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Systematic Review Paper
Craniofacial Anomalies

Mandibular reconstruction in the growing patient with unilateral craniofacial microsomia: a systematic review

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Abstract. The purpose of this systematic review is to provide an overview of the surgical correction of the mandible in unilateral craniofacial microsomia (UCM) performed in the growing patient, and its long-term outcome and stability. The following databases were searched: PubMed, Embase, Cochrane, and Web of Science. Articles reporting prospective and retrospective studies of patients not older than 16 years ($N \geq 4$) who had undergone surgical correction of a craniofacial microsomia spectrum condition using grafts, osteotomies, distraction, or combinations of these, were reviewed. The period of follow-up was selected to be $\geq$1 year. After inclusion, the articles were evaluated on short- and long-term outcomes, relapse, and any increase in asymmetry following treatment. Thirty of 1611 articles were included in the qualitative synthesis. Analysis of the surgical mandibular correction of UCM showed that the outcome is not so much treatment-dependent, but patient-dependent, i.e. deformity gradation-dependent. The type I–IIa Pruzansky–Kaban patient had the best results with regard to minimal relapse and/or minimal increase in asymmetry. Single-stage correction of the asymmetry should be postponed until the permanent dentition stage. It can be concluded that in the treatment of the severely hypoplastic mandible, the patient will benefit from a multi-stage treatment protocol if indicated for functional or psychological problems.

Keywords: craniofacial microsomia; hemifacial microsomia; goldenhar; craniofacial dysostosis; distraction osteogenesis; graft; mandibular; reconstruction; osteotomy.

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Deformities of the unilateral craniofacial microsomia (UCM) spectrum have proven difficult to treat in the growing patient, due to their heterogeneous presentation. The structures of the first and second branchial arches involved are the maxilla, zygoma, mandible, external and middle ear, facial and trigeminal nerves, muscles of mastication, and overlying soft tissues.[1] A straightforward classification system is essential to improve our knowledge of the deformity. The most commonly used classification system is that provided by Pruzansky[2] and later modified by Kaban et al.,[3] although other systems have been reported.[4,5] The Pruzansky–Kaban system consists of four types. Type I is a small mandible with normal morphology. Type

IIa is a mandibular ramus abnormal in both size and shape; type IIb is a mandibular ramus and temporomandibular joint (TMJ) abnormal in size, morphology, and location. A type III deformity consists of an absent ramus, condyle, and TMJ.

Correction of the asymmetric mandible by mandibular osteotomies and bone grafts has been performed in adults since 1928.[6] The first report of surgery in children with UCM dates from 1941.[2] Up to the 1980s, the overall tendency was for reconstruction of the severely hypoplastic mandible with autogenous grafts, mostly costochondral grafts; however rib, iliac crest, fibula, and temporal bone were also used.[7,8] In the early 1990s, McCarthy et al. were the first to report successful lengthening by gradual distraction of the mandible.[9] This changed the approach to craniofacial correction in a revolutionary way.[10] However, two decades after the first report, it has become clear that distraction osteogenesis (DO) is not the ideal solution for every patient. Extensive work by Nagy et al. showed a lack of statistical evidence to support the use of DO before the permanent dentition stage as a single treatment modality.[11] There have been increasing numbers of reports on multistage correction of the facial deformity with the use of both DO and grafts.[12–15]

The purpose of this systematic review is to provide an overview of the surgical correction of the mandible in UCM in the growing patient, and its long-term outcome and stability.

Methods

The PRISMA statement[16] was used as a guideline.

Search strategy

The following databases were searched: PubMed (until 18 October 2012), Embase (until 18 October 2012), Cochrane (until 18 October 2012), and Web of Science (until 18 October 2012).

We focused on search terms for the congenital deformity of interest and the surgical intervention. Both free text words and medical subject heading (MeSH) terms were used. The heading sequence (hemifacial microsom*[tw] OR hemi facial microsom*[tw] OR hemifacial hypoplas*[tw] OR Hemi facial hypoplas*[tw] OR craniofacial microsom*[tw] OR facial asymmetr*[tw] OR face asymmetr*[tw] OR asymmetric fac*[tw] OR (goldenhar*[tw] OR otomandibular dysostosis[tw] OR oculoauricul*[tw] OR facioauricul*[tw] OR facio-auricul*[tw] OR oculo-auricul*[tw] OR OAV[tw] OR FAV[tw] OR ((lateral[tw] OR unilateral[tw] OR hemilateral[tw]) AND (facial[tw] OR craniofacial[tw])) OR (branchial[tw] AND (arch[tw] OR arches[tw])) AND (syndrom*[tw] OR defect*[tw] OR anomal*[tw] OR dysplas*))) AND (distract*[tw] OR osteodistract*[tw] OR osteotomy[mesh] OR osteotom*[tw] OR bone transplant*[tw] OR bone graft*[tw] OR bone autograft*[tw] OR bone allograft*[tw] OR osseous flap*[tw]) NOT (animals[mesh] NOT humans[mesh]) AND eng[la] was selected. Reference lists of included studies were hand-searched for additional studies of interest.

Inclusion criteria

Articles reporting prospective and retrospective studies of children and adolescents not older than 16 years of age ($N \geq 4$) who had undergone surgical correction of a craniofacial microsomia spectrum condition by grafts, osteotomies, DO, or a combination of these interventions, were included. The period of follow-up was selected to be $\geq$1 year.

Data extraction and analysis

Duplicates were removed. Two authors reviewed the literature individually (BIP and CJJMC). Reports on heterogeneous patient groups and/or study groups with fewer than four patients were excluded due to possible bias and an expected low level of evidence.[17] Articles that met the inclusion criteria or for which the abstract was lacking information, were obtained as full-text articles. Next, full-text articles were reviewed in accordance with the inclusion criteria. Only the data of patients with objective information were included in the quantitative assessment. If there were multiple publications by the same author (group), the author was contacted and the studies were combined.

Articles were graded on quality of evidence using the Oxford Centre for Evidence-Based Medicine (OCEBM) criteria.[18] Data on the number of patients, classification, type of surgery, type of implanted material, average age during the intervention, average length of follow-up, number of relapses, increases in asymmetry, and the number of complications, where available, were tabulated.

Results

The initial search identified 2471 papers. Thirty articles met the inclusion criteria[3,14,15,19–45] (Fig. 1; Table 1). To prevent outcome bias, a total of 17 articles had to be combined.[3,19,21,22,25,27,30–35,38,40–43] An overlap in patient data was observed in six different combined article study groups. For example, Meazzini et al. have published three studies–in 2005, 2008, and 2012.[19,21,25] The patients in these articles were likely to be the same

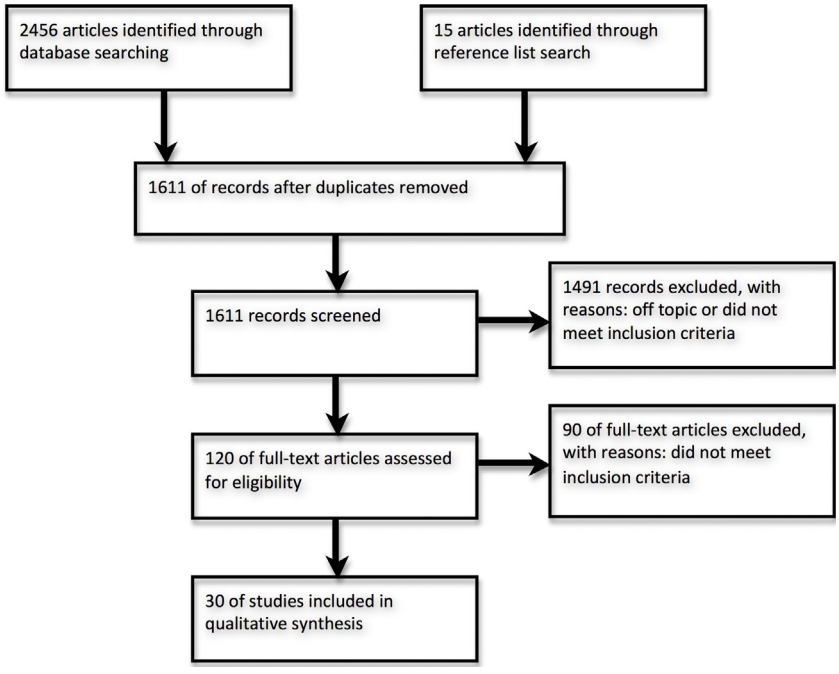

Fig. 1. Data extraction flowchart, according to the PRISMA statement.[16]