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Case report

Recurrent congenital double upper lip: A case report

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ABSTRACT

Double lip is a rare anomaly characterized by hyperplastic mucosal tissue. It may occur either congenital or acquired. Surgical treatment, carried out under either local or general anesthesia, is indicated for cosmetic reasons and produces good functional and cosmetic results. Several surgical methods have been used, such as transverse elliptical excision, W-plasty, electrosurgical excision. Post-operative recurrence is rare. The present report describes a rare case of post-surgery recurrent double upper lip, necessitating a second operation. A literature review presents the origin of this anomaly, the method of surgery, and the histology of the double lip, and the two operations are compared.

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1. Introduction

Double lip is a rare oral anomaly caused by hyperplastic tissue of the labial mucosa, affecting the upper lip more commonly than the lower [1]. The mucosal fold is not usually visible with the mouth closed, but becomes more prominent with tension is caused by smiling or talking [2]. Clinically, the double lip has a horizontal running duplicate located between the inner and outer zones of the lip, and surgical excision of the mucosal fold produces good functional and cosmetic results. However, few cases of post-operative recurrence of the double lip have been reported. Here, we report a case of recurrent double lip in a 17-year-old male patient who was treated by excision of the mucosal fold and submucosal tissues.

2. Case report

A 17 year-old male patient was referred to the Niigata University Medical and Dental Hospital Department of Oral and Maxillofacial Surgery with the chief complaint of prominent upper labial mucosa when he smiled. His medical history included asthma when he was child, but was otherwise unremarkable. There was no family history

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of double lip and no previous history of trauma. One year before his visit to this hospital, he had undergone surgery with Z-plasty on the upper labial frenum to improve his double lip at a previous hospital a year before his first visit to ours; nevertheless, there was no improvement. The form of the lip at rest was nearly normal (Fig. 1A), but the mucous membrane of the upper lip was prominent when the patient smiled or showed his teeth (Fig. 1B).

We planned to perform surgery with the patient under general anesthesia. We placed a symmetrical spindle-shaped incision line along mucosal prominence that appeared when the patient smiled and showed his teeth before the induction of general anesthesia. After administration of infiltration anesthesia around the incision line, the hyperplastic tissue above the submucosal tissue was removed because we could not identify the hyperplastic tissue volume (Fig. 2A, B). The wound was closed by non-resorbable monofilament suture at the incision site. Histologic examination revealed small amounts of nearly normal lip mucosa with hyperkeratosis, coarse collagen fiber, and occasional lymphocyte. These findings indicated the presence of chronic inflammation that, likely caused by mucosal irritation associated with lip movement (Fig. 3).

However, 6 months after surgery the patient's upper labial mucosa again became prominent (Fig. 4). Therefore, 2 years and 5 months after the first surgery, we performed a second surgery. The patient was kept under bilateral infraorbital nerve blocks and infiltration anesthesia around the lip to determine the appropriate incision line, and under intravenous sedation to evaluate the

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Fig. 1. Clinical finding of lip at rest before surgery. The form of lip at rest was almost normal (A), but the mucous membrane of upper lip was prominent when the patient smiled or showed his teeth (B).

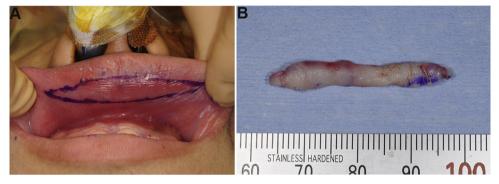


Fig. 2. The spindle-shaped incision line and a removed tissue at the first surgery. The hyperplastic tissue was removed above the ubmucosal tissue (B) by a symmetric spindle-shaped excision (A).

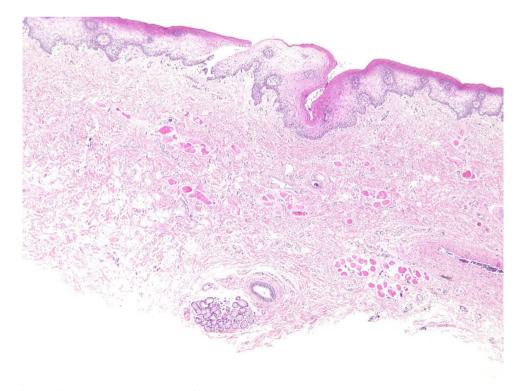


Fig. 3. Histopathological findings of the excised lip specimen at the first surgery.

Small amounts of almost normal mucosa of lip with hyperkeratosis and coarse collagen fiber, occasionally coming across the invasion of lymphocyte [H&E 10×].

lip prominence when the patient showed his teeth. An elliptical incision line was used, and the hyperplastic tissue above the muscle layer was removed, and the wound was primary closed

(Fig. 5A, B). Histology revealed a section of soft tissue covered by stratified squamous epithelium with parakeratosis, fibrous granulation tissue with hyperplastic capillaries, and numerous minor or

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