: Two shallow paramedian depressions on the vermilion border. There was no evidence of a sinus tract or fistula and no associated cleft lip or cleft palate

history of any facial, lip or palate anomalies in any of his relatives. A diagnosis of isolated congenital lower lip pits without associated anomalies was made. The patient and his parents were reassured and surgical removal was suggested if cosmetic improvement was desired.

Congenital lower lip pits are often inherited as an autosomal dominant trait with variable penetrance and are more common in females. Interstitial deletion of the chromosomal segment Iq32 - 41 has been reported to be associated with the condition.[5] In normal development, fusion of the mandibular arch and sulcus lateralis of the lower lip occurs at 5.5 weeks and fusion of the maxillary and nasofrontal processes occurs at 6 weeks. It is theorized that a common event may simultaneously disrupt fusion in both locations accounting for the strong association between lip pits and cleft lip and/or palate.[3] Small invaginations appear on the embryonal mandibular process at the stage of 7.5-12.5 mm. The origin of lip pits is related to these lateral sulci which usually get obliterated completely. If obliteration does not happen at the cephalic end and the sulci become deeper as growth proceeds, they become congenital lip pits.[1] Associated anomalies include syndactyly of the hands, club foot, ankyloglossia, symblepharon, oral-facial-digital syndrome, genitourinary abnormalities, popliteal pterygium syndrome, cardiovascular anomalies[1] and fusion of deciduous teeth.[6] Lip pits form canals which extend into the orbicularis oris muscle. Fistulography has shown that these sinus tracts can be long and bifurcated with a total length of 5-6 cm. Accessory salivary glands frequently drain into these tracts and due to the action of the orbicularis oris, the saliva is propelled to the surface leading to mucous secretion from the pits.[3] However in our patient, no connecting fistula was found. Histologically, the pits are lined by mucosal epithelium with normal appearing salivary glands lining the sinus tracts. Mild mucosal hyperplasia, inflammation and fibrosis have also been noted. Skeletal muscle (orbicularis oris) surrounds the tract.[3]

The incidence of lip pits may be higher than reported and they can often be mistaken for depressions caused by the patient's teeth (bilateral maxillary incisors). Evaluation for other associated anomalies is important but is usually not undertaken because the pits are often not recognized by the patient's primary care physician. The pits require no treatment if they are mild, but they may be excised for cosmetic reasons.[5]

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