

SCIENTIFIC
SECTION

Severe retrognathia as a risk factor for recent onset painful TMJ disorders among adult females

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Objective: To evaluate the importance of severe retrognathia as a risk factor for the development of recent onset painful TMJ disorders among adult females.

Design: Case-control study.

Setting: This study was conducted in a large health maintenance organization between 1998 and 1999 [Kaiser Permanente Northwest (KPNW), Portland, OR, USA].

Participants: Adult females with recent onset painful TMJ disorders ($n=29$) and normal controls ($n=104$).

Methods: Cases were recruited from the TMD clinic at Kaiser Permanente Northwest (KPNW). Controls were recruited from a dental clinic at KPNW. Case status was determined using a questionnaire; mandibular sagittal position was determined by measuring a research angle on facial photographs. The mean research angle for cases was compared to the mean for controls. Multivariable exact conditional logistic regression analysis was used to examine the demographic characteristics of cases and controls, and to determine the strength of association between recent onset painful TMJ disorders and severe retrognathia. The population attributable risk percentage (PAR%) and the attributable risk percentage (AR%) were calculated to further evaluate severe retrognathia as a risk factor.

Results: The mean research angle among cases (67.7° ; 95% CI=66.0–69.4) was smaller than among controls (71.6° ; 95% CI=70.7–72.5, $P<0.001$). The odds ratio for the association between case status and the presence or absence of severe retrognathia was elevated (OR=6.3; 95% CI=1.1–47.5, $P=0.039$). The PAR% and AR%, associated with severe retrognathia, were 13.3 and 84.1%, respectively.

Conclusions: Severe retrognathia is strongly associated with recent onset painful TMJ disorders (OR=6.3). Only a small proportion of these disorders are attributable to severe retrognathia among the total population of adult females (PAR%=13.3%). However, a large proportion of these TMJ disorders are potentially attributable to severe retrognathia among adult females with severe retrognathia (AR%=84.1%).

Key words: Retrognathia, temporomandibular joint

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Introduction

Several studies suggest an association between abnormal mandibular morphology and temporomandibular joint (TMJ) disorders, a subset of temporomandibular disorders (TMD).^{1–11} Previous research indicates that age, race, gender, and socio-economic status are potentially associated with both retrognathia and TMJ disorders.^{10,12–22} Taking into account these potential

confounders, the association between various forms of occlusion and TMD, and more specifically between mandibular retrognathia and TMJ disorders, still appears to exist.^{9–11}

Some authors indicate this association may occur because TMJ disorders alter mandibular development.^{5,11} Other authors indicate that the temporal relationship may be in the opposite direction, with abnormal mandibular morphology influencing the

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development of TMJ disorders.^{1,7,10} Twenty years ago Moyers²³ recognized that the temporal relationship of this association was uncertain. Nebbe *et al.*⁸ summarize this concept, 'Although associations between TMJ disk displacement and facial pattern have been demonstrated, a cause-effect relationship cannot be assumed. Disk displacement may affect facial growth or disk displacement may occur as a consequence of biomechanics associated with an altered facial pattern. It is also possible that both disk displacement and facial pattern are the result of another influencing factor not yet identified.' Furthermore, studies have questioned the importance of any purported association by noting that many factors are related to TMD and that only a small proportion of the overall occurrence of TMD is potentially explained by occlusal factors.^{24,25}

The objective of this study is to evaluate the importance of severe retrognathia as a risk factor for the development of recent onset painful TMJ disorders among adult females. The hypothesis of this study is that severe retrognathia is strongly associated with recent onset painful TMJ disorders. It is further suggested that, while only a small proportion of these disorders may be attributable to severe retrognathia among the total population of adult females, a much larger proportion is attributable to severe retrognathia among adult females with severe retrognathia.

Methods and materials

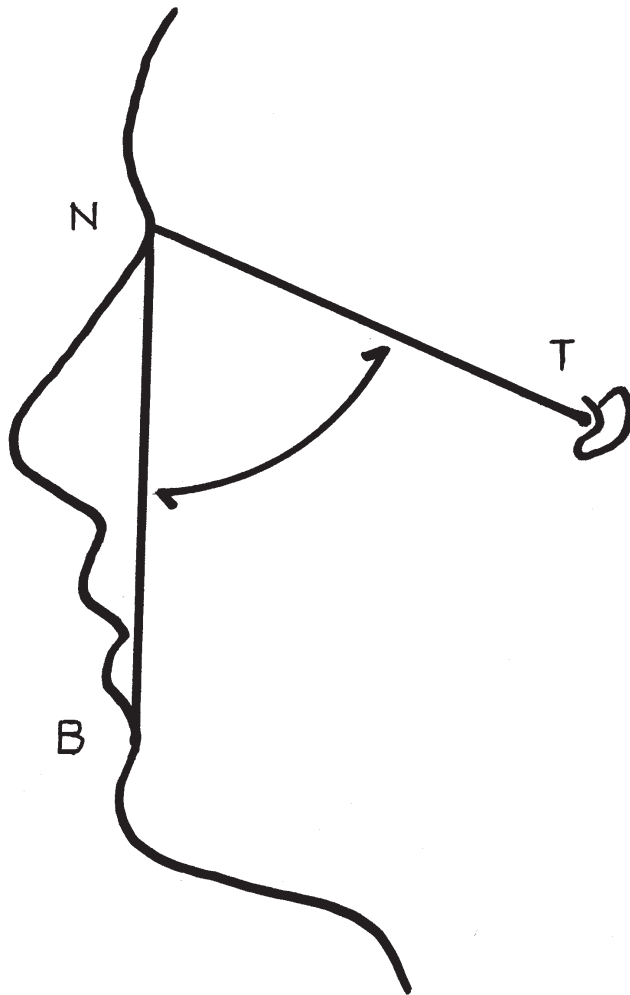
A case-control study was conducted between 1998 and 1999 according to procedures approved by the Human Subject Review Committees of KPNW and the University of Washington (UW, Seattle, WA). Subjects came from a previous multi-centre study on mandibular retrognathia and TMJ disorders.¹⁰ Subjects in the current study were restricted to 18–70-year-old females from a single centre (KPNW).

Cases were recruited from enrollees who presented to the TMD clinic for evaluation and treatment, and were restricted to participants that reported the onset of TMJ pain within the previous 3 years. Controls were recruited from adult female enrollees being seen at a KPNW dental clinic for a routine dental prophylaxis. Case status was determined by the use of a study questionnaire that solicited information pertaining to a history of TMJ symptoms (clicking, catching, joint pain) and also surveyed for general demographic characteristics (age, race, level of educational attainment). In order for a woman to be eligible as a case, she had to report the presence of recent onset TMJ pain (within 3 years) on the study questionnaire; controls

could not report any history of clicking, catching, or joint pain. To the best of our knowledge, no participants in this study reported or were diagnosed with rheumatoid arthritis. In the KPNW TMD Clinic, the attending dentist for the cases provided clinical diagnoses.²⁶ In the KPNW dental clinic, the normal protocol for examining dental hygiene patients was followed. One research assistant at KPNW (blinded) was responsible for recruitment and questionnaire administration, as well as obtaining facial photographs for both cases and controls.

A profile photograph of each participant's face was taken using a Polaroid Spectra camera to record mandibular sagittal position. The participants were instructed to stand with a natural head position, lips lightly touching in a relaxed posture and teeth slightly apart. Each photograph was developed, digitized and stored in a computer file. The following three soft tissue landmarks were identified on the digitized images: tragus, soft tissue nasion and soft tissue B-point. A research angle was constructed by the intersection of two lines, one from tragus to soft tissue nasion, and the other from soft tissue nasion to soft tissue B-point (Figure 1). This angle was measured twice by two individuals (JRM and another research assistant, also blinded). The inter- and intra-rater reliability of these measurements both exceeded 0.95 as determined using the intra-class correlation coefficient. The two measurements for each participant, obtained by the research assistant, were then averaged. Each participant was categorized as either being severely retrognathic (less than 63.2°) or not (63.2° or greater). The cut-off value for severe retrognathia was determined by using an estimate of the population mean of the research angle for adult females (mean=70.6°) and then subtracting two times an estimate of the standard deviation of the skeletal pattern underlying the research angle (SD=3.7°).¹⁰

The mean research angle for cases was compared to that for controls using a two-sample *t*-test. Potential interactions between the research angle and other variables, including age, race and education level were examined. These variables were also examined for their potential confounding effect on the association between case status and the presence or absence of severe retrognathia. Multivariable exact conditional logistic regression analysis was used to calculate the odds ratios (OR) and associated 95% confidence intervals (CI) to determine the association between case status (dependent variable), and the presence or absence of severe retrognathia (independent variable), conditional on age, race, and education level (potential confounders). The population attributable risk percentage (PAR%) and the



T, tragus; N, soft tissue nasion; B, soft tissue B-point

Figure 1 Soft tissue landmarks and research angle used to measure mandibular sagittal position

attributable risk percentage (AR%) were calculated using the odds ratio (OR) as an estimate for relative risk (RR) and the prevalence of severe retrognathia among controls (Pe).²⁷ All data analyses were performed using SPSS (SPSS Inc., Chicago, IL).

Results

Among those 18–70-year-old women approached by the research assistant, 30 cases and 108 controls agreed to participate in this study, with response rates of 95 and 80%, respectively. The power to detect an elevated odds ratio ($OR \geq 7.0$) indicating a strong association between recent onset painful TMJ disorders and severe retrognathia is 72–81% using a two-sided test at a 0.05 significance level, given the current sample size and

assuming a 3–4% prevalence of severe retrognathia among controls. Because five participants had missing data, the actual number of cases ($n=29$) and controls ($n=104$) included in the statistical analysis was slightly less than the total number of participants recruited.

Cases tended to be younger, more likely to be white, and had fewer years of education (Table 1). For the participants that were classified as cases according to the study questionnaire, we obtained the TMD diagnoses made by their dentist. All cases reported a history of recent joint pain on the study questionnaire; however, they had diverse clinical diagnoses upon examination by their dentist, including muscle pain, disk displacement, joint pain and/or degenerative joint disease. Twenty-five of the 29 cases included in the analysis had a TMJ disorder upon examination, while four had only muscle pain. Sixteen of the 25 cases diagnosed with a TMJ disorder had joint pain as one of their diagnoses; the other nine had muscle pain associated with a disk displacement disorder (Table 2).

The mean research angle among cases (67.7° ; 95% CI=66.0–69.4) was smaller than among controls (71.6° ; 95% CI=70.7–72.5, $P < 0.001$). No significant interactions were detected between the research angle and the other covariates (age, race or education level). The odds ratio for the association between case status and presence or absence of severe retrognathia was elevated (OR=6.3; 95% CI=1.1–47.5, conditional on age, race and education level, $P=0.039$; Table 1). When we removed

Table 1 Association between recent onset painful TMJ disorders and severe retrognathia, among adult females, conditional on age, race and education level

	Cases ($n=29$)	Controls ($n=104$)	Odds Ratio† (95% CI)
	<i>n</i>	<i>n</i>	
Age			
41–70	9	53	0.4 (0.1–1.2)
18–40	20	51	1.0
Race			
Non-White	2	31	0.1 (< 0.1–0.7)
White	27	73	1.0
Education Level			
Post-Baccalaureate	5	28	0.3 (0.1–1.2)
College	11	47	0.5 (0.1–1.5)
High school or less	13	29	1.0
Retrognathia			
Severe retrognathia	6	3	6.3 (1.1–47.5)
No severe retrognathia	23	101	1.0

†Conditional on all variables shown, using exact conditional logistic regression analysis

Table 2 Prevalence of combinations of clinical diagnoses for 29 cases of recent onset painful TMJ disorders among adult females

M.p	Disk	J.p	DJD	Cases
√	√	√		9
√	√			9
√				4
√	√	√	√	3
	√	√		2
	√	√	√	1
√		√		1

M.p, muscle pain (right or left);
 Disk, disk displacement (right or left);
 J.p, joint pain (right or left);
 DJD, degenerative joint disease (right or left)

the four cases diagnosed with only muscle pain and re-analysed the data, the odds ratio remained substantially the same. When we reanalysed the data using the 16 cases diagnosed with joint pain, the point estimate of the odds ratio became markedly more elevated.

Using the odds ratio (OR=6.28) as an estimate for the relative risk (RR) and the prevalence of severe retrognathia among controls (Pe=0.029), the population attributable risk percent (PAR%) and the attributable risk percent (AR%), associated with severe retrognathia, were determined to be 13.3 and 84.1%, respectively (Table 3).

Discussion

We found a stronger association between severe retrognathia and recent onset painful TMJ disorders than previously suggested in the literature.^{1-11,24} This may be due to the very restricted case definition we used. Since we restricted the cases in this study to recent onset (≤3 years) painful TMJ disorders among adult females, we believe it is reasonable to assume that mandibular sagittal position was established prior to the development of the TMJ disorders. If this assumption is correct, then PAR% and AR% can be used to further investigate the importance of severe retrognathia for the development of recent onset painful TMJ disorders among adult females. The small value for PAR% observed in our study, reinforces the current consensus that there are many factors that contribute to the development of TMD (or TMJ disorders) and it is unlikely that any one variable would explain much of the total occurrence of TMD.^{24,25} However, when we examined the occurrence of recent onset painful TMJ disorders, among adult females with severe retrognathia, the picture changed. The proportion of recent onset

Table 3 Proportion of recent onset painful TMJ disorders among adult females potentially attributable to severe retrognathia

Measurements of risk	Formula*	Estimated value
Population attributable risk percentage	PAR%= [(It - Io)/It] × 100	13.3%
Attributable risk percentage	AR% = [(Ie-Io)/Ie] × 100	84.1%

*Used alternative formulas not dependent on incidence rates. PAR%={Pe)(RR-1)/[(Pe)(RR-1) + 1]} × 100 and AR%=[(RR-1)/RR] × 100, where relative risk (RR) is estimated by the odds ratio (OR=6.28), and prevalence of severe retrognathia among controls (Pe) is estimated by the value 3/104 (Pe=0.029)

painful TMJ disorders among adult females with severe retrognathia that might be attributable to severe retrognathia was large (AR%=84.1%). Thus, among adult females with severe retrognathia, if an individual develops a recent onset painful TMJ disorder it is quite possible that the severe retrognathia was an important risk factor. However, the findings need to be put in context.

These results do not mean that clinicians should try to prevent TMJ disorders by screening and treating severe retrognathia among adult females. Cause and effect have after all not been shown. Furthermore, our previous multi-site study demonstrated that incidence rates for TMJ disorders among adult females seeking treatment are small.¹⁰ Since all eligible adult females at the KPNW TMD clinic may not have been approached, and because some symptomatic enrollees may not have been referred to or sought treatment at the TMD clinic, any incidence rates based on this study would probably under-estimate the true rates. Taking this into account, it is still likely that the actual number of recent onset painful TMJ disorders that might be attributable to severe retrognathia is quite small. Even if it were practical to screen most adult females, addressing the risk associated with severe retrognathia would probably only prevent a very small number of recent onset painful TMJ disorders. Calculating the population attributable risk (PAR) and the attributable risk (AR) using hypothetical, but reasonable, incidence rates illustrate this concept (Table 4).

Our study had several limitations. A case-control study design was used instead of a prospective cohort design. The participants were restricted to adult females. A questionnaire was used to determine case status and facial photographs were used to measure the research angle to determine the presence or absence of severe retrognathia. The reasons for these limitations and their potential effects on this study follow.

Table 4 Illustration of calculation of measurements of risk using hypothetical incidence rates for recent onset painful TMJ disorders

Measurements of risk	Formula	Values
Incidence among total adult females	It	8 per 10,000*
Incidence among adult females with S. Retrog.	Ie	47 per 10,000*
Incidence among adult females without S. Retrog.	Io	7 per 10,000*
Population attributable risk	PAR=It-Io	1 per 10,000†
Attributable risk	AR=Ie-Io	40 per 10,000†
Relative risk (OR ≈ RR)	RR=Ie/Io	6.7†
Population attributable risk percentage	PAR%=[(It-Io)/It] × 100	12.5%†
Attributable risk percentage	AR%=[(Ie-Io)/Ie] × 100	85.1%†

*Hypothetical incidence rates, for illustrative purposes only

†Calculated values using hypothetical incidence rates, for illustrative purposes only
S. Retrog., severe retrognathia

During the planning of this study, we anticipated that the incidence of recent onset painful TMJ disorders would be small, which precluded the use of a prospective cohort study design. This decision is supported by a recent prospective longitudinal study where girls with untreated Class II malocclusions and normal occlusions were followed for 2 years and very few new cases of TMJ tenderness were observed.²⁸

An exact conditional logistic regression analysis was used to help address the issue of low cell counts. This still resulted in an elevated point estimate for the odds ratio associated with severe retrognathia, but the 95% CI was quite wide. The latter is noteworthy as it is indicative of the uncertainty associated with the estimate for the odds ratio associated with severe retrognathia observed in this study.

We restricted the study to females since they seek treatment for temporomandibular disorders (TMD) much more frequently than men.^{13,14} Because our study was limited to adult females seeking treatment, it is unknown whether our results can be generalized to adolescent females, adult females not seeking treatment or males. Less was known about the relationship of age, race and socio-economic status to TMJ disorders, so we chose not to restrict or match on these variables. The logistic regression analysis was conditional on several variables (age, race and education level) that were known or suspected to be related to both the exposure and disease, and thereby qualified as a potential confounder. Certainly, there could be some unknown confounders that could have contributed to the strong odds ratio we observed, but it is unlikely that an unknown confounder could explain all of this association.

Examination by an experienced TMD clinician, augmented by ancillary images or tests, is the gold standard for diagnosing temporomandibular disorders.^{29,30} However, it was not possible to have TMD

clinicians examine the controls because of the physical separation of the clinics. A study questionnaire, that proved accurate in classifying cases when compared to the diagnoses provided by the attending dentist, was used to select cases with TMJ disorders from among TMD patients at the TMD clinic. It was also used to eliminate potential controls with unrecognized TMJ disorders from among the dental hygiene patients at the dental clinic.¹⁰ It was not possible to compare the classification of controls via the questionnaire with diagnoses that might have occurred upon examination by a TMD clinician. However, it is unlikely that an adult female would be unaware of a recent onset painful TMJ disorder when presenting to a KPNW dental clinic for a routine dental hygiene appointment. The negative responses on the study questionnaire regarding TMJ symptoms make the likelihood of misclassification of a control even more remote.

Ancillary images, including magnetic resonance images (MRIs), were not routinely available for the cases and controls. Having access to this information might have improved our classification because individuals can have undiagnosed disk displacements.^{11,31,32} If MRIs had been available for all participants, some severely retrognathic participants (exposed) and some participants without severe retrognathia (non-exposed) might have had their case status changed. These images presumably could have been obtained in a non-biased manner. Regardless, it is difficult to predict how these potential reclassifications might have changed our results or conclusions. Also, if the disk displacements in our cases had actually been present for a long time, but only became symptomatic within the 3 years prior to this study, it might have changed our assumption concerning the temporal relationship between severe retrognathia and recent onset painful TMJ disorders. Perhaps a larger, more refined, case-control study or a large prospective cohort study could better address these

issues, especially if ancillary images were available for all participants. However, neither of these study designs would resolve the issue of unknown confounders, since it is not possible to randomize with respect to the presence or absence of severe retrognathia.

The gold standard for determining mandibular sagittal position is the lateral cephalometric radiograph, but the Human Subjects Review Committees at KPNW and the UW wouldn't allow cephalometric radiographs to be taken on the controls. A standardized protocol for obtaining facial photographs was developed as an instrument to measure the research angle, and determine the presence or absence of severe retrognathia in both cases and controls.¹⁰ Photographs have proven to provide reliable and valid measurements of facial skeletal morphology, particularly when all measurements are obtained by one individual.^{33–38} The research angle was constructed to be highly correlated with the degree of abnormality in the underlying facial skeletal pattern. We chose to use the mean research angle of the controls from a previous multi-site study (70.6°) as an estimate of the mean research angle for the population of adult females.¹⁰ We used this value instead of the mean research angle for the controls from this study (71.6°) because we felt that the value from the previous and larger multi-site study was a better population estimate. We did re-analyse the data using the larger mean value and it did not change our conclusions. Since we believed that the soft tissue research angle would be more variable than its corresponding skeletal pattern, we derived the standard deviation used in this study (SD=3.7°) from previously published standard deviations for a closely corresponding skeletal angular measurement (S–N–B).^{10,39} Although we believe the research angles for the cases and controls were measured accurately using the same protocol, any misclassification that might have occurred could have distorted the observed odds ratio, producing either an under- or over-estimate, depending on the type of misclassification.²⁷

Our study had several strengths. One research assistant (blinded) recruited all of the cases and controls from a well-defined group (KPNW) with high response rates. Cases were recruited from one TMD clinic and controls were recruited from one dental clinic. The case definition was narrow and limited to recent onset painful TMJ disorders among adult females. The same research assistant that recruited the participants administered all the questionnaires, and obtained all of the facial photographs for both cases and controls. The clinical diagnoses for the cases were provided by one experienced TMD clinician. Finally, a second research assistant (blinded) measured all the research angles.

In summary, there is mounting evidence of an association between abnormal facial morphology and TMJ disorders. Our study confirms a strong association between recent onset painful TMJ disorders and severe retrognathia among adult females. Furthermore, it provides evidence that severe retrognathia may precede and be an important risk factor for the development of recent onset painful TMJ disorders among adult women with severe retrognathia.

Conclusions

- Severe retrognathia is strongly associated with recent onset painful TMJ disorders.
- Among the total population of adult females, only a small proportion of recent onset painful TMJ disorders is attributable to severe retrognathia.
- Among adult females with severe retrognathia, a large proportion of recent onset painful TMJ disorders is potentially attributable to severe retrognathia.

Authors and contributors

James Miller contributed substantially to all phases of this study and article. Cathy Critchlow contributed substantially to study design, analysis and interpretation of data, drafting and revision of the article, and final approval of the version to be published. Lloyd Mancl contributed substantially to analysis and interpretation of the data, revision of the article and final approval of the version to be published. Clinicians and staff from Kaiser Permanente recruited the participants and collected data for this study. James Miller is the guarantor.

Disclaimer

The opinions expressed in this article are those of the authors and do not necessarily represent the views of Kaiser Permanente or the University of Washington.

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