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CLINICAL RECORD

'A good shepherd, but with obstructive sleep apnoea syndrome': traditional uvulectomy case series and literature review

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Abstract

Background: In the West, removal of the uvula is predominantly undertaken as part of palatal surgery, in cases of obstructive sleep apnoea. In the developing world, such as the Middle East and Africa, uvulectomy is a more common practice. The uvula is removed for curative or preventive purposes, or as part of ritual practice. Due to immigration from developing to developed world countries, and to Western doctors working abroad, such doctors are increasingly being confronted with unfamiliar traditional healing practices, within a medical context.

Methods: The Medline and Embase online databases were systematically searched for literature on traditional uvulectomy. We present a review of this literature. We also present the first report, to our best knowledge, of obstructive sleep apnoea as a late complication of traditional uvulectomy.

Discussion: Traditional uvulectomy may be complicated by post-operative haemorrhage and local infections, among many other problems. We report cases of obstructive sleep apnoea and snoring caused by palatal stenosis resulting from traditional uvulectomy during childhood.

Key words: Sleep Apnoea, Obstructive; Medicine, Traditional; Uvula; Uvula, Surgery; Palate, Abnormalities; Nasopharynx, Abnormalities

Introduction

The uvula is a small piece of soft tissue which dangles down centrally from the soft palate. It has an abundance of salivary glands, and plays a large role in moistening the oropharyngeal and pharyngeal mucosa; it is also an important speech articulator.[1,2]

Uvulectomy is the most common term for removal of the uvula; other synonyms include uvolotomy, staphylectomy and kionectomy. The removal or shortening of the uvula (the latter comprising removal of redundant mucosa, with or without addressing the muscle) plays a role in the palatal surgery used to treat snoring or obstructive sleep apnoea (e.g. uvulopalatopharyngoplasty).[3–5]

However, uvulectomy is a more common practice in other areas of world, specifically the Middle East (Egypt (the Bedouins of the Sinai desert), Israel, Lebanon, Saudi Arabia and Yemen), Western Africa (Mali, Niger, Nigeria and Sierra Leone), Eastern Africa (Eritrea, Ethiopia, Kenya and Tanzania), Central Africa (Cameroon and Chad) and Northern Africa (Morocco and Sudan).[1,2,4,6–25] The uvula is removed for curative or preventive purposes, or as part of ritual practice.[3,12] Traditional uvulectomies are performed without anaesthesia by traditional healers or barbers, usually in children (both boys and girls), but they may also be carried out in adults to treat certain conditions.[8,12,14]

Such uvulectomies are predominantly used to treat problems of the throat, such as chronic coughing (due to a long uvula tickling the throat), sore throat, pharyngitis, tonsillitis or loss of voice. Congestion, vomiting, diarrhoea, dysphagia, fever, headache, insomnia, anorexia, poor oral intake, rejection of the breast and failure to thrive have also been reported as treatment indications.[2,6,7,11,12,14,20,23–26] In some ethnic groups, such as the Hausa group, uvulectomy is ritually performed at birth or on the seventh day after birth, during the naming ceremony.[2,12,26] Other ethnic groups remove the uvula as a preventive measure; indications include respiratory disease, diarrhoea and vomiting.[1–4] Some ethnic groups believe that removal of the uvula is necessary for health or to help children to swallow.[2,13] In addition, some groups believe the uvula can kill, by swelling, bursting and causing suffocation and death in neonates.[2,10,12,13,26,29] In Ethiopia, some groups believe that the uvula is a source of thirst, and that by removing the uvula they will need less water and tolerate thirst better.[1,26,30]

Many methods are used to perform this traditional procedure. The majority of barbers use a sickle-shaped knife to cut and remove the uvula, and use a tongue depressor, wooden spoon, wooden spatula, forceps or stick to sandwich the uvula between the latter and the knife as well as to depress the tongue.[3,6,10–13,26,28] Other methods use horsehair, a hot knife, a flattened nail, the tines of a reed fork, or scissors.[3,10,24,28] Barbers perform the procedure in the morning, as it is believed that blood is more viscous at that time of day and that the risk of haemorrhage is therefore reduced.[4,6]

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The instruments used are often unsterile and conditions are unhygienic, thus increasing the risk of infection.[13,30,31] In addition to post-operative haemorrhage and local infections (cellulitis, pharyngitis, tonsillitis, and peritonsillar and pharyngeal abscesses), many other complications have been reported: systemic infections (septicaemia, aspiration pneumonitis, lung abscess, meningitis, tetanus, hepatitis and acute nephritis), glottic oedema, severe anaemia, upper airway obstruction, passage of cut pieces of the uvula further down the respiratory tract, diarrhoea with dehydration, and death.[2,6,10–13,15,27–29] Chronic complications have also been described, such as nasal speech, palatal abnormalities, velopharyngeal insufficiency and human immunodeficiency virus infection.[8,12,13,20,25]

Eleven per cent of the Dutch population consists of non-Western, primarily from Turkey, Morocco, Suriname, and the Dutch Antilles and Aruba.[21] Due to immigration from developing to developed world countries, and to Western doctors working abroad, such doctors are increasingly being confronted with unfamiliar traditional healing practices, within a medical context.[6,10,26] Therefore, it is important that Western-trained doctors are familiar with the techniques, cultural context and epidemiology of traditional uvulectomy, and also with its possible complications and their management.[6]

In our out-patient clinic, we have been faced with (to our best knowledge) previously unreported, late complications of traditional uvulectomy. Below, we report cases of obstructive sleep apnoea in which palatal stenosis due to childhood traditional uvulectomy played a role.

Case reports

Case one

A 42-year-old man, originally from Eritrea, attended our out-patient clinic due to snoring. His partner complained about his excessive snoring, which was socially unacceptable and affected both their relationship and his own night rest. Furthermore, his partner had repeatedly observed apnoea episodes. The patient himself complained of excessive daytime sleepiness (with an Epworth Sleepiness Scale score of 12), as well as impaired concentration and forgetfulness. He has smoked one packet of cigarettes per day for the past 17 years, and rarely drank alcohol.

The patient's body mass index (kg/m^2) was 25 and his neck circumference was 39 cm. Anterior rhinoscopy revealed a straight septum, small inferior turbinates, and no nasal polyps or other pathology. Examination of the oropharynx revealed small palatine tonsils and an enlarged tongue (corresponding with Friedman palate position stage three).

More importantly, an inverted-V-shaped stenosis of the soft palate was observed, characteristic of previous uvulectomy (Figure 1).

The patient confirmed that his uvula had been removed when he was only 7 days old, in Eritrea. In his tribe, it was thought that removing the uvula in newborn boys helped them to become better shepherds.

The patient underwent polysomnography, which indicated an apnoea–hypopnoea index of 14, an apnoea index of 4.6, a desaturation index of 3, a mean oxygen saturation of 97 per cent and a minimum oxygen saturation of 88 per cent. The patient snored for 59 per cent of his total sleep time (576.5 minutes), especially in the supine position.

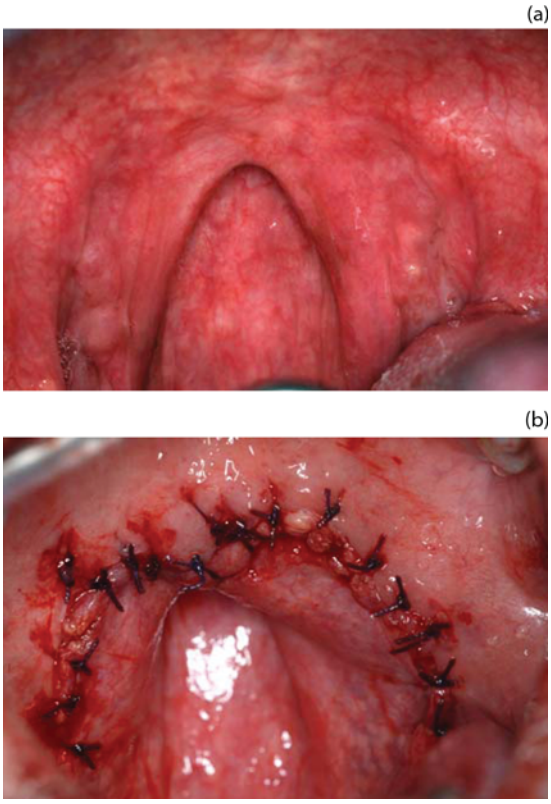

FIG. 1

Clinical photographs of case one showing (a) inverted-V-shaped stenosis of the soft palate resulting from traditional uvulectomy, and (b) the result of a Z-palatoplasty and tonsillectomy performed to treat his obstructive sleep apnoea.

To observe the site of obstruction, drug-induced sleep endoscopy was performed. A single, anteroposterior, velopharyngeal-level obstruction was observed.

In conclusion, this patient suffered from mild obstructive sleep apnoea due to velopharyngeal obstruction, caused by an inverted-V-shaped stenosis of the soft palate resulting from traditional uvulectomy.

The patient refused continuous positive airway pressure treatment. Therefore, we advised him to consider a Z-palatoplasty and tonsillectomy, in order to achieve maximal airway enlargement. The patient consented, and underwent this procedure.

Case two

Severe snoring was the chief complaint of a 51-year-old woman, originally from Morocco, with a history of traditional uvulectomy performed in childhood. In addition, apnoeas had been observed by her partner, and there were complaints of occasional daily hypersomnolence.

The patient's body mass index was 33. Nasal examination revealed a slight deviation of the septum towards the right, and small inferior turbinates. Examination of the oropharynx revealed small palatine tonsils and an absent uvula (Figure 2).

Polysomnography showed severe obstructive sleep apnoea, with an apnoea–hypopnoea index of 36, a desaturation index of 33, a mean oxygen saturation of 93 per cent and a minimum oxygen saturation of 80 per cent.

This patient was not motivated to pursue palatal surgery; therefore, continuous positive airway pressure was prescribed.

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FIG. 2
Clinical photograph of case two showing small palatine tonsils and an absent uvula.

Case three

A 33-year-old man attended our out-patient clinic complaining mainly of snoring. His partner had repeatedly observed apnoea episodes. The patient himself complained of restless sleep and morning fatigue (with an Epworth Sleepiness Scale score of nine). He reported smoking 30 cigarettes and drinking three units of alcohol per day. He had previously undergone external rhinoplasty (in 2008) and turbinate surgery (in 2007).

The patient's body mass index was 29 and his neck circumference was 41 cm. Anterior rhinoscopy revealed no evident abnormality. Examination of the oropharynx revealed small palatine tonsils (grade II), an absent uvula, a stenotic palatum and an enlarged tongue (Friedman palate position stage three) (Figure 3).

The patient underwent polysomnography, which revealed an apnoea–hypopnoea index of 18.6, a desaturation index of 4.2, a mean oxygen saturation of 93 per cent and a minimum oxygen saturation of 89 per cent. The patient snored heavily during the investigation.

During drug-induced sleep endoscopy, an anteroposterior, velopharyngeal-level obstruction was observed, as well as partial anteroposterior obstruction of the tongue base.

In conclusion, this patient suffered from moderate obstructive sleep apnoea caused by multilevel obstruction and soft palate stenosis, resulting from previous traditional uvulectomy.

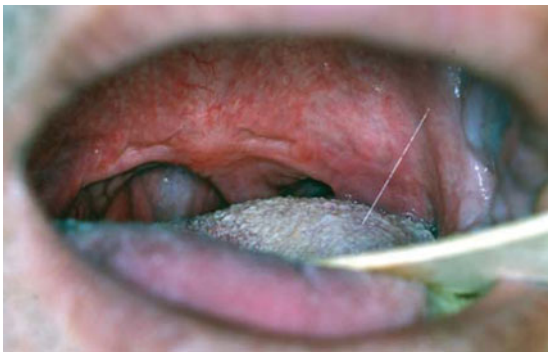

FIG. 3
Clinical photograph of case three showing small palatine tonsils, an absent uvula and a stenotic palate, due to traditional uvulectomy during childhood.

After considering his options, the patient chose continuous positive airway pressure treatment.

Discussion

The Medline database was systematically searched using the following search string: (uvul* OR 'Uvula/surgery'[Mesh]) AND ('Medicine, Traditional'[Mesh] OR ritual* OR tradition*). Of 75 articles identified, 44 were found to be relevant. The Embase database was also systematically searched, and one additional article found.[11] To our best knowledge, neither snoring nor obstructive sleep apnoea has previously been reported as long-term complications of traditional uvulectomy.

Despite no conclusive evidence that our patients' obstructive sleep apnoea was caused by traditional uvulectomy, it most probably played a role. It is known that nasopharyngeal stenosis can cause obstructive sleep apnoea.[33]

Our first patient was motivated to undergo surgery, after refusing continuous positive airway pressure treatment. We decided to treat this patient with a Z-palatoplasty and tonsillectomy, with the following goals in mind: (1) to reduce nasopharyngeal stenosis, (2) to increase retropalatal diameter, (3) to increase the lateral dimension of the oropharyngeal airway, and (4) to improve or eliminate obstructive sleep apnoea and snoring (see Figure 1b).[34]

Z-palatoplasty, originally introduced as a 'modified uvulopalatopharyngoplasty for patients without tonsils', is also used to treat patients who have previously undergone an unsuccessful uvulopalatopharyngoplasty procedure.[34,35]

Nasopharyngeal stenosis is a dreaded complication of uvulopalatopharyngoplasty. Using the grading system described by Krespi and Kacker, it may be divided into three stages: type I (mild), type II (moderate) and type III (severe, also known as a subtotal nasopharyngeal stenosis).[36] Nasopharyngeal stenosis occurs as a complication not only of uvulopalatopharyngoplasty and traditional uvulectomy, but also of tonsillectomy and adenoidectomy, radiotherapy, syphilis, rhinoscleroma, lupus, diphtheria, tuberculosis, trauma, acid burn, and unspecified infection; it may also be congenital.[26–28] Various treatment methods exist: stents, nasopharyngeal obturators, balloon dilatation, topical mitomycin-C, laser excision and palatal surgery.[36,39]

We believe that Z-palatoplasty can be effective in cases of types I and II nasopharyngeal stenosis (see Figures 1a, 2 and 3), while laser excision of the stenosis and subsequent maintenance therapy with obturators should be reserved for type III cases.

Our first patient repeatedly missed his follow-up appointments. Without warning, more than a year after his surgery this patient made an appointment for further polysomnography. This investigation revealed an apnoea–hypopnoea index of 15.1, a desaturation index of 9.5, a mean oxygen saturation of 94.8 per cent and a minimum oxygen saturation of 86 per cent. This represented a disappointing result; however, his snoring had decreased from 59 to 29 per cent of total sleep time. Furthermore, adopting a supine position had a marked influence on his apnoea–hypopnoea index: he spent 33 per cent of total sleep time in the supine position, during which his apnoea–hypopnoea index rose to 33.9, but this parameter fell to 5.6 during non-supine sleep.

We conclude that this patient had residual positional obstructive sleep apnoea. However, the patient had not slept in a supine position pre-operatively. This raises the question of whether this patient's pre-operative

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apnoea–hypopnoea index would have been more severe if he had slept in a supine position. Positional obstructive sleep apnoea is often due primarily to tongue base collapse.[40] Did this patient's pre-operative stenosis inhibit a latent tongue base obstruction? It may have been the case that, following surgery which led to enlargement of the lateral dimension of the palate, this patient's already enlarged tongue had more room to cause an anteroposterior obstruction of the airway.

- **Traditional uvulectomy is commonly practiced in Africa and the Middle East**
- **The uvula is removed for curative or preventive purposes, or within the scope of ritual practice, often in unhygienic conditions**
- **Many acute and chronic complications have been reported**
- **This study reports late complications of childhood traditional uvulectomy, including obstructive sleep apnoea due to palatal stenosis**
- **Western doctors are increasingly confronted with unfamiliar traditional medical practices and their consequences, due to professional mobility and immigration**

No surgical treatment of obstructive sleep apnoea has a 100 per cent success rate. Revision uvulopalatopharyngoplasty by Z-palatoplasty has been demonstrated to be successful in 68 per cent of patients, using Sher's success criteria.[34]

Unfortunately, in many of the countries mentioned above the local population continues to prefer the help of traditional healers, who are generally more 'socio-culturally accessible and acceptable'.[16] Western surgery may not be an option for such people, due to lack of availability and/or high costs.[19,41] Furthermore, western-trained surgeons usually refuse to perform uvulectomy when asked.[4] Finally, religious and/or tribal beliefs play a large role in the continuing practice of traditional uvulectomy.[12] This practice is significantly correlated with a low maternal education level.[11,12] As education improves, we hope that the frequency of this traditional practice will decrease.

Conclusion

Traditional uvulectomy is widely practised throughout many parts of the world. It is associated with many complications, partly due to unsanitary conditions, and also due to lesser known, long term adverse effects such as obstructive sleep apnoea and snoring.

Due to immigration from developing to developed world countries, and to Western doctors working abroad, such doctors are increasingly being confronted with unfamiliar traditional healing practices, within a medical context.[6,10,26] Therefore, it is important that Western-trained doctors are familiar with the techniques, cultural context and epidemiology of traditional uvulectomy, and also with its possible complications and their management.[14] It is essential to perform adequate patient history-taking and physical examination in order to establish whether traditional uvulectomy has previously been performed, in order to facilitate correct diagnosis and timely treatment.

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