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Systematic Review Paper

Complications of mandibular distraction osteogenesis for congenital deformities: a systematic review of the literature and proposal of a new classification for complications

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Abstract. A systematic review of English and non-English language articles on the complications of mandibular distraction osteogenesis (MDO) for patients with congenital deformities was performed, in accordance with the PRISMA statement. Search terms expressing distraction osteogenesis were used in 'AND' combination with search terms comprising 'mandible' and terms for complication, failure, and morbidity. A search using PubMed (National Library of Medicine, NCBI), EMBASE, and the Cochrane Central Register of Controlled Trials yielded 644 articles published between 1966 and mid October 2013. Clinical articles that reported complications related to MDO were included. Finally 81 articles on MDO in congenital deformities were eligible and were screened in detail. Complications including minor infection (6.0%), device-related problems (7.3%), skeletal open bite (2.4%), hypertrophic scar formation (2.1%), facial nerve palsy (1.8%), neurosensory disturbances of the inferior alveolar nerve (1.9%), and (fibrous) non-union (0.7%) were seen. A new index for more detailed classification of complications in MDO is proposed based on six categories that indicate the impact of the complication and its further treatment or final results. The proposed complication index may be a useful tool to classify complications related to MDO.

Keywords: distraction osteogenesis; lengthening; complication; complicated; congenital; syndromic; failure; morbidity; mandible; mandibular; systematic review; PRISMA statement.

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Craniomaxillofacial distraction osteogenesis (DO) is a well-described surgical reconstructive technique that was first reported in the peer-reviewed literature by McCarthy et al.; the technique was applied in the treatment of the hypoplastic mandible in four patients.[1] Since then, several systematic reviews on the clinical application of craniomaxillofacial DO[2–4] and DO in infancy[5] have been published. Master et al.[6] published an article on complications in mandibular DO (MDO). Nevertheless, evidence-based reports on the long-term results, relapse, and complications of MDO are limited.

Paley introduced a classification in which complications arising in the orthopaedic application of DO are divided into problems, obstacles, and complications.[7] This classification was adopted by Neyt et al. for transpalatal DO.[8] Mofid et al.[3] grouped the complications of craniofacial DO into five major categories: technical failure of the distraction process, injury to a vital structure, failure to guide the distraction process along the appropriate vector, infection, and 'other'. Shetye et al. reported a stratification system for MDO in which incidents related to hardware or hard and soft tissue were subdivided into minor, moderate, and major.[9] In 2010, Davidson et al. developed a similar classification for complications in MDO.[10] However, the authors think there is a need for a standard classification that is more detailed with regard to the relevant clinical situation and possible further treatment, and is more widely applicable for use by clinicians.

The aims of this study were (1) to perform a systematic review of the literature on complications of MDO for congenital deformities, and (2) to introduce an index for the classification of complications in (mandibular) DO in general.

Materials and methods

Literature search

A comprehensive systematic review of the literature was performed in the bibliographic databases PubMed (National Library of Medicine, NCBI), EMBASE, and the Cochrane Central Register of Controlled Trials from inception to 15 October 2013; the review was performed in accordance with the PRISMA statement.[11] Search terms included controlled terms from medical subject headings (MeSH) in PubMed and Emtree in EMBASE, as well as free text terms. We used free text terms only in the Cochrane register. Search terms expressing distraction osteogenesis were used in 'AND' combination with search terms comprising 'mandible' and terms for complication, failure, and morbidity (Table 1). The references of the identified articles were searched for additional relevant publications.

Table 1. Primary and secondary key words used for the systematic research.

Primary key words	Secondary key words
Distraction	Mandible
Distraction osteogenesis	Mandibular
Lengthening	Alveolar
Complication	
Complicated	
Failure	
Morbidity	

Study selection and inclusion criteria

Two reviewers independently screened all potentially relevant titles and abstracts for pre-specified eligibility criteria.[11] If necessary, the full text article was checked for the eligibility criteria. Differences in judgement were resolved through a consensus procedure. The full text of articles was then obtained for further review.[11]

The articles were included if they met the following eligibility criteria: (1) clinical article, (2) mandibular distraction osteogenesis (MDO), (3) congenital deformity, and (4) a report on complications. Studies were excluded if data on complications were insufficient, no translation was available, or the publication was a non-clinical article (Table 2).

Articles that were found clinically relevant to the study subject were included in the systematic review. According to their emphasis, these relevant papers were included if they described MDO in the treatment of congenital mandibular deformities. The articles were screened for the following data: type of deformity, number of patients, type of DO, distraction device, vector, and type and number of complications. The latter were classified according to the proposed classification index shown in Fig. 1. This classification emphasizes on the severity and clinical consequences of a complication by dividing events according to spontaneous resolving or permanent complications, hospitalization or general anesthesia required for correction of the complication.

The initial literature search identified a total of 973 references: 521 in PubMed, 437 in EMBASE, and 15 in the Cochrane register. After removing duplicate references ($n = 329$) that were selected from more than one database, 644 papers remained. Titles and abstracts were screened for eligibility by the two reviewers; 335 articles were excluded from the review based on the abstract. The full text was obtained for 309 papers and analyzed thoroughly. Subsequent categorization produced the following clusters (Table 2): (1) 124 articles concerned non-congenital deformities; (2) 57 had insufficient or no information on complications and/or methods; (3) 24 papers were non-clinical (eight scientific, 16 synopsis); (4) four papers were not relevant (three non-DO, one maxilla); (5) 11 papers had no available translation (one Russian, eight Chinese, one Japanese, one Polish); (6) five articles had an edited publication type (three discussion, two letters to the editor/authors); (7) three papers were not available in the international libraries. These seven groups were excluded from further evaluation. In the case of a paper that reported complications in a mixed population (congenital, developmental, or acquired), in which the complications could not be traced back to the exact patient subgroup, the article was excluded on the basis of insufficient data. In total 228 articles were excluded based on the eligibility criteria. Eighty-one articles on MDO for congenital deformities were included. The flowchart of the literature search and selection process through the different phases of the systematic review (PRISMA) is shown in Fig. 2.[11]

Table 2. Inclusion and exclusion criteria.

Condition	Article types	Number of papers (n)
Excluded from the systematic review	Non-congenital deformities	124
	Insufficient or no information on complications and/or methods	57
	Non-clinical articles (experimental, scientific, synopsis)	24
	Non-(mandibular) distraction osteogenesis	4
	No translation available	11
	Publication type, e.g. letter to the editor, discussion	5
	Not available in international libraries	3
Included in the systematic review	Clinical articles on complications in mandibular distraction osteogenesis for congenital deformities	81