Sialometaplasia of the Soft Palate in a 2-Year-Old Girl

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SOMMAIRE

Nous présentons un cas de sialométaplasie du palais mou qui s'est manifesté chez une fillette de 2 ans, 3 mois après une adénoïdectomie. Il est important que les dentistes connaissent les causes possibles des enflures intra-buccales chez les enfants et les adultes et sachent que certaines affections peuvent se manifester différemment chez les enfants et les adultes. Chez un adulte, la sialométaplasie nécrosante peut se présenter sous forme de lésion agressive, évocatrice d'un carcinome spino-cellulaire. Chez notre jeune patiente, toutefois, l'examen clinique ou histologique n'a révélé aucun signe manifeste de nécrose. Il est donc possible que la nécrose ne fasse pas partie des manifestations de la sialométaplasie chez des enfants aussi jeunes.

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Necrotizing sialometaplasia is a benign condition that may be found at any site in the body that contains elements of the salivary gland, from the paranasal sinuses to the lung.[1–6] Numerous cases[7–17] have been reported in the oral cavity since the condition was first described in 1973. The lesion also occurs in other mammals such as dogs and cats.[18,19] Recognizing necrotizing sialometaplasia is important because this lesion mimics the appearance of malignant disease, both clinically and microscopically.[1,7,20,21] Failure to recognize necrotizing sialometaplasia may result in unnecessary radical surgery because of an erroneous preoperative diagnosis of squamous cell carcinoma or mucoepidermoid carcinoma.[1,7] In this paper, a case of sialometaplasia of the soft palate in a 2-year-old girl 3 months following adenoidectomy is reported.

Case Report

An otherwise healthy 2-year-old girl was referred by her dentist for investigation of an intraoral paramedian submucosal swelling of the left side of the soft palate posterior to the hamulus. The patient had a history of troublesome recurrent infections of the middle ear and had undergone adenoidectomy about 3 months before the referral. Results of the examination showed a slightly pale smooth-surfaced non-ulcerated firm nodule in the left side of the soft palate just posterior to the hamulus. The nodule measured 1.5 × 1.0 cm (**Fig. 1**). Within 1 week of her referral, the patient was taken to the operating room to have an excisional biopsy. An elliptical piece of tissue containing the entire submucosal mass with a cuff of normal mucosa was removed. The wound was closed with interrupted resorbable sutures.

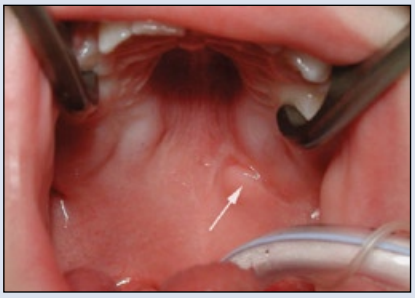

Figure 1: Paramedian firm submucosal swelling (white arrow) of the left side of the soft palate.

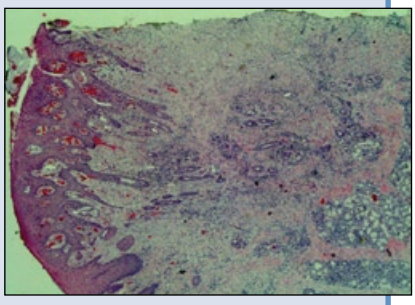

Figure 2: Low-power view of an hematoxylin and eosin-stained (H&E) section showing pseudoepitheliomatous hyperplasia overlying minor salivary glands of the left side of the soft palate.

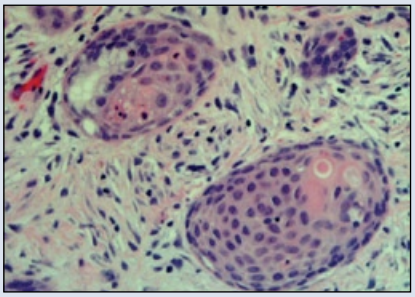

Figure 3: High-power view of an H&E-stained section with intact elements of lobular salivary glands with squamous metaplasia of ducts.

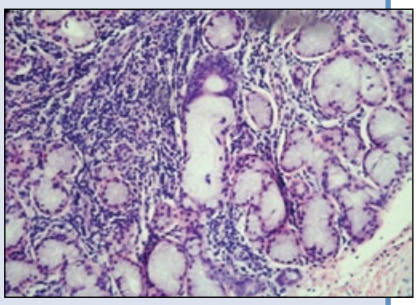

Figure 4: Lobular necrosis of minor palatal salivary glands adjacent to intact salivary gland lobules.

Results of the histological examination of the specimen indicated that it contained pseudoepitheliomatous hyperplasia of the overlying epithelium. Squamous metaplasia of the salivary duct epithelium and lobular necrosis with preservation of the lobular architecture of the neighbouring minor salivary glands was also found (**Figs. 2–4**). Based on the histological findings and the timing of the appearance of the lesion after a recent surgical procedure in the same anatomical area, a diagnosis of sialometaplasia was made. The patient's biopsy site healed without complication over 2 weeks. Results of several postoperative examinations during 18 months of follow-up showed no reoccurrence of the lesion.

Discussion

Etiology and Pathogenesis

The initiating event of necrotizing sialometaplasia is believed to be related to ischemia that is secondary to alteration in the local blood supply.[22,23] In the case reported here, the girl's adenoidectomy may have affected branches of the ascending pharyngeal, tonsilar and lesser palatine arteries in the soft palate, compromised the underlying vascular supply and likely caused infarction of the salivary gland tissue. Salivary gland tissue may make greater metabolic demands and may be more prone to necrosis after vascular alterations than the overlying mucosa. Acinar cells may then become necrotic, although ductal preservation is usually noted within the infarcted lobules, and squamous metaplasia of ductal remnants eventually appears.[7]

This condition may occur after local trauma, including surgical manipulation or injection of local anesthesia into the involved area.[22] For patients who have had surgery, the lesion becomes clinically evident in about 3 weeks or longer. Necrotizing sialometaplasia may also appear spontaneously, often without the history of a prior surgical or traumatic event.[1–7] No particular oral condition or habit has been associated with this condition to date.

Clinical Features

Intraorally, necrotizing sialometaplasia is characterized by a seemingly spontaneous presentation, most commonly at the junction of the hard and soft palates.[24] Early in this evolution, the patient may note that the lesion is a tender swelling. Subsequently, the mucosa breaks down as a sharply demarcated deep ulcer with a yellowish-grey lobular base form. In the palate, the lesion may be unilateral or bilateral; the diameter of individual lesions ranges from 1 to 3 cm.[22,23] Symptoms are generally disproportionately slight, compared with the size of the lesion. Most patients indicate surprisingly mild complaints of tenderness or dull pain. Healing is generally slow and protracted, ranging from 6 to 10 weeks.[25]

Histopathology

The microscopic features of necrotizing sialometaplasia are consistent and unique.[1,7] The overlying mucosa may become ulcerated in the early phases. Lobular necrosis of the salivary glands, pseudoepitheliomatous hyperplasia of adjacent epithelium, and prominent squamous metaplasia of the epithelium of the salivary ducts are typically seen.[20,21] The recognition of lobular necrosis and the preservation of lobular architecture distinguish this process from neoplasia.[1,7] The characteristic squamous metaplasia of ductal elements may be misinterpreted as squamous cell carcinoma. When this metaplasia is seen in the presence of residual viable salivary

glands, the lesion may be mistaken for mucoepidermoid carcinoma.[1–7]

Differential Diagnosis

Squamous cell carcinoma or malignant minor neoplasms of the salivary glands must be ruled out clinically and histologically.[1,7] Granulomatous diseases such as syphilitic gumma and deep fungal infections must also be ruled out because they may appear as punched-out lesions of the palate with a sharp demarcation.[9] In medically compromised patients, such as those with poorly controlled diabetes, opportunistic fungal infections with mucormycosis may also cause a similar clinical picture.[9]

Treatment and Prognosis

Necrotizing sialometaplasia is a benign self-limiting process that does not require specific treatment.[20] If the lesions are large, incisional biopsy should be done to establish a definitive diagnosis.[7] Healing takes place over several weeks by secondary intention. Reassuring the patient and lavaging the wound with irrigation or rinsing may be helpful.[25] Once the lesion has healed, recurrence and functional impairment are not anticipated. In this reported case, an excisional biopsy was done and the sutured wound healed without complication.

Conclusion

Although necrotizing sialometaplasia is classically characterized by areas of frank necrosis and ulceration, the child reported here had no signs of necrosis. Clinically, however, the child had intraoral swelling, and histological results showed evidence of minor salivary gland ductal metaplasia.[25] The child's preceding adenoidectomy may account for the surgical trauma, similar to the trauma that others[22,23] have reported in cases of necrotizing sialometaplasia. The child's uneventful postoperative healing and the lack of recurrence of the lesion during 18 months of follow-up also support a diagnosis of sialometaplasia. It is possible that frank necrosis and ulceration may not be a feature of this lesion in children this young because the lesion may resolve spontaneously before such necrosis has a chance to develop.

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