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Traumatic Acquired Oronasal Palatal Fistula in an Adult Patient with an Undiagnosed Submucous Cleft Palate

I.J.L. See[1], S. Martin[2], T.G. Thorburn[2], M. C. Swan[2]

1. University of Auckland, Faculty of Medical and Health Sciences, Auckland, New Zealand.
2. Spires Cleft Centre, Oxford University Hospitals NHS Foundation Trust, Oxford, United Kingdom.

Abstract

Presentation

The patient presented with symptomatic nasal regurgitation after developing a traumatic oronasal fistula whilst eating a Pringle® on Christmas Day.

Diagnosis

The underlying submucous cleft was diagnosed by intra-oral examination and confirmed intra-operatively.

Treatment

Surgical intervention to repair the fistula and submucous cleft palate.

Conclusion

This case illustrates the development of an acquired oro-nasal fistula following the consumption of an innocuous festive snack in an adult and highlights the need to have a high degree of suspicion of an underlying submucous cleft palate.

Keywords: Cleft palate, submucous cleft palate (SMCP), palatal fistula, velopharyngeal insufficiency, palatal perforation.

Introduction

A submucous cleft palate (SMCP) is the mildest phenotype of a congenital cleft palate and tends to be diagnosed later in childhood as it becomes symptomatic either in terms of nasal regurgitation or velopharyngeal dysfunction. In contradistinction to an overt cleft palate, which has an obvious defect that is typically diagnosed postnatally, a SMCP has intact mucosa that obscures the underlying abnormal muscular anatomy. Calnan described a triad of features that herald a SMCP: a bifid uvula, a zona pellucida (a thin, almost translucent, area of central mucosa due to the absence of muscle within the midline), and thirdly, a notch in the hard palate.[1] However, all three features are not always present, and an isolated bifid uvula is relatively common with a prevalence of 2%.[2,3]

There are scant reports in the literature of SMCPs presenting following traumatic penetration of the zona pellucida. We report an interesting case of a SMCP diagnosed in an adult after consuming a festive morsel on Christmas Day.

Case Report

A 54-year old lady was referred with a spontaneous palatal fistula that appeared after eating a Pringle®. The patient experienced symptomatic nasal regurgitation with fluids and solids throughout the festive period as well as a change in speech quality due to the persistent air leak through the fistula

Intraoral examination revealed a 20mm x 20mm soft palate fistula and the classic features of Calnan's triad (Figures 1 and 2). Formal speech assessment confirmed hypernasality and nasal emission that was consistent with a large oronasal fistula.

Fig 1. Oronasal fistula within the zona pellucida; note the bifid uvula.

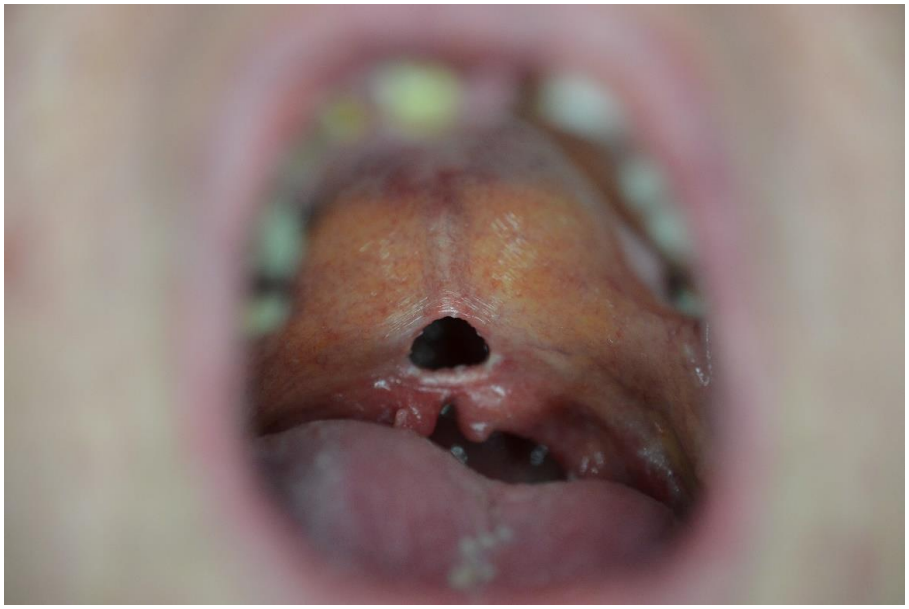
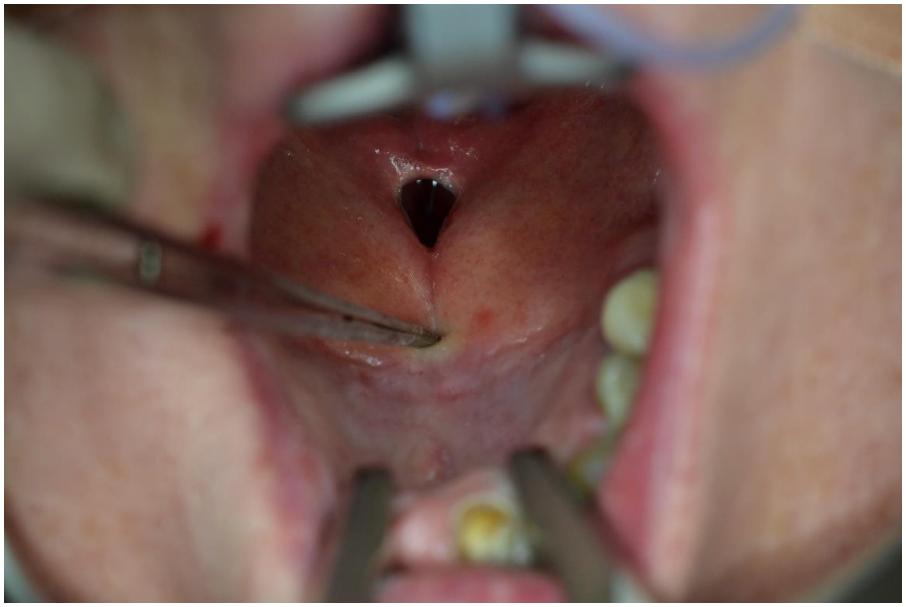


Fig 2. Examination under anesthesia revealed a hard palate notch.



Given the rarity of acquired palatal fistulae, a biopsy was performed to exclude malignancy. Histology demonstrated normal stratified squamous epithelium with mild hyperkeratosis indicating chronic inflammation. Candida was also detected and treated with fluconazole.

Subsequently, the patient underwent formal repair of the fistula with posterior re-positioning of the levator muscles to correct the SMCP.

Postoperatively, the patient obtained an excellent functional result with good palate elevation and resolution of her hypernasality and nasal regurgitation without the need for speech therapy.

Discussion

Late presenting palatal fistulas remain rare in adults and should arise suspicion of an underlying SMCP. Controversy exists over the terminology of acquired palatal fistulae, impacted by the sparse number of cases. Some authors term them congenital fistulae as the majority of cases described occur in the neonatal or infant period, with some present at birth.[4,5] Mehendele and Sommerlad reported four patients with a palate perforation spanning three decades, with only one presenting in adulthood secondary to trauma from denture use. Two cases were diagnosed within months of birth and the third was detected on the day of surgery for repair of a known SMCP.[6]

Apart from direct trauma to the palate, acquired fistulae have been documented to occur secondary to an aphthous ulcer, with or without superimposed infection.[7,8] Park and colleagues describe an infant with a bifid uvula and feeding difficulties who was treated for Candida infection of a palatal ulcer; on follow up, the ulcer progressed to a palatal fistula.[8] Interestingly, Candida was also isolated from our patient but she had no evidence of mucosal ulceration prior to fistula development.

There has been a rise in oronasal fistulae secondary to cocaine use[9] but this is the first report in the literature of an acquired fistula caused by a popular potato-based snack with a significant impact on eating and speech. Both of these functions are particularly pertinent over the festive season whilst socialising with friends and family and chanting yuletide hymns. Hwang and Kim reported another food induced palatal fistula secondary to an intra-oral burn sustained while eating hot food on a background of an undiagnosed SMCP.[10]

It can be concluded that the seemingly mundane activity of eating potato snacks may be a risky occupation for those with underlying palatal deficiency.

Declaration of Conflicts of Interest:
There are no conflicts of interest to declare.

Corresponding Author:
Marc Christopher Swan
Spires Cleft Centre,
Oxford University Hospitals NHS Foundation Trust,
Oxford,
United Kingdom.
E-Mail: marc.swan@nds.ox.ac.uk

References:

1. Calnan J. Submucous cleft palate. Br J Plast Surg 1954;6:264.
2. Wharton. P and Mowrer. D.E, Prevalence of cleft uvula among school children in kindergarten through grade five, Cleft Palate Craniofac J 1992 Jan;29(1):10-2; discussion 13-4
3. Rivron. R.P, Bifid uvula: prevalence and association in otitis media with effusion in children admitted for routine otolaryngological operations, J Laryngol Otol 1989 Mar;103(3):249-52.
4. Fara M. Congenital defects in the hard palate. Observation of five cases. Plast Reconstr Surg. 1971;48:44–47.
5. Cheng N, Zhou M. Congenital fistula of the palate. J. Maxillofac. Surg. 1998 Dec 1;26(6):391-3.
6. Mehendale FV, Sommerlad BC. Submucous cleft palates presenting with a perforation. Cleft Palate Craniofac J 2003 Mar;40(2):203-6.
7. Weijerman. J.E and Heybroek. G, Perforation in a case of submucous cleft palate due to an aphthous ulcer. Br J Oral Maxillofacial Surgery 1972;10:217–219.
8. Park. I.H, Chung. J.H, Choi. T.H, Han. J and Kim. S.W, Acquired palatal fistula in patients with submucous and incomplete cleft palate before surgery, Arch. Plast. Surg., 2016 Nov; 43(6): 582-585
9. Silvestre. S.J, Perez-Herbera. A, Puente-Sandoval. A and Bagan. J.V, Hard palate perforation in cocaine abusers: a systematic review, Clin Oral Invest (2010) 14:621–628
10. Hwang. K and Kim. Y.S, Perforation in Submucous Cleft Palate Due to Thermal Injury, J Craniofac Surg. 2010 Jan;21(1):280-1