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Systematic Review
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Surgical correction of dentofacial deformities in juvenile idiopathic arthritis: a systematic literature review

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Abstract. The aim of this study was to assess current evidence for the surgical correction of dentofacial deformities in patients with temporomandibular joint (TMJ) involvement from juvenile idiopathic arthritis (JIA). A systematic literature review, according to the PRISMA guidelines, was conducted. Meta-analyses, randomized controlled trials, cohort studies, observational studies, and case reports were eligible for inclusion. Exclusion criteria were no JIA diagnosis, no clearly defined outcomes, dual publications (except meta-analyses), non peer-reviewed studies, non English language publications, and animal studies. The outcome measures assessed were TMJ function, skeletal alignment, and morbidity. The database search identified 255 citations, of which 28 met the eligibility criteria. Of these, 24 were case reports or case series with a low level of evidence that did not allow for meta-analysis. Extrapolated evidence supports orthognathic surgery in skeletally mature patients with controlled or quiescent JIA and a stable dentofacial deformity. Distraction osteogenesis was recommended for severe deformities. Some authors demonstrated unpredictable postoperative mandibular growth with costochondral grafts. Alloplastic TMJ reconstruction was efficacious, but should be used cautiously in skeletally immature patients. TMJ function and skeletal alignment was improved with reconstruction by any technique and morbidity was low. The surgical correction of arthritis-induced dentofacial deformities is indicated but the level of evidence is low. Prospective multicenter studies are needed.

Key words: juvenile idiopathic arthritis; temporomandibular joint arthritis; juvenile chronic arthritis; orthognathic surgery; Le Fort I; bilateral sagittal split osteotomy; vertical ramus osteotomy; bimaxillary surgery; genioplasty; reconstructive surgery; TMJ prosthesis; costochondral graft; distraction osteogenesis.

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Juvenile idiopathic arthritis (JIA) is the most common chronic rheumatic disease in children, with an annual incidence of 1.3 to 22.6 per 100,000[1–7]. The temporomandibular joint (TMJ) is involved in 11.6–80% of cases[8–11]. TMJ arthritis can lead to reduced mouth opening, impaired mastication, pain, disruption of condylar growth, and dentofacial deformities[12]. Unilateral TMJ arthritis may cause facial asymmetry and/or an occlusal cant[13]. Bilateral involvement may lead to clockwise rotation of the mandible, resulting in micrognathia, anterior open bite, and a reduction in posterior airway space (PAS)[14,15]. A small PAS is associated with obstructive sleep apnea (OSA) and related comorbidities[16].

TMJ arthritis is difficult to diagnose and control[14]. Uncontrolled arthritis may lead to severe dentofacial deformities. In some patients, growth deviation resulting from TMJ arthritis can be improved with orthopedic/orthodontic treatment during growth[17]. Indications for reconstructive surgery include the following: improvement in TMJ function and/or occlusion, skeletal alignment, desire for facial esthetic improvement, and/or improvement of OSA.

There are two strategies for the surgical correction of JIA-induced dentofacial deformities: (1) TMJ preservation (i.e., orthognathic surgery, distraction osteogenesis (DO)), and (2) TMJ reconstruction (i.e., resection of the remaining condyle, synovial lining, and disc, and reconstruction with an autologous graft[18] or an alloplastic prosthesis[19]). The choice of operational strategy depends on the patient's age, disease activity, extent of the skeletal deformity, and surgeon preference and experience.

The aim of this study was to assess the level of evidence for surgical correction of dentofacial deformities in patients with JIA-related TMJ involvement.

Materials and methods

This study was conducted by the TMJaw Surgical Task Force. TMJaw is an international, multidisciplinary research network founded in Oslo, Norway in 2010, which focuses on research of TMJ arthritis related to JIA. The terminology of the present systematic review adheres to the TMJaw consensus-based standardized terminology[20].

Search strategy

An electronic database search strategy was designed based on the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines[21]. References were archived in a RefWorks database (ProQuest LLC, Ann Arbor, MI, USA). The search strategy was developed for MEDLINE and modified for Embase. The search was limited to these two databases because they are the most inclusive. The primary search was conducted in June 2017 and updated in February 2018. Search terms included the following: juvenile idiopathic arthritis OR juvenile chronic arthritis AND orthognathic surgery OR Le Fort 1 OR bilateral sagittal split OR vertical ramus osteotomy OR bimaxillary surgery OR genioplasty OR distraction osteogenesis OR temporomandibular joint surgery.

Outcome variables

The PICO criteria (patients, intervention, comparison, and outcome) for the study are presented in Table 1. Outcome variables included: (1) TMJ function (i.e., maximum mouth opening measured between the incisal edges of the central incisor teeth, in millimeters), (2) pain, (3) occlusion and skeletal alignment, and (4) operation-related morbidity.

Meta-analyses, randomized controlled trials (RCTs), cohort studies, observational studies, case series, and case reports were eligible for inclusion. Case reports were also included due to the limited number of publications with a high level of evidence identified with the predefined terms. Studies were excluded if they did not include information on JIA diagnosis, had poorly defined outcomes measures, or were dual publications (except meta-analysis), non peer-reviewed studies (e.g., conference abstracts), animal studies, or written in a language other than English.

Study selection and bias

Two authors (PF, SEN) independently assessed the studies identified in the literature search on three occasions. The first selection was based on titles only. The second selection included an assessment of the abstracts. The third and final assessment involved full text review (Fig. 1). Any disagreement between the reviewers was resolved through discussion and the involvement of a third reviewer, until consensus was reached.

The following data were collected from the eligible studies: study design, level of evidence, patient selection criteria, number of subjects, patient sex, mean patient age, surgical procedure, outcomes, adverse effects, and other relevant findings.

The selected studies were assessed for risk of bias on the basis of the following variables: prospective study design, sufficient description of the outcome variable, uniform inclusion criteria, standardized examination protocol, outcome assessor blinded to imaging findings, and information on outcome variable variation. Risk of bias was classified as 'high' if a 'no' was obtained in three or more criteria. The level of evidence was scored according to the Oxford Centre for Evidence-based Medicine (OCEBM) level of evidence guide (http://www.cebm.net/oxford-centre-evidence-based-medicine-levels-evidence-march-2009/), as follows: 1a: systematic review with homogeneity of RCTs; 1b: individual RCT with narrow confidence interval; 1c: all or none case series; 2a: systematic review with homogeneity of cohort studies; 2b: individual cohort study including low quality RCT; e.g., <80% follow-up; 2c: 'outcomes' research and ecological studies; 3a: systematic review (with homogeneity) of case–control studies; 3b: individual case–control study; 4: case series (and poor cohort and case–control studies); 5: expert opinion without explicit critical appraisal, or based on physiology, bench research, or 'first principles'.

Results

Two hundred and fifty-five articles were identified in the first search; 85 remained

Table 1. PICO criteria.

PICO	
Patients	Patients with a JIA diagnosis and involvement of the TMJ
Intervention	Receiving a reconstructive or orthognathic surgical intervention
Comparison	Efficacy and safety of various reconstructive or orthognathic surgical modalities in JIA
Outcome	Assessment of treatment outcomes and safety profile
Databases included: MEDLINE via PubMed and Embase. Search terms were constructed for MEDLINE and modified for Embase[a]	

JIA, juvenile idiopathic arthritis; TMJ, temporomandibular joint.

[a] The MEDLINE search terms: ((juvenile idiopathic arthritis) OR (juvenile chronic arthritis)) AND ((orthognathic surgery) OR (Le Fort I) OR (bilateral sagittal split) OR (vertical ramus osteotomy) OR (bimaxillary surgery) OR (genioplasty)).

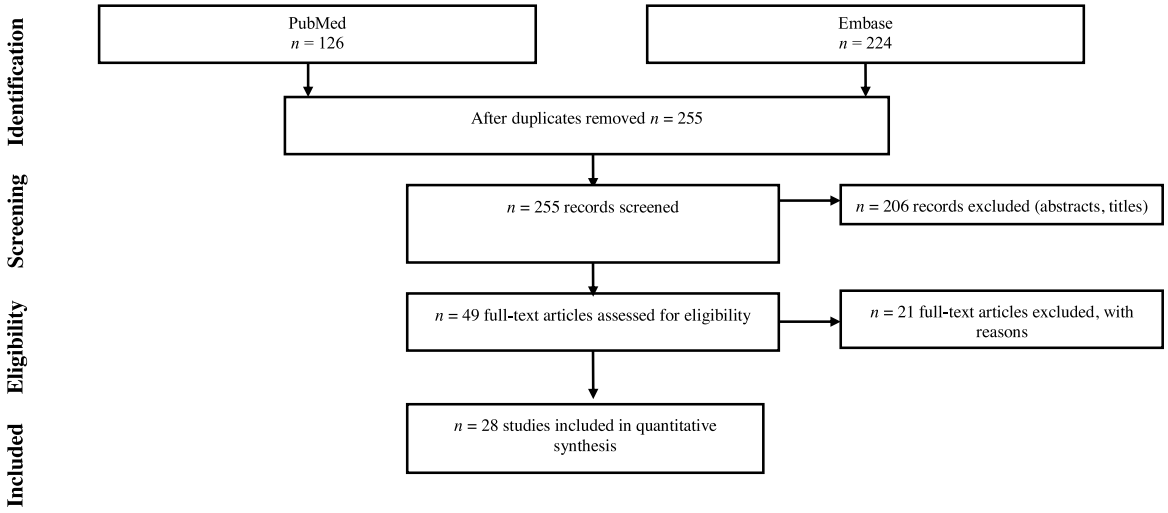

Fig. 1. PRISMA flow diagram of the study selection process.

after title screening. Abstract evaluation resulted in 49 articles. Articles were excluded during full-text assessment for the following reasons: animal studies ($n = 1$), studies not in English ($n = 3$), meta-analysis ($n = 1$), or lack of clearly defined outcome variables ($n = 16$). The final sample included 28 papers for full text evaluation (Fig. 1, Tables 2 and 3)[18,22–48]. The inter-rater agreement for study selection was 100% for all of the variables (Supplementary Material, Table S1). Of the 28 articles included, 24 were case reports or case series[18,22–44], three were retrospective chart reviews[45–47], and one was a prospective cohort study[48]. Many of the included articles described surgical procedures in a mixed group of patients. The included studies described a total of 172 subjects with JIA who underwent a reconstructive surgical procedure.

The risk of bias was rated 'high' in 26 (93%) of the included studies; two (7%) articles had a 'medium' risk of bias30,48 (Supplementary Material, Table S1).

No study included a control group. Four studies did not have uniform inclusion criteria, i.e. not all study subjects had a JIA diagnosis[24,29,30,32]. Various methods were used for the evaluation of outcomes, such as cephalometric analysis and clinical photographs when evaluating skeletal alignment, polysomnography when evaluating OSA, and mouth opening (in millimeters) to evaluate function. The examination protocol was unclear in two studies[26,28]. No outcome assessors in the studies were blinded to imaging findings. Detailed information on outcome variables, such as mean jaw advancement, mean relapse, and mean mouth opening in millimeters, were presented in 23 studies[18,23–25,27,29–32,34,35,37–48]. According to the OCEBM level of evidence, all included articles were grouped in category four (i.e., case series and poor cohort and case–control studies).

Surgical intervention and outcomes

TMJ preservation—orthognathic surgery

Nine case reports and case series discussed orthognathic surgery in 80 patients (43 with JIA)[30–32,34,36,37,42,45,46]. Age at operation ranged from 15 years to 53 years. Follow-up varied from <1 year to 10 years. All studies reported improved function, occlusion, and skeletal alignment. All studies reported few complications; these included infection, wound dehiscence, skeletal relapse, and continued TMJ pain.

Kahnberg and Holmstrom described maxillary and mandibular orthognathic surgery with chin augmentation (implant) in 19 of 34 analyzed patients (37 in total, three with JIA). In 15 patients, a chin implant was the only procedure. All patients reported high postoperative satisfaction[30]. Two patients had postoperative dehiscence and one patient had infection of the implant. Pagnoni et al. described Le Fort I osteotomy and genioplasty in five patients with quiescent JIA[37]. Occlusion and skeletal movements were stable 8 months postoperatively. All patients reported relief of TMJ pain, increased bite strength, and a reduction of headache and sleep respiratory distress postoperatively. These authors advocated for Le Fort I osteotomy with autorotation of the mandible and genioplasty, without a mandibular osteotomy, theorizing that a mandibular operation exacerbates condylar resorption, TMJ pain and dysfunction. According to the same authors, postoperative exacerbation of TMJ arthritis did not occur.

Leshem et al. reported skeletal relapse of 2.1 mm after a mean 9.6 mm mandibular advancement at 1 year postoperatively in eight patients with JIA[45]. They also reported an improvement in quality of life for all patients. These authors recommended a surgical splint with a small posterior open bite and edge-to-edge incisor relationship to compensate for expected relapse. One patient in their series continued to suffer from TMJ pain postoperatively.

Kreiborg et al. described a 15-year old patient with ''some relapse'' 1 year after bimaxillary surgery[34]. They hypothesized that dysplastic mandibular growth leads to occlusal instability and poor masticatory muscle function. Therefore, they recommended early orthognathic surgery (i.e., prior to skeletal maturity) to maintain occlusal stability throughout growth.

One of seven subjects (age 10–24 years) described by Myall et al. required a reoperation at age 18 years due to relapse after sagittal ramus osteotomy and genioplasty at age 10 years[36]. Oye et al. described mean skeletal relapse of 2.3 mm after average mandibular advancement of 5.3 mm in 16 patients[46]. Half of these patients had TMJ pain before surgery (mean visual analog scale (VAS) score 2.4), and 38% reported pain postoperatively (mean VAS score 1.3). These authors therefore concluded that orthognathic surgery reduces pain.

TMJ preservation—distraction osteogenesis

Seven studies described DO in 40 patients with JIA[23,25,33,35,39,47,48]. One study was a prospective cohort study[48] and one was a

Table 2. Summary table of articles on TMJ preservation surgery (i.e., orthognathic surgery and distraction osteogenesis) included in the review.

Source	Study design	LOE[a]	Patient selection criteria	Number of patients	Sex Age (years)[b]	Surgical procedure	Outcome[c]	Adverse effects	Follow-up Other relevant findings
Orthognathic surgery									
Kahnberg and Holmstrom[30]	CS	4	Consec.	37 (3 JIA)	20 F, 17 M 25 (16–43)	Chin augmentation: acrylic implant combined with orthognathic surgery	Stable position of chin, high patient satisfaction	2 dehiscences, 1 infection	FU: <1 year for 3 pts, min 1 year for 34 pts; 5 years for 17 pts
Kasfikis et al.[31]	CR	4	Consec.	1 (mandibular hypoplasia, OSA, Crohn's disease)	F 18	BSSO + genioplasty	Stable function and esthetics at 2 years postop.	No	FU: 2 years
Kennett and Curran[32]	CS	4	Selected	4 (1 JIA)	3 F, 1 M 16 (12–18)	JIA pt: VRO with iliac BG, onlay BG to the chin, 6 weeks of MMF; other pts: BSSO, condylectomy + rib graft, sliding genioplasty	Improved occlusion and profile	Resorption of the onlay BG to the chin	FU JIA pt: 1 year (3–18 months) The need for postop. jaw exercises is stressed
Kreiborg et al.[34]	CR	4	Consec.	1 (9–17 years, baseline to FU)	F 15	LFI + BSSO + genioplasty	Facial growth/facial appearance and oral function improvement	Some relapse 1 year postop., but it did not affect the soft tissue profile	FU: 1 year postop.
Leshem et al.[45]	Retrosp. CS	4	Consec.	8	5 F, 3 M 18 (17–22)	BSSO (8), LFI (6/8), genioplasty (4/8)	Improved occlusion, facial esthetics, QOL; continued TMJ pain (1 pt) Mean MA 9.6 mm: Co–Gn; mean mandible relapse 2.1 mm (did not significantly affect the clinical outcome)	Some relapse 1 year postop., but it did not affect the soft tissue profile	FU: min 8 months (mean 36 months)
Myall et al.[36]	CS	4	Consec.	7	5 F, 2 M Age at onset of JIA 1–11 Age at surgery 10–24	All: BSSO and genioplasty; 2 pts LFI; MMF 6 weeks and wire/screws	6 patients with satisfactory function and esthetic results; all 7 had normal sensitivity and TMJ function	1 pt underwent reoperation (1 st surgery at age 10, 2nd at age 18)	FU: 6 months No patients had ankylosis Not the authors experience that children with JRA are predisposed to this condition Prefer to delay surgery until growth has finished
Oye et al.[46]	Retrosp. CS (1991–2000)	4	Consec.[d]	16	12 F, 4 M 24.25 (16–53)	BSSO + genioplasty	Improved profile, all patients satisfied; may reduce TMJ pain Mean relapse 2.3 mm of MA 5.3 mm (48%) (Pog–Co); 50% of pts (8) relapsed ≥2 mm	Safe treatment; reoperation for 4 pts, infection in 2 pts; negative tooth sensitivity in 3 pts, scar in 3 pts; plates removed from 3 pts; extraoral changes in 5 pts	FU: 1–10 years

Table 2 (*Continued*)

Source	Study design	LOE[a]	Patient selection criteria	Number of patients	Sex Age (years)[b]	Surgical procedure	Outcome[c]	Adverse effects	Follow-up Other relevant findings
Pagnoni et al.[37]	CS	4	Consec.	5	4 F 21.75 (17–29)	LFI + genioplasty	Improved occlusion and facial profile Mean MA rotation 5.6 mm Mean posterior–anterior face height ratio 63.9 (S–Go/N–Me)	None	FU: min 8 months (8 months–8 years) ''Mandibular procedures may evoke further condylar resorption with pain and functional impairment of the TMJ''
Turpin and West[42]	CR	4	Consec.	1	F 9	BSSO + alloplastic chin implant	Improved facial esthetics and occlusion	Good function Some relapse; chin implant migration, some increased overjet	FU: 30 months postop., 14 months after removal of orthodontic appliances
Distraction osteogenesis									
Cattaneo et al.[23]	CR	4	Consec.	1 (unilateral hypoplasia of right mandibular ramus, JIA)	M 12.5	Unilateral MDO	Evaluation of load transfer mechanism to the TMJs before, under, and after DO	No long-term FU	Before DO: high peak stresses to the affected TMJ After DO: more symmetrical loading to the TMJs Under DO: the reaction forces in the TMJs were low
de Zee et al.[25]	CR			simulation model CBCT/MRI	4	Consec.	1 pt with JIA	M 12.5	Unilateral MDO
Shape of articular eminence (load, remodeling fossa): reduced load after DO	No	FU: 6.5 years							
Mackool et al.[35]	CR	4	Selected	1	F 26	Bilateral extraoral ramus DO, 24 mm, consolidation 85 days	Improved esthetics, sleep apnea; MIO: 10 mm to 12 mm	Scar tissue in skin	FU: not stated Cephalometric measures reported
Kofod et al.[33]	CR	4	Selected	1 (asymmetry secondary to JIA)	M Young	Unilateral vertical ramus DO 16 mm	Skeletal deformity corrected Stress load is higher on distracted side during distraction	NR	No FU data
Nørholt et al.[48]	Prosp. cohort study	4/3	Consec.	23	9 M, 14 F Age at onset 6.4 $\pm$ 3.9 Age at surgery 15.8 $\pm$ 4.7	Unilateral ramus DO 18 mm At removal of DO device: 7 LFI, 1 genioplasty Later surgery: 2 LFI and 1 genioplasty	No aggravating symptoms from TMJ, predictable correction of asymmetry, good function	Minor problem with pain and activation of the device, 2 pts mild dysesthesia, 2 pts trismus	FU: min 12 months

Singer et al.[39]	CR	4	Consec.	1	F 18	BMDO + LFI + genioplasty	Improved esthetics, overjet (remained positive),chin, occlusal plane	Stable results 1 year postop., MIO 32 mm, unchanged postop.; no TMJ pain or muscle pain postop.	FU: 1 year
Stoor et al.[47]	Retrosp.	4	Consec.	12	9 F, 3 M 27.4 (at 1 st surgery)	MA (11) and additional DO (3), or additional AJR, unilateral/bilateral (3); bilateral AJR (1)	TMJ function and healing good, stable occlusion, improved facial appearance	Mean MA 10.1 mm and mean relapse 2.1 mm (pogonion)	FU: mean 2.3 years

AJR, alloplastic joint reconstruction; BG, bone graft; BMDO, bilateral mandibular distraction osteogenesis; BSSO, bilateral sagittal split osteotomy; CBCT, cone beam computed tomography; Consec., consecutive; CR, case report; CS, case series; DO, distraction osteogenesis; F, female; FU, follow-up; JIA, juvenile idiopathic arthritis; JRA, juvenile rheumatoid arthritis; LFI, Le Fort I; LOE, level of evidence; M, male; MA, mandibular advancement; MDO, mandibular distraction osteogenesis; MIO, maximum incisal opening; MMF, maxillomandibular fixation; MRI, magnetic resonance imaging; NR, not reported; OSA, obstructive sleep apnea; postop., postoperative; Prosp., prospective; pts, patients; QOL, quality of life; Retrosp., retrospective; TMJ, temporomandibular joint; VRO, vertical ramus osteotomy.

[a] Level of evidence was scored according to the Oxford Centre for Evidence-based Medicine level of evidence guide (http://www.cebm.net/oxford-centre-evidence-based-medicine-levels-evidence-march-2009/).

[b] Presented as the mean (range) for case series studies; presented as the mean ± standard deviation for the prospective cohort study.

[c] S–Go, sella–gonion; N–Me, nasion–menton; Pog–Co, pogonion–condylion; Co–Gn, condylion–gnathion.

[d] Patients referred from Rheumatology Department, Rikshospitalet.

retrospective study[47]. Five of the studies were case reports[23,25,33,35,39]. Three of the included studies evaluated load transfer to the TMJs before, during, and after DO[23,25,33].

Nørholt et al. prospectively followed a cohort of 23 patients with JIA after unilateral DO, with or without a maxillary osteotomy or genioplasty, for at least 12 months postoperatively[48]. They found predictable correction of asymmetry, improved function, and decreased TMJ pain after DO. Two patients developed dysesthesia in the lower lip. A significant improvement in condylar translation was observed during distraction, but postoperatively this was not significantly improved when compared to the preoperative conditions.

In the retrospective study of Stoor et al., 11 patients with JIA and severe TMJ degeneration had mandibular advancements with orthognathic surgery, of whom three had additional mandibular DO and three had a unilateral or bilateral TMJ prosthetic reconstruction[47]. An additional patient underwent only bilateral TMJ prosthetic reconstruction. The mean mandibular advancement was 10.1 mm. Mean follow-up was 2.3 years. There was a mean relapse at pogonion of 2.1 mm. The occlusion was stable in 11/12 patients. The TMJ function was good and the facial esthetics improved in all patients.

Singer et al. reported an 18-year-old female who had bilateral mandibular DO, Le Fort I osteotomy, and a genioplasty[39]. At the 1-year follow-up, she had improved skeletal relationships and good function. Preoperative bilateral masseter pain resolved postoperatively.

Mackool et al. described a 26-year-old woman with JIA who had relief of OSA and an improved facial profile after bilateral mandibular DO[35]. However, MIO only improved from 10 mm to 12 mm.

Cattaneo et al. and Kofod et al. presented the case of a 12-year-old boy with JIA, unilateral mandibular hypoplasia, and condylar erosion[23,33]. Using a simulation model, the authors showed high peak stresses to the affected resorbed condyles before operation with DO. During active distraction, forces in the TMJs were low. After DO, load sharing was more symmetric to the condyles compared to the preoperative measurement. de Zee et al. also found reduced load to the affected TMJ 6 years after DO[25].

TMJ reconstruction—autologous TMJ reconstruction

Seven case reports and case series ($n = 48$ patients, 29 with JIA) discussed TMJ reconstruction using costochondral grafts

Table 3. Summary table of articles on TMJ reconstruction surgery (i.e. costochondral grafts and alloplastic joint reconstruction) included in the review.

Source	Study design	LOE[a]	Patient selection criteria	Number of patients	Sex Age (years)[b]	Surgical procedure	Outcome[c]	Adverse effects	Follow-up Other relevant findings
Costochondral grafts									
Bowler[22]	CS	4	Consec.	2	2 F 14	Bilateral CCG + dermis graft	Excellent functional and esthetic results, self-confidence improved	No	FU: 4 years (1 case)
Cohen et al.[24]	CS (OSA)	4	Consec.	20 OSA (1 JIA)	M 16 (6 days–18 years, total sample)	Skeletal expansion/ osteotomies: orthognathic surgery, CCG, DO, TMJ arthroplasty, segmental Le Fort I osteotomy and soft tissue surgery	Improved respiration, increased volume of naso-oropharynx	No	FU: 6–12 months ''Tracheostomy leads to social isolation for these children''
Felix et al.[27]	CR and review	4	Consec.	1 bilateral ankylosis	F 13	Bilateral TMJ reconstruction with CCG	MIO 40 mm = good function; good esthetics	No	FU: 24 months
Guyuron[28]	CR	4	Referred	1	F 35	Ankylosis age 10 LFI osteotomy with anterior rotation, BSSO 8 mm advancement, genioplasty 7 mm, bilateral CCG; after 6 months, lipectomy and removal of submaxillary glands	Relapse 1–2 mm first year, stable following 5 years; psychological improvement; TMJ function good, 40 mm MIO	No	FU: 5 years Diagnosis 9 years, prednisolone
Stringer et al.[18]	CS	4	Consec.	5	5 F 14–18	Orthognathic surgery + CCG (inverted L-osteotomy + LFI + iliac graft + genioplasty) = 1 surgery	Gn–N–Ba, 4 of 5 pts class I occlusion on FU, 1 pt a 3 mm open bite postop. Mean increase anterior–posterior 22.7 mm T1–T2	Relapse average 1.5 mm T2–T3 Infection ($n = 1$)	FU: mean 9.6 years (range 4–14 years)
Svensson and Adell[41]	CS, Retrosp.	4	Selected	12 (=7 pts from Svensson 1993)	11 F, 1 M 10–17	CCG 9 bilateral, 3 unilateral; disc removed in 12 joints, preserved in 9 joints	Good function: 11/12 had no pain or symptoms from TMJ; MIO 35.6 mm preop., 41.3 mm at FU; 5 pts class I, 7 pts class III at FU; 7 pts further orthognathic surgery	Overgrowth of CCG (8/12)	Mean FU: 5.3 years (age 18.9 years) Considerable risk of unilateral overgrowth of CCG ($n = 8$)
Svensson et al.[40]	CS	4	Selected	7	All F Mean age at onset 4.5 Mean age at surgery 12.5 (range 10–14)	5 bilateral, 2 unilateral, resection of condyle (disc preserved) and coronoid process; CCG from C5 or C6; fixation with screws; 6 weeks of MMF Functional treatment after 8 weeks	5/7 no functional problems or pain; MIO after 2–4.5 years: 39 mm (range 30–43 mm); good symmetry in bilateral cases; slight asymmetry in unilateral cases	No complications to CCG	FU: 2–4.5 years Extensive mandible growth from 8 weeks postop. to latest FU No report of excessive overgrowth or resorption of CCG – longer FU needed
Alloplastic joint reconstruction									
Fanaras et al.[26]	CR	4	Consec.	1	F 42	Bilateral AJR, custom-made (Biomet)	Full range of hinge movement, good function	No	FU: MIO 31 mm 6 months later

Haq et al.[29]	CS	4	Selected	5 (1 JIA)	F 43 (16-year history of trismus/ ankylosis)	Bilateral custom-made AJR	MIO increased to 25 mm	No	FU: 18 months
Paul et al.[38]	CR	4	Selected	1	M 29	Absence of both condyles; 1 st surgery AJR, 2nd surgery BSSO with BG and 10-mm plate	Improved sleep and esthetics; AHI decreased from 48 to 7	Not reported	FU: 10 months No fossa component!
Webster et al.[43]	CR	4	Consec.	1	F 18 (= time at surgery)	Bilateral custom-made AJR (Biomet) + mandibular advancement and genioplasty	Improved facial and dental esthetics	None	FU: 3 years
Wolford et al.[44]	CS	4	Consec.	56	55 F 1 M 39 (15–61)	Bilateral TMJ AJR Techmedica	Outcome groups based on clinical assessment: one or no previous TMJ surgery: good 86%, fair 14%, poor 0%	5 AJR removed; 17 pts (30%) reoperated due to heterotopic bone formation, fibrosis, calcification, inflammation	FU: 30 months (range 16–46 months)

AHI, apnea–hypopnea index; AJR, alloplastic joint reconstruction; BG, bone graft; BSSO, bilateral sagittal split osteotomy; CCG, costochondral graft; Consec., consecutive; CR, case report; CS, case series; DO, distraction osteogenesis; F, female; FU, follow-up; JIA, juvenile idiopathic arthritis; LFI, Le Fort I; LOE, level of evidence; M, male; MIO, maximum incisal opening; MMF, maxillomandibular fixation; OSA, obstructive sleep apnea; postop., postoperative; preop., preoperative; Retrosp., retrospective; TMJ, temporomandibular joint.

[a] Level of evidence was scored according to the Oxford Centre for Evidence-based Medicine level of evidence guide (http://www.cebm.net/oxford-centre-evidence-based-medicine-levels-evidence-march-2009/).

[b] Presented as the mean (range) for case series studies.

[c] Gn–N–Ba, gnathion–nasion–basion.

(CCG) in both skeletally mature and immature subjects[18,22,24,27,28,40,41]. Age at operation varied from 10 years to 35 years. Follow-up ranged from 6 months to 14 years. Following CCG, six studies reported improved functional outcomes (defined as increased mouth opening) and improved skeletal profiles[18,22,27,28,40,41]. Two studies reported improved self-confidence[22,28] and two case reports found improvement in OSA[24,28]. One paper suggested synovectomy in conjunction with CCG to prevent progression of TMJ destruction[22]. Morbidity at both the donor and recipient sites was low. All seven case reports and case series discussing autologous TMJ reconstruction reported no or few TMJ symptoms or muscle symptoms after surgery. Two studies discussed the risk of overgrowth of the CCG, resorption of the graft, and donor site morbidity[40,41]. Svensson et al. followed 12 patients with JIA for a mean of 5.3 years after CCG and demonstrated asymmetric mandibular overgrowth in eight[41]. Resorption of the graft was seen when bone screws were removed 1–2 years after CCG reconstruction.

Guyuron reported skeletal relapse of 1–2 mm at 1 year after CCG reconstruction and BSSO advancement of the mandible and genioplasty, but no further change over the subsequent 5 years[28]. Stringer et al. reported a skeletal relapse of 1.5 mm at a mean of 9.6 years following CCG and concomitant Le Fort I osteotomy[18].

TMJ reconstruction—alloplastic TMJ reconstruction (AJR)

There were five case reports and case series on reconstruction with alloplastic TMJ prostheses in patients with JIA[26,29,38,43,44]. In total, these studies included 64 subjects, 60 of whom had a diagnosis of JIA. Age at the time of operation ranged from 15 years to 61 years. Follow-up ranged from 6 months to 46 months. TMJ replacement was either by stock joint ($n = 1$)[38] or custom joint prosthesis by Biomet or TMJ Concepts ($n = 4$)[26,29,43,44]. All reported improved functional and skeletal outcomes. One manuscript reported improved OSA[38]. All five case reports and series discussing alloplastic TMJ reconstruction reported low morbidity with no or few TMJ symptoms or muscle symptoms after surgery.

Webster et al. recommended AJR as the most suitable treatment for patients with JIA, condylar degeneration, and dentofacial deformity[43]. They performed AJR

with a simultaneous mandibular advancement and genioplasty due to the extent of condylar destruction and clockwise mandibular rotation.

Wolford et al. reported good treatment results after AJR in patients who had undergone ''no or one previous TMJ surgery'' before joint replacement, based on clinical assessment of stability, function, and residual pain[44]. Paul et al. reported improved OSA and esthetics at 10 months of follow-up, after ''dual stage surgery'' using first an alloplastic TMJ prosthesis followed later by mandibular advancement with a body osteotomy fixed with a titanium reconstruction plate with an inter-positioning iliac graft bilaterally[38].

Discussion

The aim of this study was to assess the level of evidence for surgical correction of dentofacial deformities in patients with TMJ degeneration from JIA. Currently, there is no consensus regarding the most appropriate reconstructive approach for this patient population. The choice of operation may be influenced by the degree of skeletal maturity, level of TMJ arthritis activity, extent of TMJ and dentofacial deformity, and surgeon preference and experience.

The findings of this systematic review suggest that all available reconstructive surgical interventions (orthognathic surgery, DO, CCG, AJR) improve TMJ function and skeletal relationships in this patient population. Only minor to moderate complications have been reported, such as temporary nerve damage, infection, and mandibular overgrowth. However, these reports must be evaluated in the context of the low level of evidence.

The management of TMJ dysfunction and pain is important for activities of daily living such as mastication, speech, and oral hygiene. Improvements in mouth opening and micrognathia are critical for airway management under general anesthesia[50]. Also, improvements in occlusion, skeletal alignment, facial esthetics, and self-confidence are considered important for patient well-being, since facial attractiveness has been shown to influence education, relationships, and employment[51].

All of the included case reports and case series on orthognathic surgery in JIA reported improved functional outcomes, occlusion, and skeletal alignment[30–32,34,36,37,42,45,46]. Orthognathic surgery is traditionally performed after skeletal maturity to maximize stability. A major concern in this context are the high-angle cases, i.e. cases with a steep mandibular plane angle, undergoing a large mandibular advancement, with skeletal relapse secondary to soft tissue forces[52]. Children with JIA often are high-angle cases due to TMJ arthritis with a TMJ deformity causing posterior rotation of the mandible resulting in an anterior open bite. Mandibular advancement has been reported to show relapse even in non-JIA patients[14,53]. Thus, some authors recommend early surgical intervention in order to provide better conditions for normal development of the facial skeleton[34,53].

Other investigators recommend anti-inflammatory medications and stabilizing splints until the TMJs are 'stable' before orthognathic surgery, with minimal postoperative loading of the TMJs[17,54]. The rationale for this approach is to avoid or minimize the mandibular advancement in order to avoid overloading the TMJs. However, these treatment approaches may also result in significantly compromised function and esthetics, continued restriction of the oropharyngeal airway, and continued or worsening of pain.

Morbidity was low for orthognathic surgery compared to TMJ reconstruction (CCG or alloplastic prosthesis) or DO. Surgical exposure when performing CCG, AJR, or DO with an extraoral approach is usually via a combined preauricular and submandibular incision. Therefore, there is a risk of facial nerve damage during surgery[55].

In this review, improvements in functional outcome and skeletal alignment[39,47,48], OSA[35], and load transfer to the TMJs[23,25,33] were reported after DO. For patients with significant dentofacial deformities, DO may be a suitable option. DO is widely used to improve the morphology of the facial skeleton in patients with congenital or acquired deformities[56]. DO may be more stable over time with less skeletal and dental relapse compared to conventional orthognathic surgery[48,57]. However, DO requires careful vector planning and patient collaboration during device activation. In asymmetric cases, JIA patients will have a period with potential overloading of the TMJ on the affected side. Therefore, it is argued that DO performed during adolescence reduces the time period of load to the affected TMJ, limits the risk of nerve damage to one side compared to both when performing the BSSO osteotomy, and reduces the need for a maxillary osteotomy by allowing normal growth of the maxilla concomitantly with the mandible[48]. However, a second operation is required to remove the distraction device.

In skeletally immature patients, autologous reconstruction may be preferred because the graft has the potential to grow with the child. This review of available articles found an improvement in functional outcome and esthetics[18,22,27,28,40,41], self-confidence[22,28], and OSA[24,28], after CCG. However, there is controversy in the literature regarding the use of CCG, primarily due to the potential for overgrowth[40,41]. Additionally, the CCG surgical procedure carries the risk of graft resorption and ankylosis, and is associated with donor site morbidity[55,58]. Nevertheless, other authors found no difference in outcomes of TMJ reconstruction with CCG or alloplastic graft in rheumatoid arthritis patients, except that the alloplastic graft group had fewer additional operations[55].

Some authors favored AJR in adult patients, advocating that this approach provides stable long-term results and facilitates early mobilization. However, long-term outcomes of AJR beyond 15 years are unknown[19]. Patients who have not reached skeletal maturity are expected to have a long life span, and thus there is a risk of mechanical failure requiring reoperation or replacement of the AJR. The included articles reported improved OSA and esthetics[38] and good functional outcomes[26,29,38,43,44], with the longest follow-up of 46 months[44].

Some authors suggested staged surgical procedures with AJR first, followed by BSSO with bone graft and plate osteosynthesis[38]. Others recommended a single AJR procedure[43]. The authors claimed that failure is likely to result when performing mandibular advancement surgery alone without TMJ surgery in patients with active TMJ arthritis disease, because of mandibular relapse from continued condylar resorption and/or increased pain levels. In cases of disease remission, maxillary surgery can be performed for closure of open bites; however, the esthetic results are often suboptimal[59,60].

This systematic review had some limitations. First, the studies included had a low level of evidence and a high risk of bias. Most of the studies had a small patient sample, and no study had a control group. Additionally, the mixture of JIA and non-JIA patients in many publications introduces confusion for the reader. Only some of the included patients had JIA (232 patients, 172 with JIA). Thus, it is difficult to ascertain that the results are specific to JIA and not to another condition. Also, the majority of the outcome assessors were not blinded in the included studies. Furthermore, only two databases were used for the literature search and this may have

been a bias at the review level. Therefore, the study heterogeneity did not allow for meta-analysis. The strength of this review is that it includes all manuscripts on JIA and dentofacial reconstruction.

The research and treatment of JIA-induced TMJ and dentofacial deformities requires multidisciplinary collaboration between rheumatologists, orthodontists, and oral and maxillofacial surgeons, and many other healthcare providers (e.g., radiologists, general dentists, physical therapists, etc.). There is a significant need for prospective studies with standardized variables in patients with JIA and dentofacial growth deviations.

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Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at https://doi.org/10.1016/j.ijom.2019.01.007.

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