

# Non Syndromic Upper Double Lip: A Rare Case Report.

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## ABSTRACT

Double lip is a rare developmental anomaly. It may present as a single entity or as a feature of a syndrome. It usually causes aesthetic and functional problems, with aesthetic consideration the major reason for seeking medical intervention. We present a report of a 35-year-old male with this rare anomaly treated surgically.

**Keywords:** Double lip, upper, non-syndromic, elliptic incision.

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## INTRODUCTION

Double lip is a rare developmental anomaly, which affect the upper lip more commonly than the lower lip.<sup>1</sup> It is characterized by the presence of fold of excess or redundant labial tissue which is apparent at rest or on smiling.<sup>2</sup> It is also referred to as macrocheilia or hamartoma.<sup>3</sup>

It manifests as two fold of hyperplastic tissue on either side of the midline,<sup>1,4</sup> though a unilateral double lip has been reported.<sup>1</sup> Sometimes, bilateral hyperplastic tissues are asymmetrical with one side bigger than the other.<sup>5</sup> There is no race or gender predilection,<sup>6,7</sup> but Palma and Taub<sup>5</sup> documented a male predilection of 7:1 with a prevalence rate of <1/1,000,000.<sup>8</sup>

Double lip may be congenital anomaly or an acquired deformity.<sup>9</sup> The congenital type is a developmental anomaly<sup>10</sup> and usually affect the upper lip more than the lower lip.<sup>11</sup> Although, cases of simultaneous involvement of both upper and

lower lips have been reported.<sup>5,7</sup> It may be present at birth and become more prominent as the patient grows.<sup>4</sup> It may occur in isolation or as a part of a syndrome<sup>12</sup>. While the acquired type may be secondary to trauma<sup>10</sup> or oral habit such as sucking lip between diastema<sup>1</sup> or between ill-fitting dentures.<sup>1,6</sup>

The treatment of choice for double lip is surgery and usually indicated for aesthetic reason when it leads to facial disfigurement or for functional reasons when it interferes with mastication and speech. Recurrence is said to be extremely rare in congenital cases.<sup>13</sup>

We present a report of a case of a 35-year old male with this rare anomaly treated surgically.

## CASE REPORT

A healthy 35-year-old male presented to our clinic with a complaint of poor aesthetics due to upper lip

swelling since his teenage age of 17 years, which gets accentuated during smile and speech.

Lip swelling was not associated with pain, discharge, history of trauma or surgery in the past. Family and social history, past medical history and dental history were non-contributory. The patient was systemically healthy, no history of systemic disease in particular thyroid or renal disease.

Clinical examination revealed visible two folds of redundant mucosal tissues bilaterally on either side of the midline of the upper lip, which is accentuated when the patient smiled or beared his teeth (Figure 1). The swelling was soft in consistency and painless on palpation. There was no drooping of the upper eyelids and no thyroid enlargement.

Intraorally, occlusion was normal, with no midline diastema, vestibular depth appeared normal with adequate width of attached gingiva in the maxillary anterior region. A provisional diagnosis of non-syndromic upper double lip was made and surgical excision was planned under local anaesthesia.

After the infiltration of local aesthetic agent using 2% lignocaine hydrochloride with 1:80,000 adrenaline, a transverse elliptical incision was made using No.15 Bard Parker blade on the vestibule of the upper lip, blunt and sharp dissection was done to excise the redundant hyperplastic tissue, while care was taken not to excise normal lip tissue to avoid loss of lip dimension and asymmetry postoperatively (Figure 2).

Surgical site was irrigated with normal saline, homeostasis achieved with gauze pack and was closed with 3/0 vicryl suture (Figure 3).

Post-operative instruction was given and Amoxicillin 500mg 8 hourly for five days, Metronidazole 400mg 8 hourly for five days and Paracetamol 1g 8 hourly for 3 days were prescribed for the patient. Sutures were removed 7 days post-operatively and healing was satisfactory with patient satisfied with the result of the surgery and his smile was thoroughly improved (Figure 4).

a



b



Figure 1: Preoperative clinical photograph showing a) Upper lip at rest. (b) Accentuation of upper double lip on smiling



Figure 2: Surgical correction of upper double lip



Figure 3: Sutured wound after excision of upper double lip



**Figure 4:** Seven (7) days postoperative view photograph

## DISCUSSION

Double lip is a non-inflammatory enlargement of the lip either due to glandular tissue hyperplasia or due to persistence of the horizontal sulcus between the developing parts of lip, namely pars glabrosa and pars villosa during the second to third week of gestation.<sup>14</sup>

Double lip may present as a single entity or as a feature of a syndrome. The exact cause is unknown but may be transmitted as an autosomal dominant disorder.<sup>10</sup> Double lip has been reported as a part of syndromes.<sup>14-16</sup> However the case presented here was a solitary presentation.

Clinical features of congenital double lip though present at birth become apparent after the eruption of the permanent teeth.<sup>1</sup> In this report the patient started noticing this anomaly during his teenage years which is in line with the literature.<sup>1</sup> It is generally documented that the upper double lip is not evident at rest but when the lip is tensed as during laughing or attempting to show teeth,<sup>4,10</sup> this is attributed to the contraction of orbicularis oris during smiling causing the lip to retract and the mucosa to be positioned on the maxillary teeth,<sup>1</sup> however few cases of double lip showing deformity at rest have been documented.<sup>15</sup> In this case report, it became evident during smiling or at an attempt to bear the teeth (Figure 1b).

Most cases of this condition present with aesthetic concern,<sup>1</sup> very few show functional interference.<sup>16</sup> Our patient presented with aesthetic concerns only. Management is aimed at both the functional and aesthetic improvement. Surgical excision is the treatment of choice for double lip.

Surgical excision is carried out under either local anaesthesia, using infraorbital nerve block with or without deep ring infiltration or under general anesthesia.<sup>16</sup> Deep ring infiltration without infraorbital nerve block was used for our patient and the procedure was well tolerated by the

Patient and no anatomical distortion was observed.

A variety of surgical approaches for correction of double lip have been described which include transverse elliptical incision,<sup>11</sup> Z-plasty,<sup>4</sup> W-plasty,<sup>18</sup> triangular excision, electrosurgical excision<sup>3</sup> and laser<sup>17</sup>.

The choice of surgical approach depends on preferences and experience of the surgeon. In this reported case, transverse elliptical incision was used. The result after seven (7) days postoperatively was highly acceptable with patient satisfaction (Figure 4).

Histopathological examination show hyperplastic mucous glands, loose areola tissue, numerous blood filled capillaries and perivascular infiltration with plasma cells and lymphocyte.<sup>12</sup> The histopathology examination for our reported patient showed normal labial mucosa with abundant mucosa glands and capillaries.

## CONCLUSION

Non-syndromic upper double lip is of special interest in the head and neck region due to its rare occurrence and the attendant aesthetic and functional consequences. Surgical management gives a satisfactory result.

## CONFLICT OF INTEREST

The authors declare that they have no conflicting interest in whatever form in the publication of this article.

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