

Congenital double lip: A report of two cases, one associated with Ascher's syndrome, highlighting clinical presentation and surgical management of a rare orofacial deformity

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Informed Consent

The authors stated that the written consent was obtained from the patient presented with images in the study.

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Abstract

Double lip is an uncommon oral anomaly characterized by an additional fold of redundant mucosal tissue on the inner aspect of the lip, most frequently affecting the upper lip. It may present as an isolated finding or as part of Ascher's syndrome, which comprises the triad of double lip, blepharochalasis, and non-toxic thyroid enlargement. This condition primarily causes esthetic and functional concerns, affecting speech and smiling. Diagnosis is clinical, and surgical excision is the standard treatment to restore normal esthetics and function. This report describes two adult male patients with congenital double upper lip, their clinical presentation, and surgical management, which yielded excellent esthetic and functional results. The association of this condition with Ascher's syndrome is also presented and reviewed.

Keywords: double lip, Cupid's bow shape, Ascher's syndrome, transverse elliptical excision

Introduction

Double lip is one of the rarest forms of lip anomaly. It most commonly occurs in the upper lip [1], however, cases involving the lower lip or both lips have been reported. The anomaly is characterized by the presence of an excessive fold of areolar tissue and hypertrophy of the minor salivary glands without involvement of the surrounding muscles [2]. Double lip manifests as redundant tissues on either side of the midline, often asymmetrically, with one side being larger than the other [3]. The prevalence of double lip in the upper lip far exceeds that of the lower lip [4].

Double lip can be either congenital or acquired. The acquired form primarily arises from trauma or oral habits, such as lip sucking between a diastema or ill-fitting prosthetic restorations. The congenital variant typically develops during the first trimester of fetal growth. The upper lip mucosa is divided into an outer *pars glabrosa* and an inner *pars villosa*; between the second and third months of gestation, the *pars villosa* may hypertrophy due to the persistence of an exaggerated horizontal sulcus between the two layers, leading to its development [5].

The congenital type may also occur in association with a syndrome first described in 1920 by an ophthalmologist from Prague named Ascher [6]. The syndrome is characterized by recurrent edema of the upper eyelid and upper lip, resulting in blepharochalasis and double lip, with concurrent non-toxic goiter. This constitutes the classical (complete) Ascher syndrome. However, 10%-50% of cases do not present with non-toxic goiter (Incomplete Ascher's syndrome), and its absence does not preclude the diagnosis [7]. No definitive racial or gender-related predilection has been observed, but one study by Palma and Taub in 2009 showed a male predilection of 7:1 [8].

Surgical intervention remains the mainstay of treatment for double lip, with excision undertaken to improve esthetics and function. This case report aims to contribute to the limited literature on double lip by presenting its association with Ascher's syndrome, thereby enhancing awareness and guiding the management of this rare anomaly. Here, we discuss two patients with double lip managed by the author.

Case presentation

Case 1

A 35-year-old male presented to the Dental department at Bugando Medical Center with a chief complaint of gradual upper lip enlargement over the past 15 years. The condition was first noticed at age 19 and had progressively enlarged. He reported an unpleasant appearance that caused embarrassment during speech, which was problematic as he is a high school teacher. The patient denied any history of trauma, parafunctional oral habits, or previous surgery on the upper lip. The patient reported no family history of a similar condition. His medical history was unremarkable.

On clinical examination, the patient had a visible double upper lip that was more accentuated when he smiled. A central constriction dividing the upper lip was observed, with more tissue prominence on the right side than the left. The excess tissue was less conspicuous when the patient's lips were at rest (Figure 1). The labial sulcus depth and frenal attachment were normal. The patient also had drooping upper eyelids with excess skin folds, giving him a "tired" appearance; this confirmed the presence of blepharochalasis (Figure 1A). Thorough clinical examination revealed no signs of thyroid enlargement. All routine bloodwork and thyroid function tests were normal. A clinical diagnosis of incomplete Ascher's syndrome was established. The patient was planned for lip reduction surgery under local anesthesia after obtaining informed consent.

Anesthesia was administered via deep bilateral local infiltration with 2% lignocaine. Subsequently, a transverse elliptical incision was made from one commissure to the other using a size 15 scalpel blade, crossing the midline of the frenulum (Figure 2). Blunt dissection was performed submucosally with scissors, sparing the orbicularis oris muscle and avoiding over-extension to ensure excellent post-operative lip competence and symmetry. The minor salivary glands in the surgical field were removed to prevent future mucoceles. Hemostasis was achieved, and the surgical defect was sutured in layers using interrupted Vicryl 3-0 sutures. The patient was prescribed analgesics and antibiotics for one week and provided with post-operative instructions. The patient was followed up after two weeks (Figure 3). There were no complications, healing was uneventful, and the patient was satisfied with the postoperative results.

Figure 1. Pre-operative clinical presentation of Case 1. (A) Note the blepharochalasis of both upper eyelids and the double upper lip at rest. (B) The double upper lip is more accentuated on the right side, with a central constriction.



Figure 2. Intra-operative view (Case 1) showing the transverse elliptical incision line extending from commissure to commissure.



Figure 3. Post-operative view (Case 1) at two-week follow-up, showing satisfactory esthetic outcome.



Case 2

A 29-year-old male presented to our facility with a chief complaint of "thick bulging tissues" under his upper lip, causing esthetic concern when speaking and smiling. According to the patient, the anomaly had been present since he was young and became more apparent as he aged, having progressively enlarged over time. Similar to the first case, the patient was systemically healthy with no prior history of trauma, oral habits, or surgery on the upper lip. There was no reported family history of the condition. The patient had neither blepharochalasis nor thyroid gland enlargement. A clinical diagnosis of congenital double upper lip was established (Figure 4A-B).

On clinical evaluation, he presented with an obvious double upper lip at rest and an inflated appearance when smiling. The double lip manifested as two soft folds of excess mucosal tissue on either side of the midline, featuring a central constriction that gave a characteristic "Cupid's bow" appearance (Figure 4A). The mucosal folds were soft and non-tender on palpation. The maxillary labial frenal attachment was positioned at the gingiva with a normal sulcus depth. The patient was scheduled for reduction cheiloplasty under local anesthesia.

A bilateral infraorbital nerve block with 2% lignocaine was performed, rather than local infiltration, to prevent tissue distortion during excision. Using a size 15 surgical blade, bilateral elliptical incisions were made around the excess labial tissues, with 2-mm extensions beyond the commissure on both sides, without involving the frenum. Blunt dissection followed, extending inferiorly to the submucosa so that a wedge-shaped block of tissue could be removed. The orbicularis oris muscle was preserved, and the accessory salivary glands were removed to prevent the development of mucoceles (Figure 5). Hemostasis was achieved through local compression, and the resulting defect was closed in layers using interrupted Vicryl 3-0 sutures. Post-operatively, the patient was prescribed oral analgesics and

antibiotics for one week, with reinforcement of post-operative and oral hygiene instructions. At the two-week post-operative follow-up (Figure 6), the surgical site was free of complications, and the patient's esthetic and functional concerns were satisfactorily achieved.

Figure 4. Pre-operative clinical presentation of Case 2. (A) Bilateral double upper lip with a characteristic "Cupid's bow" appearance at rest. (B) Symmetrical appearance with a central constriction.

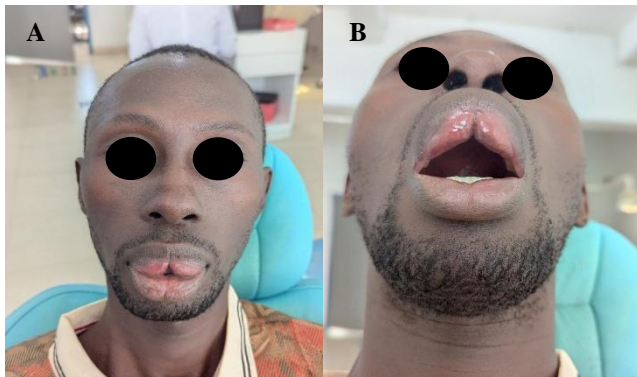


Figure 5. Intra-operative view (Case 2) showing the bilateral transverse elliptical excision, sparing the frenum. Note that the orbicularis oris muscle fibers are preserved.



Figure 6. Post-operative view (Case 2) at two-week follow-up, showing excellent esthetic results.



Discussion

Congenital double lip is a benign, non-inflammatory hypertrophy of the lip [9]. The exact etiology is not recognized, but studies propose it results from the persistence of the horizontal sulcus between the *pars glabra* and *pars villosa* [5]. Such enlargements are usually bilaterally symmetrical and typically take the form of a "Cupid's bow" when the upper lip is tensed [10]. The involvement of the upper lip far exceeds that of the lower lip, as seen in the two cases presented here. In both cases, there was no history of trauma or mentioned oral habits.

Although it is generally reported that the double lip is not noticeable when the patient's lip is at rest but becomes apparent when the lip is tensed (e.g., smiling), the deformity in both our

reported cases was obvious even at rest. Differential diagnoses for double lip should encompass hemangioma, cheilitis glandularis, and angioedema. It is particularly important to rule out cheilitis glandularis, as it is associated with a higher risk of squamous cell carcinoma [4] and thus requires histopathological clarification. These differentials typically do not present with the central constriction seen in double lip [5], which is often a distinguishing clinical feature.

In very rare cases, congenital double lip has been associated with bifid uvula and cleft palate [11]. Although double lip is present at birth, it often becomes more obvious after the eruption of permanent teeth and can be exacerbated by oral habits, leading to its presentation during adulthood. This impacts self-esteem and provides the rationale for its correction [12]. It is possible that persistent, unrecognized oral habits aggravated the condition in our patients. Double lip has been reported by various authors to have a male predilection; this is consistent with our case report, in which both patients were male.

Congenital double lip can present as part of Ascher's syndrome, characterized by the additional presentation of blepharochalasis and non-toxic thyroid enlargement [13]. The syndrome often does not involve all three presentations and may be termed incomplete Ascher's syndrome. Thyroid enlargement is present in only 10%-50% of cases and is seldom involved [14], as was true in this article where the patients' thyroids were normal. In addition, blepharochalasis has been reported to present in 80% of cases. It usually manifests during puberty but is often not of as much cosmetic concern to the patient as the double lip [15], which is consistent with our first case. This condition can mimic other conditions, such as Melkersson-Rosenthal syndrome and Mounier-Kuhn syndrome [16].

Ascher's syndrome is frequently undiagnosed because of its rarity. In most cases, a dental surgeon or an ophthalmologist is the first to diagnose it [17]; this could be due to the patient's chief complaints relating to the head and neck region. Initial management with Dapsone 50 mg daily has been documented in an attempt to slow the syndrome's progression [18]; however, the mainstay of treatment is surgical correction for esthetic reasons.

Several surgical methods have been described for the correction of double upper lip, including W-plasty, electrosurgical removal, and triangular excision [19]; however, the most commonly recommended technique is transverse elliptical excision due to its ease of implementation and excellent post-operative results. In cases where significant tissue excision is required, a W-plasty can be performed for better anatomical results [20]. In both our cases, a transverse elliptical excision was carried out, as primary closure of the defect was feasible and yielded satisfactory results. This aligns with suggestions by most authors, where the mucosal and submucosal parts of the redundant tissue are excised while sparing the orbicularis oris muscle to prevent functional loss and allow primary closure [21].

Conclusion

Dental surgeons are often the first to diagnose congenital double lip. Their expertise allows them to identify related syndromic features, such as in Ascher's syndrome, which may not be the patient's primary complaint, facilitating a multidisciplinary management approach. Although congenital double lip is rare, diligent follow-up remains essential, particularly in suspected

Ascher syndrome cases, to ensure optimal outcomes. Further research on syndromes related to double upper lip is needed for better clinical judgment in distinguishing these closely related conditions, thereby tailoring management for patients based on an accurate diagnosis.

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