Idiopathic condylar resorption: A systematic review of etiology and management

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Objective. The aim of this study was to evaluate the level of evidence in the literature on etiology and management of idiopathic condylar resorption (ICR) of the mandible.

Study Design. A systematic search of articles published from 1982 to 2019 was conducted via PubMed, according to the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guidelines. English language reports of human studies that addressed the etiology or management of ICR were included. Case reports, opinion or perspective articles, and nonhuman or non−English language articles were excluded. The quality of the included studies was assessed by using the Oxford Center for Evidence-Based Medicine criteria.

Results. The search yielded 230 studies, and 40 met the criteria for inclusion. The etiology of ICR was the focus of 33 studies; the remaining 8 assessed its management. The total number of patients in the included studies was 1399. The level of evidence assessing the etiology of ICR averaged 3.16 (range 2b−to 3b). Proposed etiologies and contributing factors included female gender, 17β-estradiol levels, condylar anatomy, and history of orthognathic surgery. The level of evidence of studies assessing the management of ICR was 3.7 (range 2b−4). Treatment strategies included disk repositioning, autologous condylar reconstruction (costochondral graft), and alloplastic total joint reconstruction. Average follow-up was 46.8 ± 38.2 months.

Conclusions. The proposed etiology and management of ICR, as reported in the literature, vary considerably. Condylectomy with autogenous or alloplastic reconstruction appears to be the most stable management strategy. The levels of evidence regarding the etiology and management of ICR are low. (Oral Surg Oral Med Oral Pathol Oral Radiol 2020;130:632−639)

Idiopathic condylar resorption (ICR), a pathologic entity of unknown etiology, results in destruction of the mandibular condyles. As it overwhelmingly affects females in the second and third decades of life, it was termed "cheerleaders' syndrome" by Wolford et al.[1] ICR is characterized by progressive, bilateral, symmetric volume loss of the mandibular condyles and rami, resulting in diminished posterior facial height, steepening of the mandibular plane angle, loss of chin projection, and anterior open bite.

ICR remains a diagnosis of exclusion. Other causes of joint destruction, such as juvenile idiopathic arthritis and osteoarthritis, must be ruled out. Demographic, hormonal, anatomic and treatment-related etiologies have been proposed. Given the observed preponderance in young females, an abnormal level of 17β-estradiol has been theorized to be a factor in the development of this disease.[2] A similar pattern of condylar resorption has been reported after orthognathic surgery,[3,4] with proposed initiators including limited preoperative condylar volume, excessive force applied to the condyle intraoperatively, and significant downward repositioning of the pogonion. Despite almost 6 decades since the first description of this disorder,[5] its etiology and pathogenesis remain unclear.

Management of ICR depends on the activity of the disease and the severity of condylar resorption. With inactivity and enough condylar mass to provide a reliable centric relationship, routine orthognathic surgery may be considered. If the disease is active and/or the extent of condylar destruction precludes a reproducible centric-relation position, then condylar/ramus reconstruction with autogenous or alloplastic material is required. There are no standard guidelines for treatment, and reports of outcomes vary considerably in the literature.

The purpose of this study was to assess the proposed etiology and management strategies for ICR and to determine any consistent factors. The specific aims were to (1) identify studies that propose theories on the etiology of ICR, (2) review the outcomes of management strategies for ICR, and (3) characterize the quality of the studies according to the Oxford Center for Evidence-Based Medicine (CEBM) criteria.

Statement of Clinical Relevance

Idiopathic condylar resorption, as the name implies, remains poorly understood. Reports of both etiology and management vary considerably within the literature. Management typically consists of joint reconstruction with autogenous grafts of alloplasts. The levels of evidence regarding etiology and management are low.

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MATERIALS AND METHODS

Search strategy

This study followed the PRISMA (Preferred Reporting Items for Systematic Review and Meta-Analyses) guidelines for systematic review of existing literature. A literature search was conducted by using PubMed. The search algorithm used was as follows: ((“Temporomandibular Joint Disorders”[Mesh]) AND “Bone Resorption/etiology”[Mesh]) OR (“condylar resorption”[tiab]) OR (“progressive condylar resorption”[tiab]) OR (“idiopathic condylar resorption”[tiab]) OR (“condylar atrophy”[tiab]) OR (“condylysis”[tiab]) OR (“condylar resorption”[tw]) OR (“progressive condylar resorption”[tw]) OR (“idiopathic condylar resorption”[tw]) OR (“condylar atrophy”[tw]) OR (“condylysis”[tw]).

Inclusion and exclusion criteria

English language reports of human studies pertaining to the etiology or management of ICR were included. The following types of reports were excluded: case reports, reviews, opinion or perspective articles, nonhuman studies, non−English language reports, and studies not relevant to the etiology and management of ICR.

Data extraction and analyses

Data extracted from eligible studies included year of publication, number of patients, location of study, proposed risk factors for development of ICR, and management strategies for ICR. The average follow-up after operative intervention was determined from the reports of studies, when available. We reviewed the studies and rated them according to CEBM guidelines for level of evidence.[6] CEBM ranks studies from levels 1 to 5, with 1 being the highest level of evidence.

RESULTS

In total, 230 abstracts were identified on the initial search. Of these, the following were excluded: 34 case reports, 8 commentary or perspective articles, 2 articles pertaining to syndromes, 24 non−English language reports, 69 on topics other than ICR, and 32 reviews. The final sample included 40 studies containing data from 1399 patients.

Etiology

Thirty-three studies proposed etiologies for development of ICR and potential risk factors (Table I).[2−4,7−36] The average quality of the studies describing etiology was 3.16 (range 2b−3b). Among studies on etiology, 9 discussed anatomic risk factors with level 2b (n = 2); level 3b (n = 3); and level 4 (n = 4) evidence. Sixteen studies proposed surgical risk factors with level 2b (n = 6) and level 4 (n = 10) evidence. Two studies proposed systemic factors with level 2b (n = 1) and level 4 (n = 1) evidence. Six studies proposed “other” (defined as multiple factors) with level 2b (n = 3) and level 4 (n = 3) evidence.

Management

Eight studies described management of ICR (Table II).[37−44] The average quality of evidence for studies evaluating management of ICR was 3.7 (range 2b−4). The proposed management strategies included watchful waiting (n = 1; 2b); disk repositioning (n = 2; 3b, 4); condylectomy with costochondral grafting (n = 3, level 4 for all 3 studies), condylectomy with TMJ alloplastic prostheses (n = 1; level 4); and other (n = 1; level 4).

DISCUSSION

The purpose of this study was to assess etiology and management strategies for ICR as reported in the literature. The specific aims were to (1) characterize the studies assessing etiology of ICR, (2) identify management strategies for ICR, and (3) characterize the quality of the studies according to CEBM criteria. This study found that there was no consensus in the literature on the etiology of ICR, that management options for ICR were variable, and that the overall quality of evidence guiding clinicians is poor.

No clear consensus on the etiology or risk factors for ICR exists in the current literature. Most studies are case series with limited numbers patients and comparison groups and, therefore, were excluded from this study. The theories of etiology and risk factors that have been studied included demographic, hormonal, and postorthognathic surgical factors. Orthognathic surgical risk factors included the use of maxillomandibular fixation postoperatively [7] and high mandibular plane angle and posteriorly inclined condylar neck after bilateral sagittal split osteotomy (BSSO), presumably causing unfavorable joint loading.[2,45−47] Other theories reported in case reports and opinion articles (which were excluded from this review) suggested that ICR could be initiated by abnormal functional loading of the temporomandibular joints (TMJs) caused by (1) changes in occlusion (orthognathic surgery, orthodontia, prosthetic dental restoration); (2) internal derangement of TMJs; (3) parafunction of TMJs; (4) trauma (e.g., condylar fracture); and/or (5) unstable occlusion. Avascular necrosis of the condyles after orthodontic/surgical intervention is also a prevalent theory,[46,47] but no related study qualified for inclusion in this review.

A prominent proposed etiology is 17β-estradiol deficiency, which is a risk factor for ICR development.[2] Gunson et al.[2] assessed 17β-estradiol levels in 27 patients with ICR and found that those with ICR were more likely to have levels below the published norms. However, lack of a comparison or control group in the study limits the conclusions that can be drawn. The

Table I. Studies describing etiology of and risk factors for idiopathic condylar resorption

Author	*Year*	*Location*	*Etiology or treatment*	*Conclusion*	*Category*	*CEBM rating*
1. Huang et al.[7]	1982	Canada	Etiology	Mandibular lengthening of ≥ 11 mm in growing children is associated with relapse and remodeling of the condyle or posterior symphysis or both.	Surgical	2b
2. Kerstens et al.[8]	1990	The Netherlands	Etiology	Orthognathic surgery may stimulate progress of condylar resorption by increased loading, disk displacement, and immobilization	Surgical	4
3. Moore et al.[9]	1991	The Netherlands	Etiology	Preoperative factors that contribute to condylar resorption include age, gender, high preoperative mandibular plane angle, prior evidence of temporomandibular disease Intraoperative factors include amount of force and direction used by surgeon to position proximal segment	Anatomic	4
4. Cassidy Jr. et al.[10]	1993	USA	Etiology	Patients who had surgical treatment of class II, division 1, malocclusions can potentially have a risk of relapse as a result of condylar resorption	Anatomic	2b
5. de Mol van Otterloo et al.[11]	1993	The Netherlands	Etiology	Condylar atrophy seen in 2 patients with high mandibular plane angles and mandibular retrognathism The authors propose this anatomy higher risk for condylar atrophy	Anatomic	4
6. Bouwman et al.[12]	1994	The Netherlands	Etiology	Intermaxillary fixation is potentially correlated with condylar resorption after orthognathic surgery in patients who mandibular deficiency and high mandibular plane angle	Surgical	2b
7. De Clercq et al.[3]	1994	Belgium	Etiology	Female patients with high angle retrognathism had higher risk of postoperative condylar resorption The authors propose condylar resorption occurs exclusively in female patients and develops 2 months to 2 years postoperatively No correlation found with surgical condylar resorption to severity of retrognathism	Other	4
8. Scheerlinck et al.[13]	1994	The Netherlands, Belgium	Etiology	The magnitude of advancement with BSSO is related to progressive condylar resorption	Surgical	4
9. Stewart et al.[14]	1996	UK	Etiology	Inflammation with presence of local neuropeptides causes lysis of articular cartilage and condylar resorption	Systemic	4
10. Cutbirth et al.[15]	1998	USA	Etiology	Large magnitude mandibular advancements with BSSO and fixation with 3 bicortical screws per side in patients with preoperative TMJ symptoms are risk factors for condylar resorption	Surgical	4
11. Hoppenreijis et al.[16]	1998	The Netherlands	Etiology	Rigid internal fixation in bimaxillary osteotomies is a risk factor for condylar resorption 19% of patients developed progressive condylar resorption No difference between mini-plate osteosynthesis or positional screws for BSSOs	Surgical	2b

(*continued*)

Table I. Continued

Author	*Year*	*Location*	*Etiology or treatment*	*Conclusion*	*Category*	*CEBM rating*
12. Joos et al.[17]	1999	Germany	Etiology	Adjustable bone fixation system for sagittal split ramus osteotomy does not cause condylar resorptive changes	Surgical	4
13. Hwang et al.[4]	2000	Switzerland	Etiology	Posteriorly inclined condylar neck is a risk factor for condylar resorption	Anatomic	3b
14. Hwang et al.[18]	2000	Switzerland	Etiology	Counterclockwise rotation of distal and proximal mandibular segments and surgically induced (iatrogenic) posterior condyle displacement increases risk of postoperative condylar resorption	Surgical	4
15. Cabrini Gabrielli et al.[19]	2003	Brazil	Etiology	Condylar resorption developed in 6.25% of fixed condylar fractures	Surgical	4
16. Azumi et al.[20]	2004	Japan	Etiology	Condylar resorption occurred in 20% of condyles after mandibular distraction osteogenesis and correlated with preoperative articular disk displacement and with the amount of condylar displacement during treatment	Surgical	4
17. Borstlap et al. (Part II) [21]	2005	The Netherlands	Etiology	4% of patients (n = 222) treated with BSSO fixated with miniplates developed condylar resorption. Young patients (< 14 years), steep mandibular plane angle and low facial height correlated with resorption	Surgical	4
18. Hwang et al.[22]	2004	Switzerland	Etiology	Younger age, and posteriorly inclined condylar neck were risk factors for condylar resorption	Anatomic	3b
19. Eggensperger et al.[23]	2005	Switzerland	Etiology	After 12 years, relapse in patients who underwent mandibular setback resulting from condylar resorption and loss of ramus height	Surgical	2b
20. Eggensperger et al. (Part II) [24]	2006	Switzerland	Etiology	Preoperative high mandibulonasal plane angle associated with long-term skeletal relapse and loss of ramus height	Anatomic	2b
21. Gunson et al.[2]	2009	USA	Etiology	Low levels of 17β-estradiol potentially related to cortical and medullary condylysis	Systemic	2b
22. Wohlwender et al.[25]	2010	The Netherlands	Etiology	Unilateral sagittal split osteotomies do not have adverse effects in causing condylar resorption compared to BSSO	Surgical	2b
23. Kobayashi et al.[26]	2011	Japan	Etiology	Potential risk factors for ICR include clockwise rotation of mandible, retrognathism, and preoperative erosion or deformity of condyle	Surgical	4
24. Dicker et al.[27]	2012	The Netherlands	Etiology	Postoperative condylar loading is not a risk factor for condylar resorption or relapse	Anatomic	4
25. Xi et al.[28]	2014	The Netherlands	Etiology	Gender, preoperative condylar volume, amount of downward displacement of pogonion were predictors of postoperative condylar resorption	Other	2b
26. Dicker et al.[29]	2015	The Netherlands	Etiology	Pterygomasseteric sling elongation and condylar rotation after BSSO advancement was not associated with condylar resorption	Surgical	4
27. Yang et al.[30]	2015	South Korea	Etiology	In patients with preoperative ICR, patients with the lowest bone density	Other	2b

(*continued*)

Table I. Continued

Author	Year	Location	Etiology or treatment	Conclusion	Category	CEBM rating
				and those undergoing large magnitude advancements had more relapse/condylar resorption		
28. Saito et al.[31]	2016	Japan	Etiology	Risk factors for condylar resorption were unclear	Other	4
29. Sant'Ana et al.[32]	2016	Brazil	Etiology	Condylar resorption may be attributable to orthognathic surgery when there is significant condylar repositioning in fossa	Surgical	2b
30. Aneja et al.[33]	2017		Etiology	Females (age < 18 years), steep mandibular plane angle, low facial height ratio (posterior/anterior), magnitude of surgery (BSSO with > 6 mm advancement) risk factors for ICR	Other	2b
31. Nogami et al.[34]	2017	Japan	Etiology	Condylar resorption most likely after treatment of mandibular retrognathism with pre-existing TMJ pathologic abnormality	Anatomic	3b
32. Kajii et al.[35]	2018	Japan	Etiology	Compared class II patients with and patients without ICR. Those with ICR had more posterior rotation of ramus, smaller condyles, more retroclined maxillary incisors and steeper occlusal plane	Anatomic	4
33. Alsabban et al.[36]	2018	USA	Etiology	Hormonal imbalance, oral contraceptives, class II malocclusion associated with ICR	Other	4

BSSO, bilateral sagittal split osteotomy; *CEBM,* Oxford Center for Evidence-Based Medicine; *ICR,* idiopathic condylar resorption; *TMJ,* temporomandibular joint.

low estrogen level theory is supported by an in vivo animal study by Wu et al.,[48] which found that mechanical stress (resin buildup on the right molars) applied to the mandible in mice with estrogen deficiency led to more extensive condylar degeneration compared with mice with normal estrogen levels. Mice with low 17β-estradiol levels had the lowest cartilage thickness and highest levels of osteoarthritis (osteoclastic activity and apoptosis) on histologic analysis.[48] Another study by Feng et al.[49] found higher levels of 17β-estradiol and lower levels of hyaluronan in synovial fluid collected from patients with ICR compared with samples from patients with disk displacement or osteoarthritis. In cell culture, the authors showed that estradiol upregulates expression of micro-RNA, repressing hyaluronan levels, which are thought to be protective of the joint.[49] These findings further implicate the role of 17β-estradiol as a factor in the development of ICR.

Management of ICR depends on disease activity, the stability of the condyle ramus unit, and the overall trajectory of the disease. ICR is considered a self-limiting process, with disease duration of 6 months to 2 years.[50] If the disease is quiescent and enough condylar mass remains, standard orthognathic treatment may be employed.[50] However, no test or biomarker exists to determine which cases will progress or remain quiescent. Some reports of standard orthognathic surgery (without joint treatment) in patients with ICR have found an unacceptably high relapse rate.[2,51] Repositioning of the articular disk, in conjunction with orthognathic surgery, has been shown to increase the stability of orthognathic correction.[1,52]

With active ICR or inadequate condylar anatomy to provide a reliable centric-relation position, joint reconstruction is usually necessary. The most successful management, as reported in the literature, is condylectomy and reconstruction with an autogenous or alloplastic joint. A recent study assessing endoscopic condylectomy and costochondral graft reconstruction found stable overbite and mandibular length in 26 patients, followed up, on average, for 3.65 years.[43] In this study, the articular disk was maintained and not manipulated, and this argues against the necessity for disk repositioning. Mehra et al. reported the use of total alloplastic joint prostheses for condylar resorption, with stable results observed at a mean follow-up of 6.2 years (see Table 2).[42]

CONCLUSIONS

In summary, a review of the literature revealed a low level of evidence regarding the etiology and management of ICR. Removal of the diseased condyle and

Table II. Studies describing treatment of idiopathic condylar resorption

Author	*Year*	*Location*	*Number of patients*	*Follow-up duration*	*Treatment*	*Summary*	*CEBM rating*
1. Merkx et al.[37]	1994	The Netherlands	8	3.5 months to 3 years	Treatment	Revision surgery for treatment of ICR was unsatisfactory, with poor aesthetics and poor occlusal stability	4
2. Huang et al.[38]	1997	USA	18	2 years	Treatment	Condylectomy and costochondral grafting produces stable functional results	4
3. Hoppenreijis et al.[39]	1999	The Netherlands	26	16−135 months	Treatment	Progressive condylar resorption after orthognathic surgery stops after 2 years Second orthognathic surgery produces variable results but general improvement	2 b
4. Troulis et al.[40]	2004	USA	10	6 months	Treatment	Endoscopic condylectomy and costochondral graft reconstruction is possible for treatment of idiopathic condylar resorption	4
5. Bodine et al.[41]	2016	USA	22	1 year	Treatment	Disk repositioning and stabilization using Mitek mini-anchor technique showed postoperative normalization of condylar growth	3 b
6. Mehra et al.[42]	2016	USA	21	6.2 years (range 5−12 years)	Treatment	ICR can be treated effectively with total TMJ prosthesis and maxillary orthognathic surgery	4
7. Peacock et al.[43]	2019	USA	26	3.65 years (range 1.11−17.1 years)	Treatment	Endoscopic condylectomy and costochondral graft reconstruction is stable and feasible for reconstruction of ICR	4
8. Galiano [44]	2019	Brazil	24	30.3 months	Treatment	Condylar resorption can be treated with disk repositioning and orthognathic surgery	4

CEBM, Oxford Center for Evidence-Based Medicine; *ICR,* idiopathic condylar resorption; *TMJ,* temporomandibular joint.

autogenous or alloplastic reconstruction were the most stable treatment options. Collaboration among specialties, including rheumatology, endocrinology, and oral and maxillofacial surgery, is recommended to gain better insight into the etiology and management of ICR.

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