

## Case Report

# Secondary cleft deformities: case report and review of literature

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### ABSTRACT

The aim of this paper is to highlight the iatrogenic aspect of secondary cleft deformity, methodologies to treat them and the importance of reporting such cases. Secondary deformities are common in cleft lip and palate patients. Primary aim of treatment in cleft patients is to enhance their normal growth and development and minimize morbidity and number of operative procedures. Meticulous diagnosis, treatment planning and execution of primary surgery are of utmost importance in such cases to prevent majority of secondary deformities. Treatment of secondary deformity depends on the degree of deformity and the severity of impact on normal functions and growth. Following is a case of 16-year-old female patient who incurred secondary cleft deformities after undergoing multiple surgical interventions for congenital bilateral cleft lip alveolus and palate, last of which was conducted at the age of 6 years. Lip revision and tongue flap surgeries to close the palatal fistula were performed to address the patient's complaint and improve quality of life. Performing a tongue flap for anterior palatal fistula was more successful than Bardach's palatoplasty technique. It is important that each case encountered at various congenital defect care facilities is reported in literature to make the masses aware of probable outcomes and also help maintain a database to have more accurate data of such cases.

**Keywords:** Secondary cleft deformities, Cleft palate, Palatal fistula

### INTRODUCTION

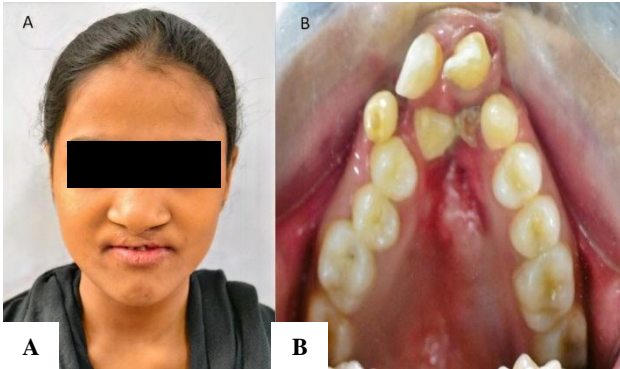
Isolated cleft lip (CP) or a combination of cleft lip and palate (CLP) is one of the most common congenital deformities reported in literature. With almost 25 million births per year, India records 27,000 to 33,000 cases of clefts per year.<sup>1</sup> No optimal time for cleft repair has been documented, however, it is generally followed that at least soft palate repair when required should be conducted between 6 months to 1 year of age to improve the child's quality of life.<sup>2,3</sup> Primary treatment of clefts if not carried out in a planned manner is often followed by complications leading to secondary deformities hampering growth and morbidity. Thus, it is important to thoroughly analyze and investigate such a defect when encountered and carry out a multidisciplinary team

approach which includes a maxillofacial and plastic surgeon, orthodontist, speech therapist and orthognathic consultation if required.

### CASE REPORT

A 16-year-old female patient was referred to the department of Oral and Maxillofacial Surgery (OMFS) from the department of orthodontics and Dentofacial Orthopedics with chief complaint of unesthetic upper lip and hole in the roof of the mouth since 10 years. The patient was diagnosed with bilateral cleft lip alveolus and palate at the time of birth. To address the same, she underwent multiple corrective surgeries, last of which was conducted 10 years ago.

A complete patient history was taken. Patient did not report any previous relevant medical and surgical complications or known drug allergies. Following this, she was comfortably positioned in a dental chair and examined for extra- and intra-oral deformities. Clinical examination revealed secondary deformities of lip and palate (Figure 1 A and B). Upper lip was short, immobile and adherent to the underlying bone with exposed premaxilla and incisors. V shaped high arched palate was present with crowding of maxillary anterior teeth. A palatal fistula, measuring 1 cm in diameter was situated immediately posterior to the premaxilla.



**Figure 1: Preoperative (A) extra-oral and (B) intra-oral findings.**



**Figure 2: Patient 1 month after cheiloplasty procedure.**

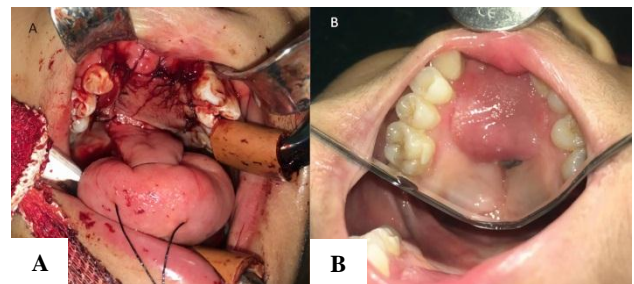
To address the patient's complaint a two-stage corrective surgery was planned. In the presence of the patient, her parents were informed and explained about the treatment plan and procedures to be undertaken for resolving the problem along with expected outcomes and the involved risks. A written, signed informed consent was taken from parents to conduct the planned and discussed treatment.

In the first stage of surgery a lip revision was done to address the immobile lip and scar from the previous

surgery. Lip musculature was relieved from the underlying bone and lined with adjacent oral mucosal flaps to form labial mucosa (Figure 2). After 6 months, the anterior palatal fistula was closed using Bardach's palatoplasty technique (Figure 3). Post-operatively the patient developed allergic rhinitis leading to flap failure and recurrence of fistula. Subsequently 1 year later, an anteriorly based tongue flap was used to close the palatal fistula (Figure 4 A and B). The tongue flap was divided after 3 weeks. The patient now awaits alveolar bone grafting and further cosmetic surgeries.



**Figure 3: Intraoral finding 1 month after Bardach's palatoplasty.**



**Figure 4: (A) Intraoperative tongue flap in situ, (B) One month postoperatively.**

## DISCUSSION

Secondary cleft deformities are routine complication in concerned patients following the primary repair which may occur at any age of which development of fistula is the most common reported complication. Depending on repair technique used and age at which primary repair was done, fistula development has a worldwide prevalence of 0-77.8%.<sup>4-7</sup> Secondary fistulas can be located starting from alveolar margin to anterior third, middle third or posterior third of the palate. Probable causes for manifestation of anterior palatal fistulas (APFs) are anatomically shorter lesser segment, wide palatal shelves, improper reflection of subperiosteal flap, and improper closure.<sup>7</sup> Most patients with primary cleft lip deformities are operated up on within the first year of life, however, there is a long period of dramatic growth to

follow, leading to secondary deformities like short and tight lip and deformities in vermilion border, to name a few.<sup>2,3</sup>

Depending on site, size and tissue quality in the concerned area, various surgical techniques can be adopted for correction of such secondary deformities. To name a few, von Langenbeck's bipedicle flap technique, Veau-Wardill-Kilner pushback technique, Bardach's two-flap technique, Furlow double opposing Z-plasty are some of the most commonly used surgical techniques for palatal fistula repair.<sup>2,8</sup> Anterior palate redo with Bardach's principle by Denny and Amm uses local palatal flaps. This technique allows direct visualization of all elements essential for a successful repair.<sup>4</sup> 'Tadpole flap' proposed by Henderson is a crevicular flap used for closure of anterior palatal fistula.<sup>9</sup> Though the flap helps provide with a well vascularised and mobile tissue, the technique is not entirely reliable.<sup>10</sup> Carstens introduced sequential cleft management with sliding sulcus technique and alveolar extension palatoplasty.<sup>11</sup> Jackson utilized a combination of buccal and palatal flaps along with bone grafting.<sup>12</sup> Another technique of repair with secondary mucolized tissue involved creating a vomer flap in small figure of three which showed significantly improved healing outcome. Many techniques for fistula closure involved utilizing tongue flap, free flap, buccal mucosal graft or acellular dermal matrix.<sup>13-16</sup>

Tongue flap is one of the preferred options for correction of secondary palatal fistulas for it can be successfully used in cases like anterior defects that are too large to be managed using mucobuccal flaps, scarred palate or where direct closure of nasal layer is impossible.<sup>7,17</sup> The flap provides with admirable vascularity and ample of tissue which can be oriented or modified to fit most complicated fistulas.<sup>17</sup> This technique has success rate reaching up to 100% with few complications.<sup>18,19</sup> In our case, doing a tongue flap was extremely useful in managing the anterior palatal fistula extending to the alveolus. Abundant flap tissue available in tongue and its mobility helped in its proper adaptation at the anterior most position of the fistula.

Secondary deformities of cleft lip have esthetic as well as functional concerns to the patient. These deformities are so varied in their presentation that choosing an appropriate technique to correct them is challenging. Accurate diagnosis of the deformity and its underlying cause is an essential part of treatment planning. Surgical planning must consider the soft tissue, muscles, underlying skeleton, scar of previous surgery and presence of deficient tissue, if any. Our patient had a deficient vermilion with deformity of the Cupid's bow and a short, wide upper lip. These deformities had to be addressed specifically using a single stage cheiloplasty procedure.

Deficient vermilion may be prevented during initial lip operation by back cutting the mucosa in gingivobuccal

sulcus along medial lip element and advancing it inferiorly. Vermilion deficiency is more common in cases of bilateral cleft lip and is usually more severe. Mild deformity may be treated with V-Y advancements, double or single Z-plasty or mucosal grafts.<sup>20</sup> Severe bilateral cleft lip vermilion deficiencies require Abbé cross lip flap transfer.<sup>21</sup> Cupid's bow recreation is difficult due to either malalignment of the vermilion and white roll or a short lip scar. This can be corrected by Z-plasty or a repeat lip rotation with a focus to reset the proper height for symmetry of Cupid's bow. In our patient the deficient vermilion was combined with an invisible Cupid's bow leading us to use a single stage lip revision procedure.

To be considered as a short lip, the philtral column should be at least 3 mm shorter than the contralateral non-cleft philtrum.<sup>21</sup> Minor cases of short lip may be due to underlying scar contracture that improves over 6-8 months' time. More severe cases are usually caused by inadequate rotation of lip at the time of primary operation. These can be corrected by Z-plasty for as much as 3 mm of advancement. The wide lip deformity is mainly seen in bilateral cleft lip patient caused by designing the new philtrum too wide at time of initial lip repair. To correct this problem the excess philtrum should be excised along previous lip scar. Most severe cases of short and wide lip necessitate a repeat rotation advancement procedure as was done in our patient.

The additional iatrogenic deformity we had to address in our patient was the absence of labial vestibule. This had resulted due to improper primary surgical repair and had led to an immobile upper lip with unaesthetic teeth and gingival visibility. This deformity was corrected by releasing the orbicularis oris muscle from the premaxillary segment and lining the muscle with local mucosal flaps realigned during the cheiloplasty procedure.

## CONCLUSION

Secondary deformities are common in patients of cleft lip and palate. The main aim of treatment in these patients is to boost the normal growth and development and enhance the quality of life with number of surgical interventions. Proper diagnosis, planning and execution of primary surgery are of utmost importance in preventing majority of secondary cleft deformities. Treatment of secondary deformity depends on the degree of deformity and the severity of impact on normal functions and growth.

### *Clinical significance*

Development of secondary cleft deformities is a common complication encountered by cleft patients following primary repair. It is important that at least such cases do not occur due to iatrogenic negligence. Therefore, it is highly recommended that Government institutions, teaching centers with provision of patient care, Non-

Governmental Organizations (NGOs), private clinical setups and healthcare providers provide literature reports of such cases when encountered so that practicing dentists and surgeons can be kept updated on advances in treatment techniques and various expectant outcomes before narrowing down to a customized treatment protocol for a patient. Also, such reporting will help in creating and maintaining database of these cases providing more accurate statistical data and knowing trends of treatment outcomes.

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