



CASE REPORT

Sublingual Mass in a Neonate

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INTRODUCTION

The intraoral examination is a crucial part of the neonatal exam, as it can identify various abnormalities, including an abnormal palate, bifid uvula, and natal teeth. The differential for an intraoral mass is extensive and may include congenital cyst, infection, vascular malformation, epithelial inclusion cyst, and malignancy. Most of these masses are benign and not a cause for significant concern. This article introduces two cases, each involving the incidental finding and assessment of a distinct sublingual mass.

CASE REPORT

The first case involves a female infant born at 38 weeks and four days via cesarean section, who was admitted to the hospital on the sixth day of life for evaluation of poor feeding. Parents reported that the infant seemed disinterested in feeding via breast or bottle. History was significant for hydronephrosis and ankyloglossia requiring frenotomy in the newborn nursery. Upon initial exam, a 2cm yellow-orange nodule was noted under the tongue (figures 1.1 and 1.2). It was diamond-shaped and seemingly non-tender. No fluctuance, discharge, or surrounding erythema or swelling was noted. The family had not noticed the lesion. Occupational therapy and the newborn physician team were consulted and did not believe this interfered with feeds. Labs were significant for bilirubin of 10.6 mg/dL (normal bilirubin level 0-1.0 mg/dL). The sublingual mass decreased in size throughout the infant's hospitalization, and she was ultimately discharged without further intervention after parental teaching concerning feeds and demonstration of appropriate weight gain with feeding. The infant was examined one week later, and there was no mention of intraoral abnormality. The total bilirubin level at this visit was 2.0 mg/dL.

The second case addresses a six-day-old female born at 39 weeks of gestation via spontaneous vaginal delivery with a medical history significant for ABO incompatibility with indirect hyperbilirubinemia not requiring phototherapy and ankyloglossia with frenotomy performed on one day of life. The infant presented to the clinic on the day of life six for a bilirubin and weight check. Examination revealed a jaundiced infant with a 1cm midline sublingual yellow mass (figure 2.1). The infant seemed unaffected by the mass and was breastfeeding well. The transcutaneous bilirubin level in the clinic was 14.5 mg/dL. The infant returned at two weeks of age for a routine well-child check. At this point, the intraoral examination demonstrated resolution of the sublingual discoloration and mass.



Figure 1.1



Figure 1.2



Figure 2.1

DISCUSSION

The sublingual masses described in these cases were likely caused by the accumulation of bilirubin-rich granulation tissue at the infants' frenotomy sites. In the case of frenotomy, the lingual frenulum is surgically released and allowed to heal by secondary intention. Families are instructed on lingual stretches to prevent excessive wound contracture. The wound-healing process is complex and can be complicated by infection and delayed healing. Although the deposition of bilirubin within granulation tissue has been previously noted in the literature, there have been no prior case reports identifying bilirubin deposition at frenotomy sites.

Several studies have demonstrated that bilirubin can play a role in wound contracture, healing, and collagen deposition.¹⁻³ Bilirubin's antioxidant and anti-inflammatory properties can reduce oxidative stress and inflammation at the site of injury, promoting healing.² One study demonstrated that rats treated with bilirubin had accelerated wound closure, thus highlighting bilirubin's role in angiogenesis and collagen deposition at the wound site.⁴ Another study involving rats showed that granulation tissue from wound sites treated with bilirubin expressed upregulated anti-inflammatory and downregulated pro-inflammatory markers, thus suggesting that bilirubin can play a significant role in wound healing.⁵

These studies support the conclusion of bilirubin deposition in the sublingual granulation tissue following frenotomy and may further suggest that this bilirubin enrichment aided healing. Understanding the role of bilirubin in wound healing has essential implications for physicians caring for newborns who have undergone frenotomy or other surgical procedures. One potential benefit is preventing unnecessary consultations, referrals, or procedures. Yellow-orange granulation tissue at the frenotomy site can be concerning to both parents and medical professionals, potentially leading to unnecessary interventions if the granulation tissue is misinterpreted as an infection or congenital abnormality. It can also induce undue anxiety and stress for families if further interventions are pursued. Understanding the process and benefits of bilirubin deposition can equip physicians to reassure families that the wound is progressing as expected.

In conclusion, recognizing the role of bilirubin in wound healing is crucial for physicians caring for newborns who have undergone frenotomy or other surgical procedures. By understanding the potential benefits of bilirubin in wound healing, physicians can better reassure parents, prevent unnecessary consultations or procedures, and optimize wound care to promote healing.

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