SVOA Dentistry

ISSN:2753-9172

ScienceVolks Open Access

Case Report

Congenital Fistula of the Lower Lip: A Case Report

Adriana Valéria Rodrigues da Silva[1], Daniel Nuciatelli Pinto de Mello[2], José Carlos Pettorossi Imparato[3], Caleb Shitsuka[4] and Irineu Gregnanin Pedron[5]*

[1]*Undergraduate Student, Universidade Brasil, São Paulo, Brazil.*

[2]*DDS, Birmingham, UK.*

[3]*Professor, Department of Pediatric Dentistry, Universidade de São Paulo, São Paulo, Brazil.*

[4]*Professor, Department of Pediatric Dentistry and Cariology, School of Dentistry, Universidade Brasil and Faculdades Metropolitanas Unidas, São Paulo, Brazil.*

[5]*Professor, Department of Periodontology, Implantology, Stomatology and Therapeutics, Universidade Brasil, São Paulo, Brazil.*

***Corresponding Author:** Irineu Gregnanin Pedron, Professor, Department of Periodontology, Implantology, Stomatology, Integrated Clinic and Therapeutics, School of Dentistry, Universidade Brasil, São Paulo, Brazil, Tel: +55-11 2944-4067*

***Received:** September 14, 2021 **Published:** September 27, 2021*

Abstract:

Congenital fistula of the lower lip is considered a rare developmental anomaly, being more frequently observed in association with cleft lip and/or cleft palate in Van der Woude Syndrome. Clinically, congenital fistulas of the lower lip are depressions or a slight increase in volume with unilateral or bilateral fistula, with or without saliva secretion. The congenital fistula of the lower lip shows genetic importance to the Dentistry, because it is known that family descendants of patients who had congenital fistula of the lower lip but did not have cleft lip and/or palate showed high probability of having this malformation. In this perspective, the dental surgeon has an important role in genetic counseling for these patients. The purpose of this article is to present a case of congenital fistula of the lower lip in a female patient, 12-years-old. The clinical features, etiology, incidence and frequency, diagnosis and treatment were discussed.

***Keywords:** congenital lower lip fistula; congenital lower lip sinuses; congenital lip pits; Van der Woude syndrome.*

Introduction

Congenital fistula of the lower lip is a rare developmental anomaly first described by Demarquay in 1845[1]. When the congenital fistulas of the lower lip occur in association with cleft lip and cleft palate it is known as Van der Woude Syndrome, described in 1954[2-4].

Clinically, congenital fistulas of the lower lip are depressions or a slight increase in volume with unilateral or bilateral fistula, with or without saliva secretion[3-5]. When the fistulas are bilateral, they are usually symmetric and equidistant from the midline. Patients with this malformation complain about the esthetic of the lip or from the frequent accumulation of saliva. However, the lesions are usually painless[3].

Diagnosis is established through clinical findings. Nevertheless, the fistula path can be seen by sialography or radiography with the insertion of gutta-percha cones[6,7].

Its etiology is still not clearly defined, but theory mentions that it occurs due to failure during fusion of the lateral and median lip processes and the maintenance of embryogenic sulci that forms the fistula tract. This malformation has a hereditary characteristic, and it is determined by dominant autosomic genes. It is slightly more common in females than males and its incidence ranges from 1:75.000 to 300.000[2-4,7-12]. The purpose of this case report is to describe the clinical findings of a girl with congenital fistula of the lower lip, as well as the etiology, frequency and incidence of this anomaly.

Case Report

A Caucasian female patient, 12-years-old, attended the dental clinic complaining about a bite on her lower lip. The clinical examination showed elevation of the lower lip tissue adjacent to the central depressions on which the orifices of the fistulas were located. The fistulas were classified as bilateral and symmetric, equidistant from the midline. The patient also showed Class II Angle relationship with protrusion of the upper central incisors that traumatized the fistula (Figure 1). However, under an air jet, the fistulas showed dry, without saliva secretion (Figure 2).

Figure 1: *Congenital fistula of the lower lip in a 12 -years-old girl.*

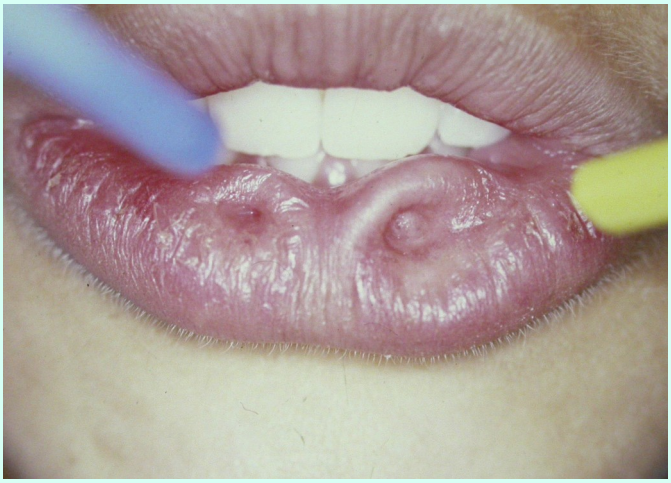

Figure 2: *Clinical findings of the fistula under air jet.*

Orthodontic treatment was performed to change the position of the upper incisors to Class I Angle relationship and removing trauma on the fistula.

After orthodontic treatment, the only complaint of the patient was the excessive salivation that caused epithelial excoriation. The patient also mentioned that these symptoms were more frequent on her left side than on the right side. The patient did not complain about esthetics in any time. Due to epithelial excoriation, the patient was medicated with miconazole gel for 15 days and was oriented to use this medication whenever the symptoms reappeared.

Discussion

Congenital fistula of the lower lip is a rare developmental malformation which has been given different names such as congenital lower lip pits[1,3,5,13], congenital sinuses of the lower lip[6,8,10,11,14,15], congenital mucosal fistula of the lip[2], accessory salivary glands[9,14], and congenital secreting cavities[2,9].

Congenital fistulas of the lower lip have an autosomic dominant mode of inheritance with a high penetrance and variable expression[2-4,7,11,15], and familiar pattern[14], that involves the gene 1q32-q41[7]. Congenital fistulas of the lower lip do not seem to be linked to gender and there is no influence of the gender on this condition[2]. Some diseases that occur during pregnancy that cause inflammation or obstruction of the glands of the lower lip due to hypertrophy or transformation may also be considered in the etiology of this anomaly. According to the embryogenic theory of Warbrick, the development of the labial fistulas apparently occurs due to persistence of a lateral sulci of the mandibular arch of human feto[2,6,8,9,11,15,16]. The lateral sulcus of the lower lip is usually obliterated, except for the final encephalic part which is located in the globular process, becoming the fistula more evident and its depth proportional to the embryogenic growth. Gradually, the sulcus edge would get more prominent and fusing itself forming a fistula. This theory may also explain why congenital fistula of the lower lip is associated to cleft lip and palate, since both of these anomalies occur at the same stage of embryogenic development.

Around 60% to 80% of lip fistulas are associated with cleft lip and/or palate, characterizing the Van der Woude Syndrome. Genetically, the congenital fistulas of the lower lip show more importance, because it is known that family descendents of patients who had congenital fistula of lower lip but did not have cleft lip and/or palate showed high probability of having this malformation[2,3,12,16]. Some authors even mention that genetic counseling should be performed whenever patient had the Van de Woude Syndrome or the congenital fistula alone[2,3,12,17].

Clinically, the fistulas may appear in different forms depending on their situation and location regarding the midline; therefore, they could be bilateral and symmetric, like in the case we have reported; bilateral and asymmetric; unilateral; median; or even microforms. They are usually situated on the vermilion border of the lip, such as in the case described above, mucocutaneous line, internal surface of the lip, or an association of these three locations[2,3,18].

On clinical examination, the fistulas appeared as nipple-shape like prominence with folds and a central depression with orifices. The hyperplasia could hide the orifices of the fistulas producing labial deformity that may even cause psychological and behavioral disorders in the patient[2]. No association among congenital fistula of the lower lip or Van der Woude Syndrome and Class II malocclusion was found.

The orifices of the fistula may be circular-shaped or slightly folded. Their depth ranges from 0,5 to 2,5 mm and the diameter of the orifice can measure up to 3 mm. Many authors mention that the fistula has the potential of salivary secretion[2,3,7,11,16,18]. However, some cases, as presented here, do not show salivary secretion[15]. The microforms of this anomaly are bilateral conical elevations with one central depression on the lower lip vermilion. In this type, there is no salivary secretion. Higher salivary secretion may be noticed at mealtimes or whenever patients are under pressure[2,6]. In our report, the patient complained of the frequent accumulation of saliva which, associated with the habit of biting her lower lip, also caused epithelial desquamation[4,15]. These anomalies usually do not show a tendency to obstruction or infection[2]. However, as seen in this patient, epithelial excoriation can be observed due to saliva stagnation. In this case, we used the miconazole instead of nystatin because it is a gel, lasts more time on the surface, and has a better effect.

The frequency in the white population is higher than the one observed in Asian and black[8,9,18]. It is more common in females than males[2,9,15,18]. The patient here reported also is white female with no other congenital alteration. In most of the cases, congenital fistulas of the lower lip are associated with the cleft lip and/or palate. The type of lip fistulas is related to the severity of the cleft lip and/or palate. Consequently, bilateral, unilateral, and mixed lip fistulas are usually associated to cleft lip and/or palate, while the microforms and conical elevations would be related to cleft palate or submucosal[18]. The most common type of this malformation is the bilateral fistulas, followed by the median and unilateral ones[15]. The patient, whose case was described, showed bilateral and symmetric fistulas.

Familial occurrence of congenital fistulas of the lower lip involving members of the same family is reported[1,13]. There were no other members family of the patient that had congenital fistulas of the lower lip.

The recommended treatment is surgical removal. When a minor salivary gland is involved, surgical removal of the fistulous path and the involved salivary gland should also be performed[4]. The diagnosis was deeply investigated, and the family of the patient was informed about this alteration. It was suggested to seek for plastic surgery for cosmetic/esthetic reasons.

Conclusion

Although congenital fistula of the lower lip is a rare developmental anomaly, it is important to the dental surgeon to have knowledge and transmit information in order to adequately direct treatment, when necessary. Additionally, the dental surgeon is responsible for providing genetic counseling to patients and their families.

References

1. Zarandy MM, Givehchi G, Mohammadi M. A familial occurrence of lip anomaly. Am J Otolaryngol 2005;26(2):132-134.

2. Onofre MA, Taga R. Fistulas Congênitas de lábio inferior na Síndrome de Van der Woude. Uma revisão. Rev Fac Odontol Baurú 1994;2(1):35-41.

3. Scheiba N, Hartschuh W. van der Woude syndrome. Dtsch Arztebl Int 2020;117(44):751.

4. Douhi Z, Mernissi FZ. Small pits of the lower lip: think paramedian fistula lip. Pan Afr Med J 2014;19:203.

5. Dissemond J, Haberer D, Franckson T, Hillen U. The Van der Woude syndrome: a case report and review of the literature. J Eur Acad Dermatol Venereol 2004;18(5):611-613.

6. Bernaba JM, Castro AL, Garrafa V. Fístulas congênitas bilaterais do lábio inferior e comissuras labiais - Relato de dois casos. Rev Assoc Paul Cirurg Dent 1973;27(1):17-21.

7. Merino CC, Ballestra CG, Sanchez REO. Congenital labial fistulas: case report. J Clin Pediatr Dent 1997;21(4):325-328.

8. Iregbulem LM. Congenital lower lip sinuses in Nigerian children. Br J Plast Surg 1997;50(8):649-650.

9. McConnel FMS, Zellweger H, Lawrence RA. Labial Pits - Cleft Lip and/or Palate Syndrome. Arch Otolaryngol 1970;91(5):407-411.

10. Mutaf M, Sensoz O, Ustner ET. The split lip advancement technique for the treatment of congenital sinuses of the lower lip. Plastic Reconstr Surg 1993;92(4):615-620.

11. Srivastava S, Bang RL. Congenital Sinuses of the lower lip reappraisal of Van der Woude Syndrome on the basis of nine patients. Ann Plast Surg 1989;22(4):316-320.

12. Taga R, Onofre MA, Brosco HB et al. Ultra-estrutura da Parede da Fistula Congênita de Lábio Inferior na Síndrome de Van der Woude. Rev Fac Odontol Baurú 1998;6(1):29-37.

13. Calista D. Congential lower lip pits. Pediatr Dermatol 2002;19(4):363-364.

14. Bowers DG. Surgical repair of congenital lower lip sinuses. Plast Reconstr Surg 1972;49(6):632-636.

15. Lopes MA, Gonçalves M, Di Hipólito Jr O, Almeida OP. Congenital lower lip pits: case report and review of literature. J Clin Pediatr Dent 1999;23(3):275-277.

16. Onofre MA, Brosco HB, Brosco JU, Taga R. Congenital fistulae of the lower lip in Van der Woude Syndrome: a histo-morphologycal study. Cleft Palate Craniofac J 1999;36(1):79-85.

17. Rizos M, Spyropoulos MN. Van der Woude syndrome: a review. Cardinal signs, epidemiology, associated features, differential diagnosis, expressivity, genetic counselling and treatment. Eur J Orthod 2004;26(1):17-24.

18. Onofre MA, Brosco HB, Taga R. Relationships between lower lip fistulae and cleft lip and/or palate in Van der Woude Syndrome. Cleft Palate Craniofac J 1997;34(3):261-265.

Citation: da Silva AVR, de Mello DNP, Imparato JCP, Shitsuka C, Pedron IG. "Congenital Fistula of the Lower Lip: A Case Report". SVOA Dentistry 2:6 (2021) Pages 267-270.