Congenital Palatal Fistula with Cleft of the Soft Palate

Abstract

Congenital palatal fistula or perforation is rare, unlike the acquired form which commonly results from cleft palate repair. Congenital palatal fistulae are often associated with submucous cleft palate. Only a few of this fistulae are diagnosed shortly after birth. We present the case of a 3-year old girl with congenital palatal fistula coexisting with cleft of the soft palate that was noticed shortly after birth. The palate was repaired using Bardach's palatoplasty and the client was subsequently referred to a speech therapist.

Keywords: Cleft palate, congenital palatal fistula, palatal fistula

Introduction

Congenital palatal fistula or perforation is rarely reported in literature unlike the acquired form which is a common complication of cleft palate repair. [1-3] Most cases of congenital palatal fistula reported in literature occurred in patients with submucous cleft palate, [1-5] but a few occurred in isolation, with absence of submucous cleft. [3,4,6]

Only a few of the reported cases of congenital palatal fistula were diagnosed shortly after birth or in early childhood.^[1] In this paper, we present a case of a girl with congenital palatal fistula that was noticed shortly after birth. She presented at the age of three years.

Case History

We present the case of a 3-year-old girl who presented with known cleft palate and a history of poor speech, and leakage of oral feeds into the nose. There was no history of cough, nor fever. The mother noticed the fistula shortly after birth. There was no history of oral trauma and no previous surgery was done. The antenatal period and delivery were uneventful. There was no family history of cleft lip or palate. Her three older siblings died within the first year of life. She had not started any formal schooling as at the time of presentation.

On examination, she was a healthy-looking child. There was a cleft of the entire soft

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palate. An oro-nasal fistula was noted in the midline, at the middle third of the hard palate. The fistula measured 8 mm x 5 mm. The intervening tissue between the fistula and the soft palatal cleft appeared pale. This intervening tissue was deficient of bone, suggestive of a submucous cleft.

The cleft palate was repaired using Bardach's technique – 2 flaps palatoplasty. The soft tissue between fistula and cleft was divided longitudinally. Levators veli palatini were dissected out on either side as part of the surgery.

An oro-nasal fistula was noticed on follow up, about 4 months after surgery, but at a different location- the junction of hard and soft palate. It was the size of a pin hole. The fistula spontaneously closed eight months after surgery.

The mother had noticed significant improvement in speech for which she was happy, although there was still a need to improve on her speech. Patient was referred to a speech therapist and was then lost to follow up.

Discussion

There has been a question of whether these fistulae are actually congenital (a true embryological malformation) or acquired (secondary to a mucosal rupture in a submucous cleft of the palate), prenatally or postnatally. Our patient's fistula was noticed shortly after birth [Figure 1]. In the embryology of the palate, fusion of palatal shelves starts at the incisive foramen

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Figure 1: Palatal fistula occurring together with a cleft of the soft palate



Figure 2: The palate after repair of congenital palatal fistula

and progresses posteriorly to the uvula.^[6,7] A typical cleft of the secondary palate is therefore found posterior to this landmark, with the typical fistula at the junction of the hard and soft palates, where tension in the palate is maximal.^[7] Congenital fistula has been associated with submucous cleft of the palate^[1] but a few cases of congenital fistula occur in the absence of a submucous palatal cleft.^[3] Localized embryological insult from trauma or infection after formation of the palate has also been considered.^[1,3,4] Very few cases occur in association with other congenital anomalies. No other congenital anomaly was noticed in our patient, but death of three older siblings in infancy was unexplained. The pattern of presentation of congenital fistulae still requires further study.

We repaired the cleft palate and palatal fistula in our patient with Bardach palatoplasty as shown in Figure 2. Varied techniques have been used in similar repairs^[2] but Furlow's double-opposing Z-plasty technique has been used in most reported cases. In one of the reported cases, surgery could not be done because of the patient's poor cardiac condition and a palatal obturator was used instead.^[3] The use of palatal obturator is also an option for patients that refuse the option of surgery.^[2]

Conclusion

Congenital palatal fistula is a defect of the palate that may be found in patients with submucous cleft palate but can also occur in isolation. They are repaired using techniques of palatoplasty employed in cleft palate repair. Furlow's technique is the preferred method in the presence of submucous cleft palate. The pathogenesis of congenital fistula is still unsettled.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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