Abstract: Double lip is a rare developmental anomaly which usually involves the upper lip than lower lip. Infrequently it may be seen in upper and lower lips concurrently. It may be congenital or acquired. Its occurrence in isolation or as a constituent of Ascher’s syndrome is well documented. Congenital double lip is a rare oral anomaly. Surgical treatment is indicated more often for aesthetic reasons when the excess tissue interferes with functionality. The results are generally very good and complications are extremely rare. We describe a case of young boy presenting with congenital upper double lip, along with etiology, clinical presentation and treatment plan and review of literature. Congenital maxillary double lip is one of the unusual forms of lip abnormality. It not only interferes with speech and mastication but also affects the facial aesthetic. Though aesthetic is prime concern, surgical correction of the anomaly may be required to assist speech and mastication.

Key words: Ascher’s Syndrome, Double lip, hamartoma, Lip anomaly, macrochelitis.

Introduction: Double lip is an unusual lip anomaly [1]. It is also known as macrocheilitis or hamartoma [2]. It may be a congenital or acquired anomaly. Usually the involvement of upper lip than the lower lip is well known. Infrequently, both upper and lower lips may be involved. Double lip can occur in isolation or as a component of Ascher’s syndrome. It usually occurs bilaterally though it can occur unilaterally also in one or both the lips [3]. There is no gender or race predilection [4]. Some authors have suggested a male predilection of 7:1[1]. The treatment is surgical and usually indicated for cosmetic reason. Recurrence is rare.

Case Report: A 16 year old male patient reported to our Oral and Maxillofacial surgery department with a chief complaint of swelling of upper lip.
Figure 1: Preoperative front view showing no evidence of double lip in relaxed posture

The swelling was congenital as narrated by patient’s mother. No relevant medical history was recorded. The boy did not present with any functional abnormalities of speech and mastication. On examination, the swelling measured 1x1.2 cm and extended up to labial mucosa intraorally.

Figure 2: Close up frontal view of double lip

The mucosa over the swelling was of same color as that of adjacent mucosa. The swelling was visible only when the patient smiled or opened mouth or stretched the lips. On palpation the swelling was soft in consistency and mobile. Associated occurrence of blepharochalasis and non-toxic enlargement of thyroid was ruled out by thorough clinical examination. A Cosmetic surgical correction was planned under local anaesthesia as day care surgery. Markings for an intraoral elliptical incision were made. Anaesthesia was obtained by bilateral infraorbital block and local infiltration with Xylocaine 2% with 1:200000 adrenaline. Excision of double lip was done and closure obtained in single layer using 3-0 Vicryl.

Figure 3: Excision of hyperplastic upper labial tissue

Figure 4: Closure

Postoperatively, patient was given antibiotics and analgesics for 5 days. The postoperative recovery was uneventful.

Figure 5: Excised tissue specimen
Histological examination of excised tissue, showed, parakeratinized stratified squamous epithelium with underlying well-vascularized fibrous connective tissue and, deep to this, numerous minor salivary glands were seen.

Discussion:
Double lip is an infrequent anomaly more commonly affecting upper lip, though lower lip or both the lips can be affected. The incidence of this anomaly is not yet known [5]. In the foetus, the mucosa of the upper lip is divided into two transverse zones namely, an outer zone [pars glabrosa] and an inner zone [pars villosa]. It is in the form of two masses of hyperplastic tissue on either side of the midline caused by excessive aerolar tissue and non inflammatory labial mucosa gland hyperplasia of the pars villosa. It is thought to arise during 2nd and 3rd month gestation, from perseverance of an exaggerated horizontal sulcus between the pars glabrosa and pars villosa of the developing lips [3, 4]. Clinically, during smiling and laughing, the lip is retracted and the mucosa gives appearance of “cupid’s bow” whereas it is not evident when the mouth is closed [2,6]. It can be congenital or acquired anomaly [1]. Acquired deformity may be secondary to trauma or habits such as sucking lips between diastema or ill fitting dentures. The congenital double lip is present since birth but becomes prominent after eruption of teeth [1,7,8]. Double lip may occur in isolation or in association with Ascher’s syndrome, bifid uvula, cleft palate and cheilitis glandularids [8]. Differential diagnosis of double lip includes hemangioma, lymphangioma, angiodema, cheilitis glandularis, cheilitis granulomatosis [3,4] vascular tumor, Meischer syndrome, mucocele, salivary gland tumors, inflammatory fibrous hyperplasia, sarcoidosis and plasma cell cheilitis [3,4,8]. It is important to distinguish double lip from cheilitis glandularis, as the latter is coupled with increased risk of development of squamous cell carcinoma.

The treatment of double lip is surgical excision and is required mostly due to cosmetic reasons. Sometimes surgery may be required when the hyperplastic tissue interfere normal function such as speech and mastication [2,4,9]. Treatment should be accomplished by excision of the mucosa and sub mucosal tissues, without involvement of the underlying muscle. Good functional and cosmetic results is accomplished by surgical excision via transverse elliptical incision or Z-plasty or W-plasty, or triangular excision using electrocautery or blade [2,4,6,8].

Conclusion:
A double lip is an infrequently occurring anomaly which may appear by itself or in combination with other abnormalities. The presence of double lip should raise a question in mind of a surgeon, to rule out various other conditions associated with it. It is imperative to distinguish double lip from cheilitis glandularis, as it is coupled with increased risk of development of squamous cell carcinoma. Evaluation and follow up of these patients should be maintained.

Acknowledgement:
Authors acknowledge the immense help received from the scholars whose articles are cited and included in references of this manuscript. The authors are also grateful to authors/editors/publishers of all those articles, journals and books from where the literature for this article has been reviewed and discussed.

Source of Funding: Nil
Conflict of Interest: None

References: