Breastfeeding Keratosis: This Frictional Keratosis of Newborns May Mimic Thrush

AUTHORS: Sudarat Kiat-amnuay, DDS, MS,^a and Jerry Bouquot, DDS, MSD, FICD, FRSM^b

^aDepartment of General Practice and Dental Public Health, and ^bDepartment of Diagnostic and Biomedical Sciences, University of Texas School of Dentistry at Houston, Houston, Texas

KEY WORDS

frictional keratosis, thrush, candidiasis, breastfeeding, oral trauma in infancy, breastfeeding keratosis

Dr Kiat-amnuay saw the patient, conceptualized and designed this report, took all clinical photos, and drafted the case workup for the initial manuscript; Dr Bouquot drafted the discussion and revised the initial manuscript to the final version, including the formation of the table and the taking of the photomicrographs; and both authors approved the final manuscript as submitted.

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Address correspondence to Jerry E. Bouquot, DDS, MSD, FICD, FRSM, Department of Diagnostic and Biomedical Sciences, Rm 5460, University of Texas School of Dentistry at Houston, 7500 Cambridge St, Houston, TX 77054. E-mail: jerry.bouquot@uth.tmc. edu

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abstract

We report the first example, to our knowledge, of a frictional keratosis from exuberant sucking in a breastfeeding infant. A 2-month-old girl was referred for evaluation of a well-demarcated, nonsloughing white keratotic plague of the lower lip mucosa, just inside the vermilion border. The plaque had a slightly irregular surface, had no surrounding erythema, and was the only such plaque in the mouth. It had been present for at least 3 weeks and had been unsuccessfully treated by her pediatrician via oral Mycostatin (nystatin). Her parents sought a second opinion when the infant was prescribed a full course of oral Diflucan (fluconazole). A cytopathology smear (Papanicolaou test) revealed abundant mature keratinocytes with no evidence of Candida. The mother admitted that the infant "worked hard" at sucking during breastfeeding and continued sucking long after feeding. The parents were unaware of any other habit or potential irritation of the lips. After 3 months of age the infant's sucking pattern became more "normal" and the keratosis disappeared; it did not recur during 3 years of follow-up. We propose the term "breastfeeding keratosis" for this entity. Pediatrics 2013;132:e775-e778

Pseudomembranous candidiasis of infancy, ie thrush, is a relatively common oral infection characterized by the presence of partially adherent and nonadherent white plaques of the mucosa. These plaques develop 1 to 3 weeks after birth and typically resemble cottage cheese or curdled milk.^{1,2} Scraping the plaques with a tongue blade or rubbing them with a dry gauze sponge removes them, leaving a normal or erythematous mucosa. They are composed of tangled masses of Candida hyphae (yeasts), desquamated epithelial cells, normal oral bacterial, and miscellaneous debris found normally in the mouth. If confirmatory histopathology

of these plaques is thought to be necessary, a simple cytology smear with Papanicolaou/periodic acid–Schiff staining will typically identify atypical keratinocytes and *Candida* hyphae or spores; a biopsy is most certainly not required.

Although culture or cytopathologic tissue staining confirms the diagnosis, these are almost never done because the distinctive clinical appearance and the lack of other look-alike lesions make the clinical diagnosis definitive. In fact, the sloughing white covers of broken blisters in epidermolysis bullosa make this the only logical additional lesion to be placed in the differential diagnosis. Nonsloughing white keratotic plaques of the oral mucosa in infants have never been reported except in certain syndromes (Table 1). In adults, such plaques are frequently seen as the precancer leukoplakia or as frictional keratosis, which occurs under a variety of diagnostic names (Table 1). The present example of a neonatal frictional keratosis is, to the best of our knowledge, the first such case reported.

PATIENT PRESENTATION

Our patient was a 2-month-old girl with no obvious or previously diagnosed

TABLE 1 Keratotic, Nonsloughing Oral Mucosal Plaques and Macules in Infancy and Childhood

Diagnosis	Plaque or Macule?	Comment
Frictional keratosis	Р	Smooth-surfaced, thick plaque of any mucosal surface from rubbing of mucosa by teeth or dental devices. Essentially equivalent to skin callus.
Linea alba	Р	Thin, sometimes crenated, white linear plaque of buccal mucosa from rubbing teeth against the cheeks; usually bilateral.
Chronic cheek bite	Р	Ragged white keratotic plaque of buccal mucosa along occlusal plane of the teeth; usually bilateral; from habit of chewing on cheeks (often during sleep). Also called morsicatio buccarum.
Chronic lip bite	Р	Ragged white keratotic linear plaque of lower lip mucosa from habit of chewing on lower lip (often during sleep).
Tongue thrust habit	Р	Ragged white keratotic linear plaque along lateral borders of tongue, often with indentations of tongue correlating to tooth contours; from habit of pushing tongue against teeth.
Breastfeeding keratosis	Р	White, thick plaque of lip mucosa in breastfeeding infants; from habit of excess sucking; habit disappears naturally over time.
Migratory stomatitis	Р	Serpiginous or circular thin white lines, often adjacent to red mucosa. Pattern of line changes on a daily or weekly schedule. Also called geographic mouth (geographic tongue if only the tongue is involved).
Lichen planus	Р	White, intersecting "spider web" lines, perhaps on a red background. Usually on buccal and labial mucosa. Small potential for malignant transformation.
Lichenoid reaction	Р	White, intersecting "spider web" lines on a red background. Usually on buccal mucosa. Hypersensitivity response to cinnamon- or peppermint-containing gum, candy, or to dental material.
Pachyonychia congenita	Р	Leukoplakia-like white plaque of the dorsum of the tongue. Rare autosomal dominant disorder with abnormal nails and palmar/plantar keratosis.
Dyskeratosis congenita	Р	Mucosal bullae and erosions, usually on the tongue and buccal mucosa, may develop into leukoplakia-like plaques. Rare, usually autosomal dominant disorder with hyperpigmentation of skin and dysplastic nails. Oral white lesions have a small risk of malignant transformation.
Leukoedema	М	Whitish-gray, diffuse macules of buccal mucosa, usually bilateral. Becomes more pronounced during childhood; more frequent in blacks.
White sponge nevus	М	Whitish-gray, diffuse, often corrugated macules of buccal mucosa, usually bilateral. Becomes more pronounced during childhood years. Familial disorder.
Witkop-Sallmann syndrome	М	Whitish-gray, diffuse macules of buccal mucosa, usually bilateral; ocular plaques may produce eventual blindness. Rare autosomal dominant disorder. Also called hereditary benign intraepithelial dyskeratosis. Caution: dyskeratotic epithelial cells may be misinterpreted as carcinoma.
Smokeless tobacco keratosis	М	Whitish-gray, diffuse macule of buccal and lower vestibular mucosa in area of chronic smokeless tobacco or snuff placement. Essentially a combined chemical burn and abrasion from the tobacco. Users usually start the habit at 8–13 years of age. Disappears completely with habit cessation. Small increased malignant transformation risk.
Listerine keratosis	М	Grayish-white macules of buccal mucosa, often bilateral, in persons who swish high-alcohol-content mouthwashes frequently throughout the day. Essentially a mild chemical burn.
Uremic stomatitis	М	Diffuse whitish-gray macules of the buccal and ventral lingual surfaces in terminal kidney failure; uremic oral odor often is present. Is transient in dialysis patients, disappearing completely immediately after a dialysis session, and then recurring before the next session.

None of the lesions are symptomatic except for occasional lichen planus and lichenoid reaction cases; geographic tongue may be sensitive to spicy or hot foods or may develop a burning sensation from secondary candidiasis.¹⁻⁴ M, macule; P, plaque.

systemic problems. Three weeks earlier she developed an 8×5 mm, welldemarcated, nonsloughing white keratotic plaque of the lower lip mucosa, just inside the vermilion border (Fig 1). The asymptomatic plaque had variable thickness, with 1 region of surface irregularity or granularity and other regions with smooth surfaces. There was no surrounding inflammatory erythema, and the plaque could not be removed via scraping. No other lesions of any type were seen in the mouth.

The girl was brought to her pediatrician for evaluation of the same plaque, at which time it was clinically diagnosed as thrush and treated with liquid Mycostatin (nystatin; Sandoz Limited; Cumberley, Surrey, United Kingdom). The treatment was unsuccessful, and so a course of oral Diflucan (fluconazole; Pfizer, Inc; New York, New York) was prescribed. Her parents, however, were concerned that the diagnosis was not appropriate and therefore sought a second opinion. They thought that the plaque might be a response to excessive sucking during breastfeeding, with considerable sucking between episodes of breastfeeding. Throughout this period the patient's mother experienced no significant breast chafing or irritation from the breastfeeding.

At the initial examination by an oral and maxillofacial pathologist, the lesion was essentially unchanged (see Fig 1).



White, moderately well demarcated keratotic plaque of the lower left lip mucosa had a roughened surface and could not be scraped off.

A cytology or Papanicolau test showed numerous mature keratinocytes with small bacterial colonies and no sign of *Candida* or any other mycotic structures (Fig 2). On the assumption that this was not a *Candida* problem, no additional antifungal therapies were used; the clinician elected to simply follow the patient on the presumption that the sucking habit would eventually diminish or disappear.

This attenuation is exactly what happened. At the 2-week follow-up visit, the keratotic lesion was smaller, and by the fourth week of follow-up it was completely gone (Fig 3). The lesion has not recurred after 3 years of follow-up.

DISCUSSION

A thorough literature review could find no similar case. Focal regions of thickened labial mucosa are not uncommon in breastfeeding infants, but







Combined Papanicolaou test and periodontic acid–Schiff staining of cells scraped from keratinized lesional surface revealed clusters of numerous mature and slightly precornified keratinocytes. Small colonies of coccal and filamentous bacteria were also numerous, but no mycotic structures were present. A, original magnification \times 100; B, original magnification \times 400.



FIGURE 3 One month later, after diminution of the sucking habit, there was no evidence of the plaque.

these appear more like pale pink "pads" than white keratotic, leukoplakia-like plaques. Presumably, our patient's lesion was yet another result of the sucking habit developed for breastfeeding, with an unusually active lip sucking habit while breast feeding and especially between feeding sessions. This habit became less pronounced over time, and with cessation of the habit the lip lesion disappeared. We have called this type of frictional keratosis "breastfeeding keratosis," but we assume that the lesion cannot develop without exuberant lip sucking activity between feedings.

The lesion is, of course, innocuous, and we believe it will always disappear as the sucking habit becomes less pronounced, just as other forms of adult-onset frictional keratosis disappear when the offending tooth or denture edge is made less traumatic. The major problem, probably the only problem, is mistaking the keratosis for thrush and treating accordingly without success, leading to the potential use of hepatotoxic systemic antifungal agents that have not been approved for neonatal infants. The recognition of the proper diagnosis will prevent this situation from occurring.

CONCLUSIONS

Active sucking during and between breastfeeding can produce a transient frictional keratosis of the lip mucosa. This lesion requires no treatment and should be differentiated

from the more common sloughing white plaque of infancy, thrush. We

propose the diagnostic term "breast-feeding keratosis" for this entity.

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