

Congenital intra-oral dermoid cyst co-existing with unilateral club foot: A case report

BDO Saheeb,* BDS, FWACS, FDSRCS(Edin), FICS
PFA Umebese, *MD, D.ORTH, FCS(A), FICS

الكيس البشري الخلفي في مقدمة الفم والمتلازم مع تقوس القدم المعتدل أحادي الجانب لدى طفلة عمرها سبعة أيام تم نشره في هذا المقال . ان حدوث هذين المرضين أثناء الولادة هي حالة نادرة وشاذة . إلا أن تلامهما وظهورهما بشكل واضح عند الولادة هي حالة أندر . الكيس كان كبيراً لدرجة كافية لإعاقة الارضاع الطبيعي في الأسبوع الأول من العمر .

A congenital dermoid cyst in the anterior floor of the mouth with concomitant unilateral equinovarus foot in a seven-day old baby girl is reported. The two developmental anomalies are individually rare conditions. The occurrence of both diseases co-existing and manifesting at birth is even rarer and no such combination case, to our knowledge, has been reported. The cyst was large enough to cause obstruction and difficulty in breast feeding In the first week of life.

Introduction

Dermoid cysts of the oral cavity may be congenital or acquired. When they are congenital, they arise from epithelial rests in the midline of the floor of the mouth and ventrum of the tongue.¹ When they are acquired, they arise from epithelium implanted following trauma and occur in many sites usually not in the midline.²

Oral dermoid cyst is most commonly located in or about the midline in relationship with mylohyoid muscle, occurring generally as a swelling in the floor of the mouth with the tongue raised towards the palate.^{3,4} Although it may originate very early in life, it does not reach any symptomatic size until later in life.

Clubfoot or *talipes equinovarus* is a common foot deformity with varying degrees of severity. It presents generally with plantar flexion, inversion and adduction deformity of the foot. More than half of the presenting cases of clubfoot are bilateral and are of the mild to moderately severe type. They are thought to be due to abnormal foetal position of the foot in-utero, in what has come to be regarded as constraint induced foot deformation.⁵

However, the unilateral clubfoot which occurs in about one-third of cases is believed to be a non-constraint induced phenomenon and is regarded as a true malformation of the foot due to errors in its morphogenesis. Therefore, it often presents as the very severe type of clubfoot. Clinically, it is more rigid than the constraint-induced

deformation type and is more likely to be due to the atrophy of calf muscles and hypoplasia of the foot in the altered form.⁶

Although the pathogenesis of both dermoid cyst and clubfoot are not well understood, their concomitant occurrence in the same patient and their early manifestation in the first week of life, may suggest a common morphopathogenesis at the stages of ectodermal tissue differentiation.⁶ We report a case of congenital dermoid cyst co-existing with unilateral clubfoot in a one-week old baby.

Case Report

A one week old baby girl was referred to the Oral and Maxillofacial Surgery Clinic of the University of Benin Teaching Hospital from a private Dental Clinic with a swelling in the anterior part of the floor of the mouth. The baby had been a normal full term vaginal delivery with a birth weight of 3.1 kg. She was the first child of a 24 year-old couple. The mother had observed that the baby was losing weight because of her inability to suckle the breast.

On clinical examination, the baby was apparently healthy looking except for a cystic swelling in the midline of the anterior part of the floor of the mouth and a unilateral clubfoot of the left. She weighed 2.5 kg. The cystic swelling was dough-like in consistency, measured 3cm in diameter and elevated the tongue impeding its free movement (Fig. 1). The oral mucous membrane overlying it was normal. A tentative diagnosis of congenital dermoid cyst was made with

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* Consultant, Department of Oral and Maxillofacial Surgery;

^ Consultant, Department of Orthopaedic Surgery, University of Benin Teaching Hospital
Benin City, Nigeria

Address reprint requests to:

Dr. BDO Saheeb
PO Box 2799
Benin City, Nigeria

congenital ranula and cystic lymphangioma considered as differential diagnoses.



Fig. 1. Congenital dermoid cyst in the floor of the mouth. Difficult mouth opening.

Under general anaesthesia with nasotracheal intubation, the cystic lesion was dissected through an incision underneath the tongue down to the floor of the mouth. Before the swelling could be freed, it was accidentally punctured and a whitish material resembling desquamated keratin squames was spilled. However, the remaining outline was traced by the insertion of a finger into the sac in order to dissect it free from the mylohyoid muscles. The patient made an uneventful recovery. There was no evidence of recurrence one year after the operation.

Histopathologic examination of the specimen showed a lining stratified squamous epithelium with sebaceous material and some keratin in a cystic space. Within the connective tissue stroma there were cystic spaces lined by flattened cells. These features are consistent with those of a dermoid cyst. The left clubfoot showed a hypotrophic leg and foot, a deepened foot medial crease and a clear resistance to corrective manipulation. (Fig. 2). An orthopaedic consultation was arranged for the patient at the orthopaedic clinic of the same hospital.



Fig. 2. Left clubfoot, shorter than right foot.

Discussion

The aetiology of either congenital dermoid cyst or clubfoot is not properly understood. However, it has been suggested that congenital intra oral dermoid cysts are derived from the enclavement of epithelial debris in the midline during closure of mandibular and hyoid branchial arches.^{3,7} They are also regarded as some forms of inclusion cyst teratoma which are formed from epithelial cell rests or from proliferation of entrapped epithelium and other germ layers during embryogenesis.⁸ Although these cysts arising from the floor of the mouth are seldom present at birth, their midline location appears to be classic.⁷ The aetiology of clubfoot malformation is not known, however, most studies still appear to support the theory of muscle imbalance from arrest or embryological development.⁹

Dermoid cysts like some form of nasolabial cysts⁸ are slow growing starting usually from a small size and growing to an appreciably large symptomatic size in the first or second decade of life. Our case however at the presentation was as large as 3 cm in diameter and was present at birth causing obstructive and feeding symptoms. Furthermore, its co-existence with a unilateral hypoplastic clubfoot deformity made it a unique clinical case.

The treatment of dermoid cysts is complete surgical excision. However, the value of ultrasound and computerized tomographic scan to confirm the size of the lesion before surgery has been highlighted in some reports.^{10,11} Although these investigations were not carried out on our patient before surgery, follow up examinations did not show any recurrence. Similarly, early surgical correction of a clubfoot often give the best result, as conservative management with serial plaster of Paris case manipulation rarely corrects satisfactorily.^{5,6} After successfully treating the congenital dermoid cyst, the patient's clubfoot is being followed up in our clubfoot clinic.

In conclusion, congenital dermoid cysts arising in the anterior part of the mouth are rare. It appears that not many cases of its co-existence with a unilateral clubfoot have been reported. The loss of weight as a result of the inability of the baby to suckle breast was more worrying to the patients than the clubfoot. Complete surgical excision of the cyst still remains the best form of treatment.

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