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Clinical Paper
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Secondary mandibular reconstruction for paediatric patients with long-term mandibular continuity defects: a retrospective study of six cases

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Abstract. Paediatric patients with long-term mandibular continuity defects following segmental resection usually present severe functional and cosmetic deformities. Secondary mandibular reconstruction for these patients is very challenging. Literature reports on how to handle these patients are scarce. The aim of this study was to describe the authors' experience in handling those paediatric patients who have not undergone primary reconstruction, for whom the final goal of treatment is to restore a symmetrical facial appearance, masticatory function, and speech. This was a retrospective analysis of the data of six patients who underwent radical mandible resection in childhood, without immediate bone restoration, and who then underwent a secondary mandibular reconstruction procedure after reaching adulthood, during the period 2009 to 2015. The multidisciplinary treatment procedure, selection of the donor site, and reconstructive approach are discussed. Key points in relation to secondary mandibular reconstruction with the aim of achieving not only good functional and cosmetic results, but also an improvement in the paediatric patient's psychological and social outcomes, are emphasized.

Key words: paediatric patient; secondary; mandible reconstruction; multidisciplinary treatment.

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Primary jaw tumours represent about 7% of paediatric malignancies. Nevertheless, paediatric malignancies usually present an aggressive growth pattern and are relatively prone to recurrence.[1,2] Thus, in such cases, an extensive resection of the jaw is necessary. These patients require particular attention, as they rarely undergo simultaneous bony reconstruction due to unpredictable continuing growth, donor site comorbidity, and the possibility of postoperative tumour recurrence.[3] At the authors' institution, the reconstruction protocol is always postponed until after the puberty growth spurt.

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Normal development of the mandible is crucial for the facial complex.[4] The mandible growth is mature at 14–16 years of age in females and at 16–18 years of age in males.[5] The growth of the cranial base, maxilla, and mandible are intimately related. A unilateral mandibulectomy performed at puberty will result in lower jaw deviation and the disruption of sub-periosteal bony apposition and secondary cartilage remodelling. An asymmetric masticatory muscle distribution and jaw deviation might result in abnormal craniofacial development and long-term functional and cosmetic deformities.[2] In the authors' experience, the severity of the facial deformity appears to be related to the timing of the segmental mandibulectomy. A radical segmental mandibulectomy performed pre-puberty results in a more severe facial asymmetry and malocclusion as compared to a post-puberty resection.

Secondary mandibular reconstruction for patients with severe facial asymmetry and malocclusion is very challenging. Re-establishing the bony continuity alone is insufficient, as the malocclusion will remain and proper function is not restored. The gold standard of secondary reconstruction is the restoration of a symmetrical facial appearance, masticatory function, and speech. A multidisciplinary treatment approach is required to resolve this clinical challenge.

The aim of this study was to describe the authors' experience in handling those paediatric patients who have not undergone primary reconstruction after mandibular segmental resection. This retrospective analysis of six cases of secondary mandibular reconstruction was performed to elucidate the importance of the multidisciplinary treatment procedure, selection of the donor site, and reconstructive approach. Key points in relation to secondary mandibular reconstruction with the aim of achieving not only good functional and cosmetic results, but also an improvement in the paediatric patient's psychological and social outcomes, are emphasized.

Materials and methods

This was a retrospective study of six patients (three male and three female) who underwent secondary mandibular reconstruction during the period September 2009 to December 2015, at the Ninth People's Hospital, Shanghai Jiao Tong University School of Medicine. All of these patients had previously undergone ablative treatment for aggressive primary jaw tumours. Patients who had undergone mandibular segmental resection without immediate reconstruction at the pre-puberty age and who underwent secondary mandibular reconstruction that was postponed to adulthood (>18 years old) were included in this study. Some of these patients had received chemotherapy and radiotherapy after the initial surgery.

Clinical and demographic data were recorded, with special attention to facial appearance, status of the occlusion and the temporomandibular joint–glenoid fossa relationship, duration of the mandibular defect (timing between resection and reconstruction), and adjuvant therapy received. Intraoral impressions were taken and the occlusion was registered. Panoramic, lateral cephalometric, and frontal radiograph views were obtained. Computed tomography (CT) scans of both the recipient site (head and neck region) and donor site were obtained for virtual surgery. The cases were discussed in multidisciplinary meetings. Records of the treatment procedures were retrieved; these included orthodontic treatment (patients 1, 2, 4, and 6), orthognathic surgery (patients 1, 2, 3, 4, and 6), reconstructive surgery (patients 1–6), plastic surgery (patient 1), and dental implant procedures (patients 1, 2, and 6). Details of the reconstructive methods and orthognathic surgery used, as well as any surgical complications, were also recorded. The outcomes of the treatment procedures were analyzed according to facial appearance, dental occlusion, and surgical complications (donor site and recipient site).

Results

The case series included three male and three female patients with a mean age of 10.7 years (range 6–13 years; Table 1) at initial presentation. The initial diagnosis was mandibular ameloblastoma for one patient, mandibular hemangioma for one patient, mandibular osteosarcoma for two patients, and mandibular Ewing sarcoma for two patients. All of these patients underwent segmental resection of the mandible without immediate bone reconstruction, except for patient 6, for whom a frozen re-transplanting technique was applied (the re-transplanted bone tissue did not grow normally and fractured 2 months after the initial surgery). Postoperatively, four patients received radiotherapy, one of whom also received chemotherapy. The mandibular defect had lasted approximately 11 years in patient 1, 5 years in patient 2, 9 years in patient 3, 6 years in patient 4, 20 years in patient 5, and 16 years in patient 6. The mean size of the bone defect was 6.9 cm (range 6–9 cm); the condyle was still present in all of these patients.

Two of the patients received an iliac crest bone reconstruction, two received a fibula flap reconstruction, and two received a scapular flap reconstruction. Orthognathic surgery was performed simultaneously in five patients: one patient underwent Le Fort I osteotomy, bilateral sagittal spilt advancement osteotomy (BSSO), and subapical osteotomy, two patients had a subapical osteotomy and sagittal split ramus osteotomy (SSRO) (one of whom also underwent a mandibular angle plasty), and two had a single SSRO for advancement. Four patients (patients 1, 2, 4, and 6) had orthodontic treatment before and after reconstruction surgery. Dental implants were placed postoperatively in three patients (patients 1, 2, and 6).

During routine follow-up, at several months after surgery, all of these patients reported being satisfied with their appearance. They all had a relatively stable and normal occlusion, except for patient 5. This patient had lost most of her teeth as a result of radiotherapy. The average hospital stay was 10 days (range 8–14 days). There were no infections of graft bone tissues and no wound dehiscence at either the donor site or the recipient site, except for patient 5, who suffered from titanium plate exposure after surgery.

Typical case

A 6-year-old boy was diagnosed with ameloblastoma in the right mandible and was treated with marsupialization in September 2003. Recurrence of the ameloblastoma was noted 2 months later, and a segmental resection of the right mandible without immediate bone reconstruction was performed in November 2003; the ramus of the right mandible was preserved. The patient returned to the hospital 11 years later requesting a mandibular reconstruction.

Physical examination revealed a severe facial deformity with facial asymmetry and hyperplasia of the soft tissues of his left face (Fig. 1A and B). Intraoral photographs and dental casts showed a non-stable occlusion with five missing teeth (43–47) and malocclusion, for which a stable occlusion was unachievable; the lower jaw was deviated to the right side. Dental analysis revealed a class II dental malocclusion of the remaining left molars, proclination of the maxillary anterior teeth, overjet and overbite of the anterior teeth, and narrowing and mild crowding of

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