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Case Report

Primary repair of binderoid bilateral cleft lip and palate: Synchronous nasolabial repair with premaxilla ostectomy and fixation

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ABSTRACT

Binderoid cleft lip and palate refers to patients who have complete unilateral or bilateral cleft lip and palate with nasolabiomaxillary hypoplasia and orbital hypotelorism without evidence of holoprosencephaly. These patients are characterized by hypoplastic nasal tip, conical columella, tiny prolabium, underdeveloped lateral labial elements, and small/mobile premaxilla. The ipsilateral central and lateral incisors are absent in patients with unilateral cleft, and single-toothed premaxilla is typically found in bilateral patients.

Special considerations and several modifications are needed not only in the operative technique, but also in the entire treatment process, starting from the preoperative period until childhood and adolescence.

A 4-year-old Indonesian girl with binderoid bilateral cleft lip and palate visited our department with the help of a relief agency.

We present the surgical management of the binderoid cleft lip and palate using synchronous nasolabial repair with premaxillary ostectomy and fixation following Mulliken's principle and special considerations for the late primary repair are discussed.

1. Introduction

In 2003, Mulliken et al. suggested the term binderoid (Binder-like) complete cleft lip and palate (CCLP) to be used for the CCLP patients with nasolabiomaxillary hypoplasia and orbital hypotelorism, without evidence of holoprosencephaly [1]. The term 'binderoid' comes from its similarity with Binder syndrome (anomaly), which is characterized by nasolabiomaxillary hypoplasia [1–3]. Mulliken et al. identified 15 Binderoid CCLP patients among the 695 patients with all forms of cleft lip and palate (CLP) [1,2]. Female to male ratio was 2:1 [1,2]. Seven of the 15 infants had unilateral CCLP, with all seven having hypoplastic septum, small alar cartilages, narrow/conical basilar columella, ill-defined Cupid's bow, thin vermilion mucosa on both sides of cleft, and diminutive premaxilla [1,2]. Eight of the 15 infants had bilateral CCLP (BCCLP), with all having more severe hypoplastic nasal tip, conical columella, tiny prolabium, underdeveloped lateral labial elements, and small/mobile premaxilla in addition to above features [1,2]. The ipsilateral central and lateral incisors were absent in all the patients with unilateral cleft, and single-toothed premaxilla was typically found in the bilateral patients [1,2]. In these patients, presurgical dentofacial orthopedics was often aborted because of the diminutive and mobile premaxilla [1,2].

In these infants, several modifications were reported to be necessary during primary nasolabial repair because of the diminutive bony and soft-tissue elements [1,2]. Special considerations and modifications are needed not only in the operative technique, but also in the entire treatment process, starting from the preoperative period until childhood and adolescence [1,2].

Abbreviations: BCCL, Bilateral complete cleft lip; BCCLP, Bilateral complete cleft lip and palate; CLP, Cleft lip and palate; CCLP, Complete cleft lip and palate; GPP, Gingivoperiosteoplasty.

☆ Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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A 4-year-old Indonesian girl with binderoid BCCLP was treated by synchronous nasolabial repair with premaxilla ostectomy and fixation under the principles of Mulliken [1,2,4]. We present the details of the surgical procedures, and discuss the special considerations for this late primary repair.

2. Case report

A 4-year-old Indonesian female was referred to our department with the help of a relief agency. She had BCCLP with marked nasolabiomaxillary hypoplasia and orbital hypotelorism. Holoprosencephaly had already been ruled out and therefore, she was diagnosed as binderoid BCCLP. There was no abnormal finding in her general condition on preoperative evaluation. She presented complete cleft of the secondary palate and protruding single-toothed hypoplastic premaxilla (Fig. 1). She also had thin vermilion in the lateral labial elements, and hypoplastic nasal septum, which are characteristics of the binderoid CLP patients. For synchronous repair of the primary palate and alveolar gingivoperiosteoplasty (GPP), the protruding premaxilla had to be set back. This can be usually achieved by the use of a preoperative orthopedic appliance, such as Latham appliance, in infants up to the age of 6–7 months [2,4–6]. However, it was impossible to use the orthopedic appliance for the 4-year-old patient because the premaxillary-vomerine suture was not pliable enough.

2.1. Premaxilla ostectomy and fixation

Therefore, ostectomy had to be performed on the septal bone in order to gain space for the setback of the protruding premaxilla (Fig. 2). After marking the incision line antero-posteriorly on vomerine/palatal mucosa (Fig. 2A), incision was made and mucoperiosteal flap was elevated. Ostectomy was performed with a fissure bur and bone rongeur to provide adequate amount of setback (Fig. 2B). After the ostectomy, fixation was done with miniplate and screws (Fig. 2D). Mucoperiosteum was sutured with 5–0 Vicryl (Ethicon, Somerville, NJ). After this procedure, the position of the premaxilla was adequately set back to enable synchronous repair of the primary palate and alveolar GPP (Fig. 3).

2.2. Markings and preoperative anthropometry

Markings were made with gentian violet dye, using a sharpened wooden toothpick. The anatomic points were allocated using the standard anthropometric initialisms (Fig. 4)[4]. Anthropometric dimensions were measured and recorded before the surgical procedure (Table 1). Normal values were based on the published data of Farkas et al. [5]. Fast-growing features were planned to be smaller and slow-growing dimensions to be larger than those of the normal age/sex-matched values. However, the median tubercle (Table 1, ls-sto) was an exception. Although median tubercle is a fast-growing feature, it does not show normal growth in patients with repaired bilateral complete cleft lip (BCCL)[2]. Therefore, it must be fashioned to be as full as possible, anticipating insufficient growth. The labial values for Asian children are reported to be the same as those for Caucasian children until the age of 6, and normal nasal dimensions are reported to differ slightly in projection, columellar length, and alar width in this age period [4,6]. Lidocaine with epinephrine was injected into the nose and labial area. After 5 min of conventional waiting time, the critical points to be preserved during repair were tattooed with gentian violet dye (Fig. 4)[7].

2.3. Incisions

Though the prolabium is smaller than that of the usual BCCL patients', the philtral flap was not drawn smaller in either height or width, but included the entire cutaneous dimension of the tiny prolabium (Figs. 4 and 5)[1], as it is known to expand very little with growth [1,4,7]. However, the interalar dimension was intended to be reduced slightly below the age-matched normal value because it would be widened after the repair of common BCCL (Table 1)[1]. A strip of skin on each side of the philtral flap was de-epithelialized to be laid beneath the lateral labial flaps and simulate philtral ridges (Fig. 5).

2.4. Labial dissection

The philtral flap was elevated up to the level of anterior nasal spine. The anterior nasal spine was ambiguous, consistent with the characteristics of binderoid CLP patients. The lateral white-roll-vermilion-mucosal flaps were incised. The lateral labial elements were dissected from the anterior maxilla, extending over the

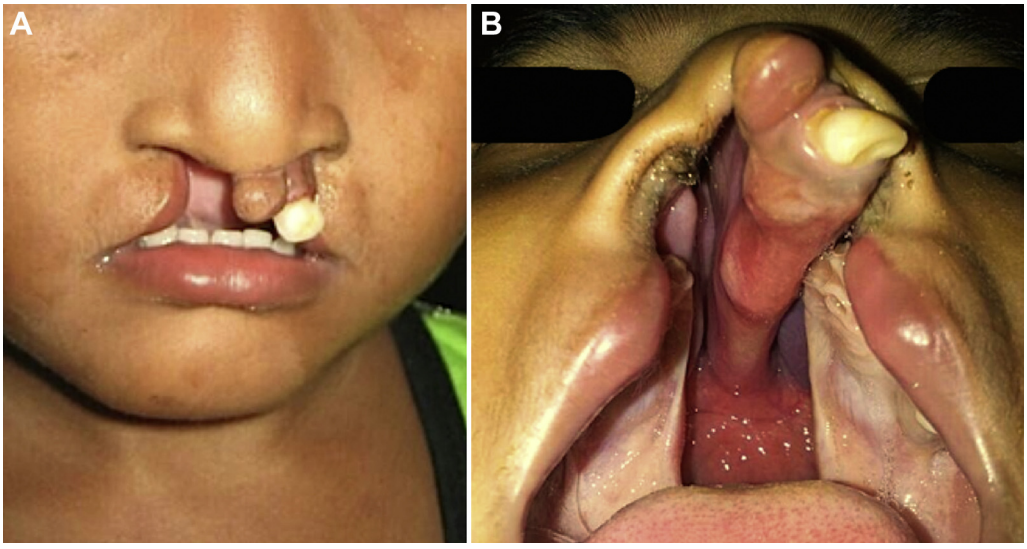

Fig. 1. Preoperative photography of the patient. Note the single toothed tiny premaxilla.