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Review

Long term complications of costochondral graft reconstruction in temporomandibular joint ankylosis of the young- a systematic review

Kavita R Wadde, Shambhavi Nadkarni*, Paul Mathai

Department of Oral and Maxillofacial Surgery, Government Dental College and Hospital, P D'Mello Road, Near CSMT, Fort, Mumbai, Maharashtra, India

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ABSTRACT

Costochondral graft has been a popular reconstruction choice in the past for temporomandibular joint ankylosis in young individuals. However, accounts of growth hampering complications have also been observed. Our systematic review aims to compile all existing evidence to determine the occurrence of these unfavourable clinical outcomes as well as factors affecting them to provide a better judgement on further use of these grafts. A systematic review was conducted following PRISMA guidelines where databases like PubMed, Web of science and Google Scholar were searched for the purpose of data extraction. Observational studies performed on patients younger than 18 years of age with a minimum follow-up of one year were selected. Incidence of long term complications like reankylosis, abnormal graft growth, facial asymmetry and others were considered as outcome variables. Eight articles with a total of 95 patients were selected where complications like reankylosis (6.32%), graft overgrowth (13.70%), insufficient graft growth (22.11%), no graft growth (3.20%) and facial asymmetry (20%) were reported. Other complications like mandibular deviation (3.20%), retrognathia (1.05%) and prognathic mandible (3.20%) were also observed. Our review concludes that the occurrence of these complications was noteworthy. Thus use of costochondral graft for reconstruction in temporomandibular ankylosis in young patients holds significant risk in development of growth abnormalities. However, modifications in surgical procedure such as use of appropriate graft cartilage thickness and the presence and type of interpositional material can favourably affect the frequency and type of growth abnormality.

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1. Introduction

Temporomandibular Joint (TMJ) ankylosis is an incapacitating condition of the craniomandibular articulation where the osseous or fibrous fusion of its anatomic elements restrains the normal jaw movements. The most common cause for TMJ ankylosis is trauma, followed by infection, congenital anomalies, previous TMJ surgery, iatrogenic causes, etc [1]. In case of traumatic TMJ ankylosis, the hematoma formed in the region of trauma undergoes fibro-osseous changes to form an ankylotic mass that grows aggressively over time [2]. In children, where mandibular growth is still incomplete, this mass not only restricts movement of the joint but hampers the overall functional, topographical, and aesthetic development of the mandible. This is responsible for facial deformities like asymmetry or retrognathia and in some cases disorders such as Obstructive sleep apnoea (OSA) [3]. Thus appropriate treatment of children with ankylosis is of paramount importance owing to the active growth of the mandible and its condyle.

Treatment protocol usually involves removal of ankylotic mass followed by reconstruction of the joint to achieve normal jaw function. There are various options for reconstruction such as using autogenous or alloplastic grafts, distraction osteogenesis, ramus osteotomies, etc [4]. In young patients, reconstruction of the joint should not only fulfil the anatomic and functional role of the joint but also aid in its adaptive growth and remodelling to match the growth of its healthy counterpart on the other side and also to maintain facial symmetry. Out of these various alternatives, an autogenous costochondral graft (CCG) remains a popular choice for reconstructing TMJ in paediatric patients owing to its autogenous nature, adaptability and growth close to that of mandibular condyle. However, costochondral graft also shows a major disadvantage of unpredictable growth at the reconstruction site which leads to abnormal development of mandible. The reporting of such long-term growth abnormalities has been sporadically observed in past and current literature which has thus formed a controversy on whether costochondral graft can be deemed as a reliable material for TMJ reconstruction.

This systematic review aims to compile and review the long-term complications of using costochondral graft for reconstruction of TMJ in children with ankylosis based on existing literature and also to

* Corresponding author.

E-mail address: shambhavin30@gmail.com (S. Nadkarni).

estimate the effect of demographics and intraoperative factors that may influence the long term prognosis of the graft.

2. Material and methods

This systematic review adhered to the PRISMA Statement guidelines (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) and is registered under the PROSPERO (International Prospective Register of Systematic Reviews) database with registration ID CRD42021229412. The following was the review question: 'What are the chances of unfavourable clinical outcomes of costochondral graft placement in young patients suffering from temporomandibular joint ankylosis on long term follow up?'

2.1. Eligibility criteria

The following inclusion criteria was generated based on PICOS framework: (P) Patients below 18 years of age suffering from unilateral or bilateral TMJ ankylosis, (I) Removal of ankylotic mass and reconstruction with CCG, (C) Not applicable, (O) Incidence of unfavourable post-operative outcomes on long term follow up of minimum one year and (S) Observational studies including clinical prospective or retrospective study designs in English language. Studies unrelated to current research topic or having study population above 18 years of age were excluded. Studies including interventions other than costochondral graft reconstruction or those having a follow up period of less than a year were also excluded. Also, articles with insufficient published data, personal communications, case series, case reports, animal studies and those only available as abstracts were not selected for the review.

2.2. Search strategy

A thorough electronic search was conducted from dates of inception till February 2021 using the following databases- PubMed, Web of science and Google Scholar with specific keywords or MeSH terms. The following terms were used:

"Temporomandibular Joint Disorders/surgery"[Mesh], "Ankylosis/surgery"[Mesh]

AND costochondral graft, "Child"[Mesh] and "Postoperative Complications"[Mesh].

Hand search was conducted for references obtained from selected articles and previous reviews. The electronic search and the PICO strategy are shown in Supplementary Table S1.

2.3. Data extraction

All procured articles were screened for eligibility by two independent reviewers and any disagreements were resolved by a third person. The articles were first screened based on their title and abstract out of which only relevant studies were selected. Full texts of these selected studies were again reviewed based on the above mentioned selection criteria. The following information was obtained: year of study, author, type of study, number of patients and joints, age, gender, unilateral/bilateral ankylosis, right/left ankylosis, aetiology, treatment protocol, rib used for autograft, thickness of cartilaginous segment, follow up period & long term complications. The procured data was tabulated on Microsoft Excel sheet (version 2016).

2.4. Assessment of risk of bias

Two review authors undertook the risk of bias assessment independently by using the ROBINS-I tool with RevMan 5.4.1 software. Seven specific domains were addressed: confounding bias, selection bias, bias in classification of interventions, deviations from intended interventions, missing data, measurement of outcomes and selection

of the reported result. The articles were thus categorized as "Low risk", "Moderate risk", "Serious risk" and "Critical risk".

3. Results

3.1. Systematic search

A thorough search on the databases resulted in retrieval of 438 articles. Mendeley software was used to remove duplicates after which 53 results were retrieved. On screening by title and abstract, 4 articles were excluded on the bases of inadequate relevancy to study topic, animal studies and pilot studies. Full texts of 49 studies were assessed based on inclusion criteria out of which 41 articles were rejected. Thus, a total of 8 studies were included in this systematic review. The process of selection of articles is recorded in the form of a PRISMA flowchart (Fig. 1).

3.1.1. Study characteristics

95 study subjects in 2 prospective studies, 5 retrospective studies and 1 ambispective study comprised our review, out of which 20 subjects were of prospective studies and 65 subjects were retrospectively availed. Mean age of the patients at the time of reconstruction was 8.13 years with the range of 3 to 16 years. Slight female predilection with male:female ratio of 0.9 was seen and the mean follow up period was 3.27 years with the range of 1 to 9 years. 79 patients were diagnosed with unilateral ankylosis out of which 20 were right sided and 16 were left sided and the remaining 43 were unspecified [1-3, 5-9]. Bilateral ankylosis was seen in 16 patients [1-3, 5, 6, 8]. Thus there were more than 111 joints included in this systematic review (Table 1).

3.2. Risk of bias assessment

Based on the quality assessment tool used, four studies had a low risk of bias while rest four studies had a moderate risk. Risk of bias for each study is represented in Figs. 2 and 3.

3.3. Complications

All the eight studies reported with some or other form of long term complication in their patients. Five studies reported reankylosis of joint in a total of six patients (6.32%) [1, 3, 5-7]. 37 out of 95 patients (38.94%) exhibited abnormal growth of costochondral graft at the recipient site on long term follow up which was reported by six studies [1, 2, 5, 7-9]. Abnormal growth of graft was determined by three characteristics: overgrowth of graft, insufficient growth of graft or no growth of graft. Out of the 37 patients, 13 patients had overgrown graft (13.70%) [1, 2, 8, 9] and 21 patients had insufficiently grown graft (22.11%) [1, 5, 7, 9] at the operated site whereas three patients presented with absolutely no growth of the graft (3.20%) [9]. 19 patients presented with facial asymmetry with or without chin deviation post treatment (20%) which was reported by five studies [2, 5, 7-9]. Apart from the above mentioned variables, other complications such as jaw abnormalities and deviation were also reported. Behnia H et al [5] in 1997, reported 3 cases with mandibular deviation (3.20%) and one case of retrognathia (1.05%). Ko EW et al [8] in 1999 reported three cases of mandibular prognathism (3.20%) (Table 2).

3.4. Secondary variables

3.4.1. Cause of ankylosis

Total five studies reported the primary cause of ankylosis in patients out of which most common cause was trauma followed by infection [2, 3, 5, 6, 8]. A total 39 patients were reported to had experienced trauma prior to ankylosis out of which three were specified to be due to trauma at birth, three were specified as road traffic

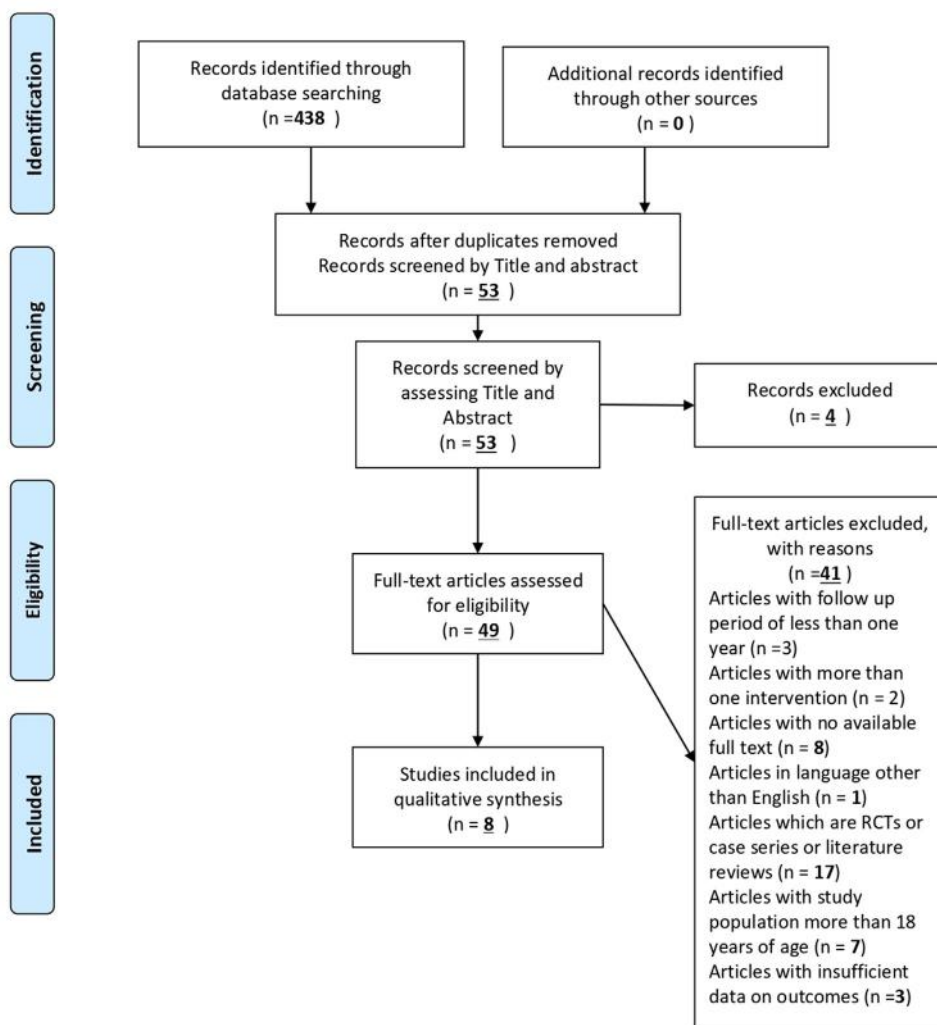


Fig. 1. PRISMA flow chart.

accidents and four were specified as fall. Four patients were reported with the primary cause of infection out of which one was specified as osteomyelitis and three were specified as ear infections. The rest 52 patients had an unknown cause. (Table 1)

3.4.2. Type of surgical procedure

All studies followed the same basic surgical procedure of arthroplasty with costochondral graft reconstruction (Table 3). However, differences were observed in the interpositional material used. Three

Table 1 Demographic data of patients in selected studies.

Sr. no.	Year of study	Author	Type of study	No. of Patient	No. of joints	Age range of patients at the time of reconstruction	Males: Females	Unilateral ankylosis	Right:Left ankylosis	Bilateral ankylosis	Cause of ankylosis
1	1997	Behnia H	Retrospective study	13	16	5 to 14 yrs [mean-8.07 yrs]	05:08	10	03:07	3	1 osteomyelitis, 11 trauma, 1 unknown
2	1999	Ko EW	Prospective study	10	12	3.8 to 12.5 yrs [mean-7.4 yrs]	02:08	8	06:02	2	6 trauma, 1 ear infection, 3 unknown
3	2014	Sharma H	Prospective study	10	12	≠/ < 14 yrs	06:04	8	Not specified	2	8 trauma, 2 infection
4	2016	Bhardwaj Y	Retrospective study	7	10	6 to 13 yrs	02:05	4	02:02	3	3 RTA, 4 Fall
5	2017	Zhao J	Retrospective study	7	7	3 to 7 yrs [mean-4.7 yrs]	03:04	7	Not specified	0	Not specified
6	2017	Balaji SM	Retrospective study	14	14	3 to 9 yrs [mean-5.2 yrs]	10:04	14	09:05	0	Not specified
7	2018	Xia L	Retrospective study	11	13	5 to 11 yrs [mean-6.5 yrs]	06:05	9	Not specified	2	7 Trauma, 4 unknown
8	2020	Lakshmanan S	Ambispective: 13 retrospective, 10 prospective	23	27	3 to 16 yrs [mean-10yrs]	12:11	19	Not specified	4	Not specified
Total				95	111	[3 to 16 yrs] mean- 8.13 yrs	Ratio-0.9	79	20:16	16	

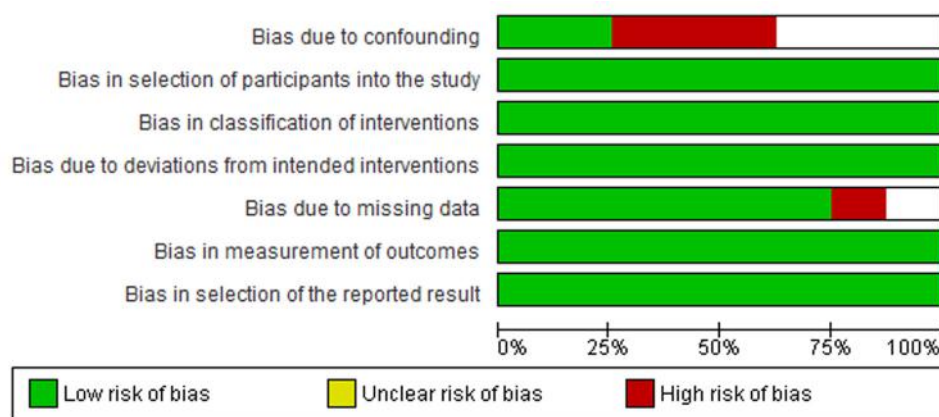


Fig. 2. Risk of bias across included studies.

studies used temporalis myofascial flap [3,5,7], two studies used temporalis muscle [2,6] and one study used buccal fat pad [1] as an interpositional material while two studies have used no interpositional material [8,9]. The relation of interpositional material with the graft growth abnormalities in the selected studies has been depicted in detail in Fig. 4. Five out of eight studies mentioned use of ipsilateral and/or contralateral coronoidectomy to help in increase of mouth opening [2,5-7,9].

3.4.3. Type of rib graft

Except for one study [1], all other studies specified the rib graft used for reconstruction. The fifth, sixth and seventh ribs were mainly used for reconstruction out of which sixth was most common.

3.5. Thickness of cartilaginous segment

All eight studies mentioned the exact thickness of the cartilaginous segment of the selected graft. Few studies maintained a minimal thickness ranging from 1 to 4 mm [1,2,7,9], while others chose a moderately thick cartilage ranging from 4 to 10 mm [3,5,6]. Ko EW et al [8] in 1999 were the only ones to use a cartilaginous segment of more than 10 mm in their study. The relation of cartilage thickness with the graft growth abnormalities in the selected studies has been depicted in detail in Fig. 5.

3.6. Post-operative protocol

Three out of eight studies gave post-operative Maxillomandibular fixation while the other four opted for early mobilization [1,5,8]. All the studies except one provided post-operative physiotherapy in their patients [1-3,5,6,8,9]. Behnia H et al [5] in 1997 was the only study to asked patients to use a removable functional appliance post surgery. (Table 3).

4. Discussion

Treating TMJ ankylosis in children requires not just anatomic but also functional replacement of the joint. The graft used for reconstructing TMJ in children should have active growth potential which aids in normal and symmetric growth of the mandible as the child grows. For this purpose, a variety of autogenous grafts are available such as sternoclavicular joint, metatarsophalangeal joint, costochondral graft, iliac bone graft, fibular head, etc [10]. Costochondral graft has been considered as a gold standard in treating TMJ defects as it fulfils the joint reconstruction goals in the younger population.

Gilles in 1920 had first used CCG on his patients which paved the way for its extensive use in TMJ reconstruction surgery [11]. Over the years CCG has been routinely used in various children with TMJ

ankylosis owing to its capability of vascularization and osseointegration in adjacent bone and low incidence of morbidity at the donor site. In addition, it has primary and secondary growth centres which lie between the cartilaginous and bony part of the graft and grows at a rate similar to that of mandibular condyle [4,10] and allow optimum development of mandible in growing individuals. However, it has been documented in the literature that costochondral grafts may exhibit abnormal growth at the recipient site and may even show reankylosis of the joint [8]. This may lead to facial asymmetry, restricted jaw movements, and abnormal development of the mandible. Other reported complications are poor bone flexibility and elasticity which may lead to fracture and chances of infection [9]. Past literature on CCG have only mentioned these complications but no definite compilation of such complications and their incidence have been reported yet. This systematic review tries to compile all the available evidence of such abnormal behaviour and long term complications of costochondral grafts after its use in young patients with TMJ ankylosis.

Of all the recorded complications, the three most common were reankylosis of TMJ, abnormal growth of graft and facial asymmetry which may or may not involve chin deviation. Abnormal growth of graft showed three different patterns- overgrowth, insufficient growth and no growth at all. In our review, we found that insufficient growth of graft had the highest incidence of 22.11% followed by overgrowth of graft which was 13.70%. Only 3.20% patients showed complete absence of graft growth. Overgrowth of graft can be accounted to the rapid growth rate of the rib during puberty. It has been reported that by the age of 10 the rib grows up to 78.6% of its adult size [9]. Past literature has also mentioned that the thickness of the grafted cartilaginous segment plays a significant role in abnormal growth of the graft. The old school of thought was to use a sufficiently thick cartilaginous segment to avoid bony reankylosis [8]. In the study conducted by Kaban et al [12], it is mentioned that a 3 to 4 mm of cartilage is enough to avoid ankylosis as well as prevent overgrowth. On the other hand, Villaneuva et al [13] mentioned that the age of the patient should govern the thickness of the cartilage. They specified that children in the age range of maximum growth need a thicker cartilage than those who have already crossed puberty. Zhao J et al [7] advocated the use of 5 mm or less than 5 mm cartilage as cartilage more than 10 mm would cause overgrowth while those with 2 to 4 mm thickness may lead to absorption. Among the studies selected for this review, Ko EW et al [8] were the only ones to use a cartilage which was as thick as 15 mm. Incidentally, this study also reported the highest number of patients with graft overgrowth. Conversely, Lakshmanan S et al [1] used a cartilage thickness of 2 to 4 mm and reported the maximum number of patients with insufficient graft growth. According to our observations, the studies which chose a cartilage thickness ranging from 4 to 10 mm were the ones

	Bias due to confounding	Bias in selection of participants into the study	Bias in classification of interventions	Bias due to deviations from intended interventions	Bias due to missing data	Bias in measurement of outcomes	Bias in selection of the reported result
Balaji 2017		+	+	+	+	+	+
Behnia 1997	-	+	+	+	+	+	+
Bhardwaj 2016	+	+	+	+	-	+	+
Ko EW 1999	-	+	+	+	+	+	+
Lakshman 2020	+	+	+	+	+	+	+
Sharma 2014		+	+	+	+	+	+
Xia 2018	-	+	+	+		+	+
Zhang 2017		+	+	+	+	+	+

Fig. 3. Risk of bias item for each included study.

who had the least cases of abnormal graft growth. Incidentally, there were dissimilarities observed in the surgical protocol used among the studies. Out of these the main variable was the use of interposition material. Studies which used temporalis myofascial flap or temporalis muscle as an interpositional material were observed to have reported with fewer complications [2,3,5-7] as compared to the ones who used none [8,9]. The number of surgical steps or phases involved

in the treatment itself can also affect the post-operative results of the graft. Ankylosis cases that are treated in two surgical phases, where the first surgery is performed to remove the ankylosed segment and a second surgery is planned for insertion of the costochondral graft on a later date, have lesser chances of reankylosis than those treated in a single surgical procedure. [14] However, in our systematic review, all the selected studies performed a single stage surgery

Table 2
Complications in selected studies.

Sr. no.	Year of study	Author	Surgical procedure used	Thickness of cartilage of graft	Rib used for autograft	Post-operative Maxillomandib-ular fixation	Post-operative Physiotherapy
1	1997	Behnia H	IGA with CCG reconstruction, IPM- Temporalis Myofascial flap, ipsilateral & contralateral coronoidectomy if needed	5-8 mm	5th, 6th or 7th	3-14 days in few patients not specified number	Functional appliance therapy in nine patients
2	1999	Ko EW	Arthroplasty with CCG reconstruction	15 mm	5th, 6th or 7th	3 to 6 weeks	given immediate post-operatively
3	2014	Sharma H	IGA with CCG reconstruction, IPM- temporalis muscle, coronoidectomy if needed	4-5 mm	5th or 6th	Not given	given immediate post-operatively
4	2016	Bhardwaj Y	IGA with CCG reconstruction, IPM- temporalis muscle, ipsilateral & contralateral coronoidectomy if needed	1-2 mm	6th	Not given, early mobilization done	given immediate post-operatively
5	2017	Zhao J	IGA with CCG reconstruction, IPM- temporalis myofascial flap, ipsilateral & contralateral coronoidectomy if needed	<5 mm	7th	Not given	Not given
6	2017	Balaji SM	IGA with CCG reconstruction, ipsilateral & contralateral coronoidectomy if needed	2-5 mm	6th	Not given	given immediate post-operatively
7	2018	Xia L	IGA with CCG reconstruction, IPM- temporalis myofascial flap	5-10 mm	7th	Not given	Given 1 week post-operatively
8	2020	Lakshma-nan S	IGA with CCG reconstruction, IPM- Buccal fat pad	2-4 mm	Not specified	7-10 days	given post Maxillomandibular fixation

IGA- interpositional gap arthroplasty, CCG- costochondral graft, IPM-interpositional material

where resection of ankylotic mass and insertion of graft was done in the same surgery. This is reflected in our review where reankylosis of TMJ was reported in five out of eight studies with an incidence of 6.32%. Five studies also reported facial asymmetry in their patients with a significant incidence of 20.0%. Other complications like mandibular retrognathia or prognathia and deviation of mandible were reported in minority. Studies of Lakshamanan et al [1], Ko EW et al [8] and Balaji et al's [9] showed the maximum number of complications while Xia et al [3] and Zhao et al [7] had the least complications. The former and latter studies mainly differed in the cartilage thickness and the type of interposition material used.

Our systematic review has few limitations as it includes only observational studies with a high confounding bias as the cases are not differentiated based on classification of ankylosis. When we compared patients suffering from unilateral ankylosis with those having bilateral ankylosis, we found no significant difference in complications as majority of the studies failed to specify the results in accordance to the type of ankylosis and the remaining showed more or less equivalent results in both. This review also has a few strengths as majority of the studies had a long term follow up with mean of 3.27 years and the longest being nine years and had a low to moderate risk of bias. Costochondral grafts are a valuable option for joint

Table 3
Operative interventions in selected studies.

Sr. no.	Year of study	Author	Follow up period	Reankylosis	Graft Overgrowth	Insufficient growth	No growth	Facial Asymmetry including chin deviation	Other long term complications	Total number of patients with complications
1	1997	Behnia H	2 to 9 years [mean-4.9 years]	1	0	4	0	2	1 Retrognathia, 3 mandibular deviation	4
2	1999	Ko EW	1.5 to 8.4 years [mean-4.54 years]	0	7	0	0	3	3 Prognathism	7
3	2014	Sharma H	1 year	2	0	0	0	0	0	2
4	2016	Bhardwaj Y	2 to 8 years	0	1	0	0	1	0	1
5	2017	Zhao J	1.6 to 6.6 years [mean-3.8 years]	1	0	1	0	1	0	2
6	2017	Balaji SM	2 to 6 years [mean-3 years]	0	4	5	3	12	0	12
7	2018	Xia L	1 to 5 years [mean-2.4 years]	1	0	0	0	0	0	1
8	2020	Lakshmanan S	1-4 years [mean-1.5 years]	1	1	11	0	0	0	13
Total			3.27 years [mean-1-9 years]	6	13	21	3	19	7	42

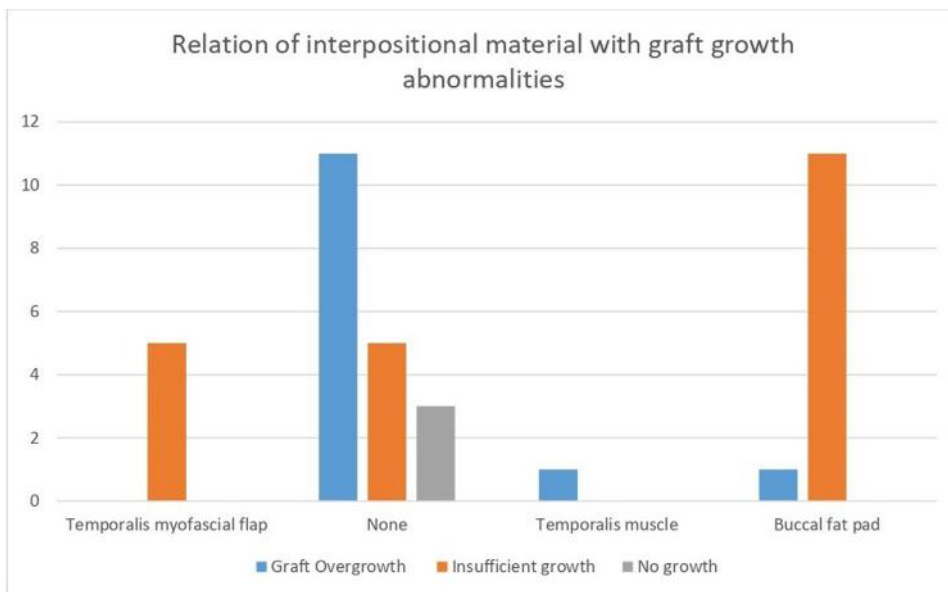


Fig. 4. Relation of interpositional material with graft growth abnormalities.

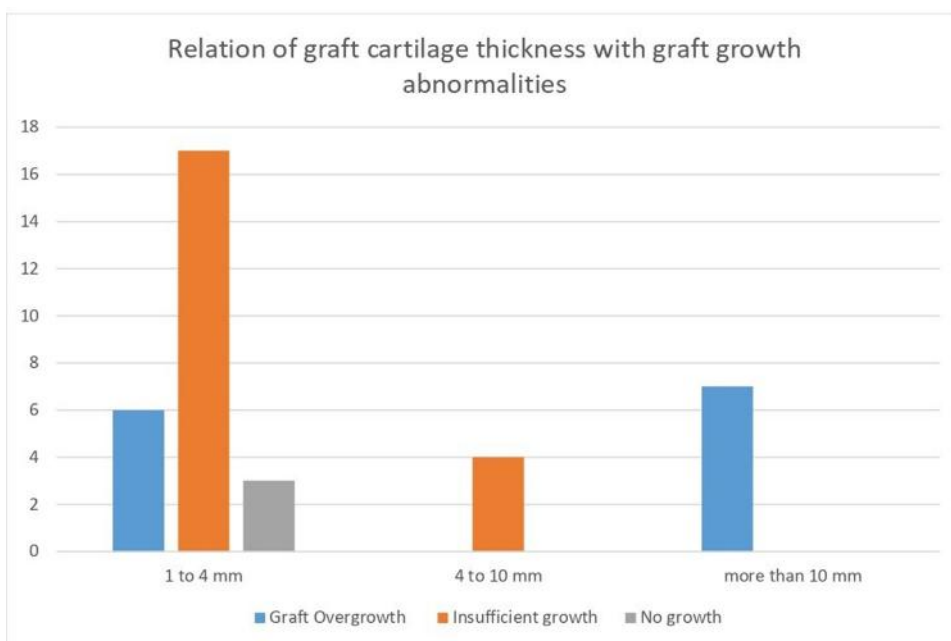


Fig. 5. Relation of graft cartilage thickness with graft growth abnormalities.

reconstruction in young patients with TMJ ankylosis but its post-operative complications make it a controversial choice.

Our systematic review tried to accumulate all relevant literature to derive the risk or chance of encountering such complications. All the eight studies showed some or the other form of complication in their patients. However, the frequency of complications in each of these studies were observed to be diverse. Based on our review we conclude that the use of costochondral graft holds a significant risk of long term growth abnormalities in temporomandibular joint ankylosis of the young. Also, factors such as graft cartilage thickness and use of interpositional material could be correlated with the occurrence of these complications. Changes in surgical procedure can be made in an attempt to reduce the complications. Management of the graft with additional orthognathic or orthopaedic procedures such as simultaneous distraction osteogenesis may further reduce the graft

uptake complications. Further detailed studies focusing on the measures that can be used for prevention and reduction of these complications could be of great help.

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This is a systematic review. The Institutional Research Ethics Committee has confirmed that no ethical approval is required.

Consent to participate

Not applicable.

Consent to publish

Not applicable.

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Declaration of Competing Interest

The authors have declared that no competing interests exist.

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Supplementary materials

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