Dental Traumatology

*Dental Traumatology* 2010; **26**: 332–337; doi: 10.1111/j.1600-9657.2010.00892.x

# Bifid mandibular condyle with tempromandibular joint ankylosis – a pooled data analysis

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Accepted 2 February, 2010

**Abstract** – *Background and objectives*: Bifid mandibular condyle (BMC) with associated temporomandibular joint ankylosis (TMJA) is extremely rare with only sixteen cases reported worldwide. This article presents the largest case series with 12 patients of BMC with TMJA with treatment results in an attempt to elucidate the morphological pattern in this rare condition. *Materials and method*: Retrospective examination of Computed Tomograms (CT) over a period of 9 years revealed that 12 cases had BMC with TMJA. Demographical, Clinical, CT features and treatment results of these were analyzed. Patients were grouped according to sides of involvement and orientation of condyles. Data of 12 Indian cases were noted and pooled from existing literature from India and analysis performed using statistical software. Descriptive statistics and one way ANOVA were used to find association. *Results*: The male: female ratio was 1:1. Twenty three cases were post-traumatic and one postinfectious. The etiology was sustained in childhood in all patients. The mean presenting age was 16.92 ± 11.05 years. Of all the cases 66.7% presented with ipsilateral chin deviation. The mean mouth opening was 3.89 ± 5.4 mm. Bilateral BMC was observed in 29% of cases and 87.5% of all cases had mesiolateral orientation. In general, ankylosed heads were mushroom shaped compared with non-ankylosed heads. The result of 1 year postoperative mouth opening was compared with treatment modalities used in this center. *Conclusions*: This series attempts to elucidate patterns of ankylosis and CT morphology in BMC with associated TMJA. Also included are treatment results of BMC with TMJA. This case series is the largest presented and includes youngest case of TMJA with BMC reported so far in English literature.

The bifid mandibular condyle (BMC) has been described as a condition of unknown etiology and uncertain pathogenesis. Many see it as the product of accidental trauma or forceps delivery, with the two heads occurring one behind the other in the sagittal plane. In bioanthropological literature, 'bifid condyle' often describes pitting in the sagittal plane, dividing the condyle mediolaterally (1). This condition involves duplication of the mandibular condyle and is increasingly being detected due to the frequent use of improved imaging techniques, particularly CT and MRI. Most cases are asymptomatic and incidentally detected. BMC associated with temporomandibular joint ankylosis (TMJA) is an extremely rare abnormality with a handful of reported cases across globe. Owing to paucity of the cases reported, this condition is not entirely understood with regard to etiology, clinical implications and morphology. An extensive search revealed only 16 cases of BMC with TMJA reported in the English medical literature (2–7).

This article presents the review of the reported Indian (6, 7) cases added with our own 12 cases of BMC with TMJA, in an attempt to elucidate the few clinical and morphological patterns in this rare condition. We also present center's experience in treating such surgeries.

## Materials and method

All archived cases treated for TMJ related problems from June 1999 to June 2008 were re-reviewed for BMC. There were 121 cases treated during the period for TMJ ankylosis. Overall 15 cases were diagnosed with BMC. There were 12 cases of BMC with marked evidence of TMJA features on CT. TMJA and developing TMJA was identified by the presence of a markedly reduced joint space and irregular, enlarged joint margins with fibrous and or bony bridging. These patients had well formed double or bifid mandibular condyle separated by a shallow or deep groove. This group of patients from this center provided the materials for the current study. Patients with small bony processes projecting from the mandibular ramus or condylar neck, without a well formed duplicated condyle, were not included. Though

these patients had undergone preliminary investigations such as plain radiography and orthopantomograms as they were neither digitalized nor stored and were not available for some of the earlier cases, hence these details were not considered. Clinical details of this group including demography, etiology, duration of symptoms and clinical findings were analyzed from medical records. CT records of TMJs in these patients were analyzed by the author. These CTs were taken by an independent private radiologist over the period of time of 10 years. Based on the CT features patients were divided into two groups: those with bifid condyles in one TMJ only (unilateral BMC) and those with bifid condyles on both sides (bilateral BMC). Patients were then classified with regard to the relation of one condylar process to the other as mediolateral (ML) or anteroposterior (AP). These were further grouped, based on the presence of TMJA, as unilateral or bilateral TMJA. Finally, the condylar process ankylosed was also considered – whether medial or lateral condylar process (in ML BMC) and anterior or posterior process (in AP BMC).

Data available from other Indian papers (6, 7) were retrieved from the articles and categorized as above and pooled. If any of the details were found to be inadequate or could not be summarized, they were marked as missing or not known.

The morphology of the BMC and treatment method and resulting mouth opening of cases treated at presented center were tabulated and compared with pretreatment values.

### Statistics

All the data were entered and analyzed using SPSS software version 16.0 (SPSS Inc., Chicago, IL, USA). Descriptive statistics were presented for all variables from all papers and overall cases. One way ANOVA was performed for finding the association of age and interincisor opening for gender, etiology, center, site of bifid and ankylosis and orientation of the bifid condyle. The difference in 1 year postoperative mouth opening was analyzed using paired t test and the effectiveness of the technique using one way ANOVA. $P$-value of $< 0.05$ was considered statistically significant.

## Results

The demography, etiology and clinical details of our cohort are summarized in Table 1. The male: female ratio in our center was 5:7. The mean age was 15 ± 8 (SD) years (range 3–29 years). All patients had the etiology and symptom onset in childhood. All of them had a restricted interincisal distance at maximal mouth opening with a mean of 0.8 ± 1.9 (SD) mm (range 0–5 mm). Clinical examination in these patients revealed varying degrees of restricted mouth opening, and deviation and retraction of the chin. The pattern of ankylosis and morphology of bifid condyles detected on CT are tabulated in Table 2. The bilateral to unilateral cases was 1:4 and all present cases were oriented ML. Of the four patients had bilateral TMJA, one case had unilateral left side deviation of chin. All the present cases of BMC exhibited mushroom shaped BMC. CT scan in Figs 1 and 2 depicts our cases.

The demography, etiology and clinical details of all Indian BMC with TMJA are summarized in Table 1.

*Table 1.* Demography, etiology and clinical details of all Indian studies with our cohort

| | Rehman et al. (6) ($n$ = 10) | Gulati et al. (7) ($n$ = 2) | This study Cohort, 2009 ($n$ = 12) | Overall Study, 2009 ($n$ = 24) |
|---|---|---|---|---|
| Age | | | | |
| Mean | 17.9 | 23.5 | 15 | 16.92 |
| SD | 14 | 12.02 | 8.32 | 11.05 |
| Minimum | 5 | 15 | 3 | 3 |
| Maximum | 32 | 44 | 29 | 44 |
| Gender | | | | |
| Male | 5 (50) | 2 (100) | 5 (41.7) | 12 (50) |
| Female | 5 (50) | – | 7 (58.3) | 12 (50) |
| Male:female | 1:1 | 2:0 | 5:7 | 1:1 |
| Etiology | | | | |
| Trauma | 9 (90) | 1 (50) | 12 (100) | 22 (91.7) |
| Infection | 1 (10) | | – | 1 (4.2) |
| Unknown | – | 1 (50) | – | 1 (4.2) |
| Presenting complaint | | | | |
| Restricted mouth opening | 10 (100) | 2 (100) | 12 (100) | 24 (100) |
| Obstructive sleep apnea | 1 (10) | – | | 1 (4.2) |
| Presence of chin deviation | | | | |
| Ipsilateral side | 8 (80) | – | 8 (66.7) | 16 (66.67) |
| Bilateral | 1 (10) | – | 1 (8.3) | 2 (8.3) |
| None | 1 (10) | – | 3 (25) | 4 (16.7) |
| Unknown | – | 2 (100) | – | 2 (8.3) |
| Inter-incisor distance (in mm) | | | | |
| Mean | 10 | * | 0.83 | 3.89 |
| SD | 4.86 | * | 1.95 | 5.4 |
| Minimum | 5 | * | 0 | 0 |
| Maximum | 18 | * | 5 | 18 |

*Data not available. Values in parentheses are expressed as percentages.

*Table 2.* The pattern of ankylosis and morphology of bifid condyles detected on CT from our cohort and Indian studies

| | Rehman et al. (6) ($n$ = 10) | Gulati et al. (7) ($n$ = 2) | This study Cohort, 2009 ($n$ = 12) | Overall Study, 2009 ($n$ = 24) |
|---|---|---|---|---|
| Bifid mandibular condyle | | | | |
| Bilateral | 4 (40) | – | 3 (25) | 7 (29.2) |
| Unilateral – left | 2 (20) | 1 (50) | 8 (66.7) | 11 (45.8) |
| Unilateral – right | 2 (20) | 1 (50) | 1 (8.3) | 6 (25) |
| Condylar orientation | | | | |
| Mesiolateral | 8 (80) | 1 (50) | 12 (100) | 21 (87.5) |
| Anteroposterior | 2 (20) | 1 (50) | – | 3 (12.5) |
| TMJ ankylosis | | | | |
| Bilateral | 1 (10) | – | 4 (33.33) | 5 (20.8) |
| Unilateral – left | 4 (40) | 1 (50) | 7 (58.3) | 12 (50) |
| Unilateral – right | 5 (50) | 1 (50) | 1 (8.3) | 7 (29.2) |
| Ankylosed head | | | | |
| Anterior | 2 (20) | – | – | 2 (8.3) |
| Lateral | 7 (70) | 1 (50) | 7 (58.3) | 15 (62.5) |
| Mesial | 1 (10) | – | 5 (41.7) | 6 (25) |
| Not known | – | 1 (50) | – | 1 (4.2) |

Values in parentheses are expressed as percentages.

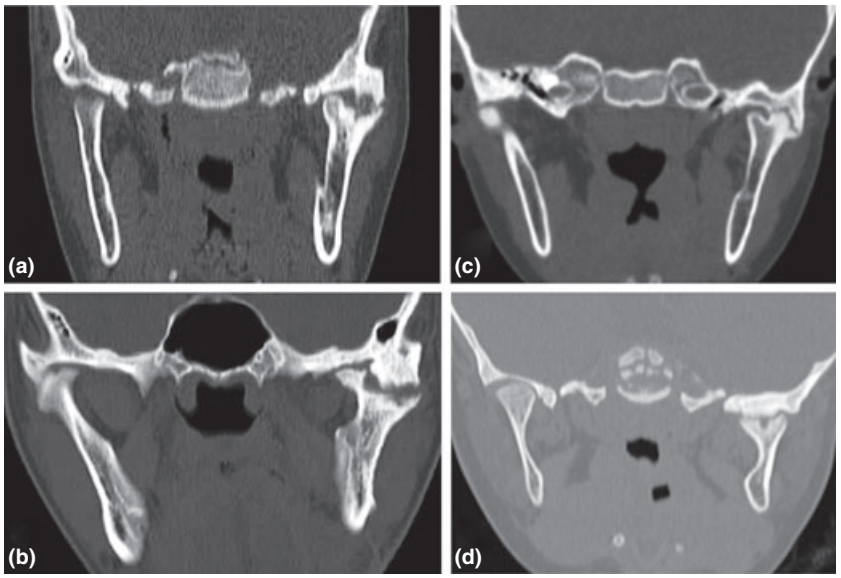


*Fig. 1.* CT of present case series. A, C: CT of cases with left side TMJ ankylosis and left BMC oriented mesio-distally with lateral head ankylosed. B, CT of a case with bilateral TMJ ankylosis and right BMC oriented mesio-distally with lateral head ankylosed. D, CT of a case with left side TMJ ankylosis and left BMC oriented Mesio-laterally with medial head ankylosed (CT, Computed Tomogram; TMJ, Tempromandibular Joint; BMC, Bifid Mandibular Condyle).

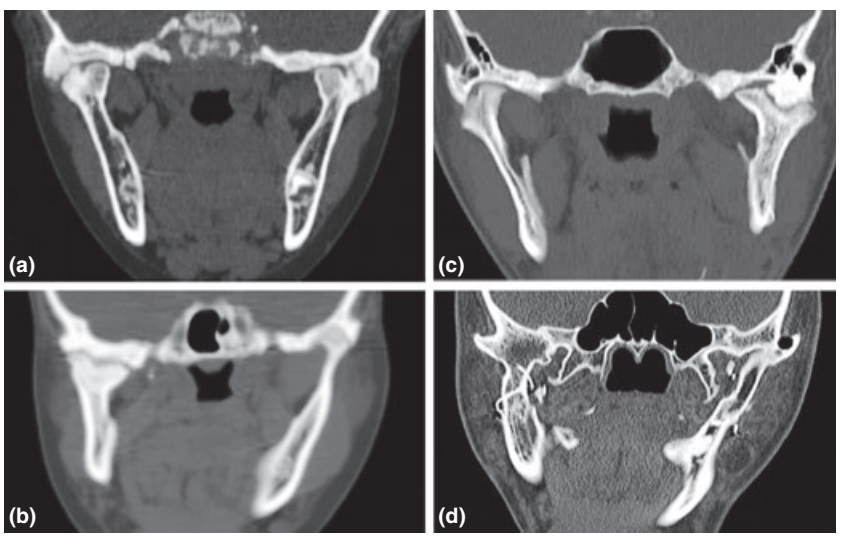


*Fig. 2.* CT of present case series. A, B: CT of cases with bilateral TMJ ankylosis and bilateral BMC oriented mesio-laterally with lateral head ankylosed. C, D: CT of cases with bilateral TMJ ankylosis and left BMC oriented mesio-laterally with lateral head ankylosed. (CT, Computed Tomogram; TMJ, Tempromandibular Joint; BMC, Bifid Mandibular Condyle).

The male: female ratio was 1:1. History of trauma was reported in 91.7% of cases and all patients had the etiology and symptom onset in childhood. Clinical examination in these patients revealed varying degrees of restricted mouth opening, and deviation and retraction of the chin. The mean age of reporting was 16.92 ± 11.05 years with a range of 3–44 years. Chin deviation to ipsilateral direction of ankylosis was seen in two-third of cases while 16.7% of cases had no deviation of chin visibly. Mean inter-incisor mouth opening was 3.89 ± 5.4 mm. The pattern of ankylosis and morphology of bifid condyles detected on CT are tabulated in Table 2. The salient features are summarized as follows. Bilateral to unilateral and ML to AP BMC patient ratios were 7:17 and 7:1, respectively. Of the seven cases with bilateral BMC, four patients had TMJA. Of these four cases, three were oriented ML and one AP. Of these cases, four were ankylosed laterally, one medially and two anteriorly. In the remaining three cases with bilateral BMC and unilateral TMJA, two condylar heads were oriented laterally and one anteriorly. The left:right side was 2:1. Only one case of bilateral TMJA had a unilateral BMC from the present case series. In the reported postinfectious case, the medial head was ankylosed to the temporal bone while the lateral head articulated with a facet in the temporal bone.

Tables 3 and 4 refers to the one way ANOVA with respect to age (in years) of reporting with Bifid Mandibular condyle and TMJ ankylosis and interincisor opening (in mm; MO) respectively compared with gender, etiology, center, site of bifid and ankylosis and orientation of the bifid condyle. Age has no significant relationship whereas for MO there was a statistical significance between gender and centers.

These cases were treated by two modalities – gap arthroplasty (GA) with either costochondral graft (CC) or temporalis flap. The developing TMJs were treated with CC whereas the developed TMJ was treated with temporalis flap. There were five cases (41.7%) treated with GA with CC graft and seven cases (58.3%) treated

*Table 3*. One-way ANOVA for age (in years) of reporting with Bifid Mandibular condyle and TMJ ankylosis compared with gender, etiology, center, site of bifid and ankylosis and orientation of the bifid condyle

| | *n* | Mean | SD | 95% Confidence interval for mean | | *P*-value |
|---|---|---|---|---|---|---|
| | | | | Lower bound | Upper bound | |
| Gender | | | | | | |
| Male | 12 | 21.00 | 11.11 | 13.94 | 28.06 | 0.07 |
| Female | 12 | 12.83 | 9.78 | 6.62 | 19.05 | |
| Etiology | | | | | | |
| Trauma | 22 | 16.86 | 11.54 | 11.75 | 21.98 | 0.95 |
| Infection | 1 | 20.00 | – | – | – | |
| Unknown | 1 | 15.00 | – | – | – | |
| Center | | | | | | |
| Rehman et al. (6) | 10 | 17.90 | 14.00 | 7.89 | 27.91 | 0.58 |
| Gulati et al. (7) | 2 | 23.50 | 12.02 | −84.50 | 131.50 | |
| Present study | 12 | 15.00 | 8.32 | 9.71 | 20.29 | |
| BMC | | | | | | |
| Bilateral | 7 | 13.57 | 8.52 | 5.69 | 21.45 | 0.35 |
| Unilateral | 17 | 18.29 | 11.89 | 12.18 | 24.41 | |
| TMJA | | | | | | |
| Bilateral | 5 | 16.8 | 9.73 | 4.717 | 28.88 | 0.98 |
| Unilateral | 19 | 16.95 | 11.62 | 11.35 | 22.55 | |
| Orientation of BMC | | | | | | |
| AP | 3 | 15.67 | 14.22 | −19.67 | 51.00 | 0.84 |
| ML | 21 | 17.1 | 10.95 | 12.11 | 22.08 | |

*Table 4*. One-way ANOVA for mouth opening (in mm) of reporting with Bifid Mandibular condyle and TMJ ankylosis compared with gender, etiology, center, site of bifid and ankylosis and orientation of the bifid condyle

| | *n* | Mean | SD | 95% Confidence interval for mean | | *P*-value |
|---|---|---|---|---|---|---|
| | | | | Lower bound | Upper bound | |
| Gender | | | | | | |
| Male | 9 | 6.67 | 6.30 | 1.82 | 11.51 | 0.02 |
| Female | 9 | 1.11 | 2.20 | −0.58 | 2.81 | |
| Etiology | | | | | | |
| Trauma | 17 | 3.41 | 5.16 | 0.76 | 6.07 | 0.13 |
| Infection | 1 | 12.00 | | | | |
| Unknown | | | | | | |
| Center | | | | | | |
| Rehman et al. (6) | 6 | 10.00 | 4.86 | 4.90 | 15.10 | 0.00 |
| Gulati et al. (7) | Data missing | | | | | |
| Present study | 12 | 0.83 | 1.95 | −0.40 | 2.07 | |
| BMC | | | | | | |
| Bilateral | 5 | 4.60 | 7.80 | −5.08 | 14.28 | 0.74 |
| Unilateral | 13 | 3.62 | 4.56 | 0.86 | 6.37 | |
| TMJ A | | | | | | |
| Bilateral | 4 | 0.00 | 0.00 | 0.00 | 0.00 | 0.10 |
| Unilateral | 14 | 5.00 | 5.67 | 1.73 | 8.27 | |
| Orientation of BMC | | | | | | |
| AP | 1 | 5.00 | | | | 0.84 |
| ML | 17 | 3.82 | 5.56 | 0.97 | 6.68 | |

with GA with temporalis flap. The 1 year follow-up revealed a mean MO in 28.58 ± 7.44 mm. The mean gain in MO, postoperatively after a year was 27.75 ± 8.38 mm.

The paired t test between preoperative and 1 year postoperative MO revealed that the mean difference between MO was 27.8 ± 8.38 mm (95% confidence interval of the mean difference −22.43 to 33.07, $P = 0.000$). The GA with CC graft exhibited a mean 1 year postoperative MO of 19.8 ± 5.31 mm while the GA with temporalis showed a mean MO of 33.43 ± 4.43 mm. The difference was statistically significant ($P = 0.001$) (Table 5).

## Discussion

Bifid mandibular condyles (BMC) are now being more frequently discovered and described with the advanced imaging and reconstructing techniques in CT and MRI. Rehman TA (6) in 2009 has reported of 67 cases living with BMC. On combining our 12 cases, this number goes to 79 cases. The exact etiology of BMC is still not

*Table 5*. One-way ANOVA depicting the difference in 1 year postoperative mean mouth opening between the two treatment methods

| | *n* | Mean | SD | 95% Confidence interval for mean | | *P*-value |
|---|---|---|---|---|---|---|
| | | | | Lower bound | Upper bound | |
| GA + CC | 5 | 19.80 | 5.31 | 13.21 | 26.39 | 0.001 |
| GA + temporalis | 7 | 33.43 | 4.43 | 29.33 | 37.53 | |

GA, Gap Arthroplasty; CC, Costochondral graft

*Table 6*. Glimpse of other studies

| Study Ref | Age | Gender | Etiology | BMC | TMJA |
|---|---|---|---|---|---|
| 2 | 14 | Male | Trauma | Left | Left |
| 3 | 40+ | Male | Trauma | Left | Bilateral |
| 4 | 32 | Male | Trauma | Right | Right |
| 5 | 9 | Female | Not mentioned | Left | Right |

elucidated. Possible etiopathological agents include developmental, traumatic, vascular, abnormal muscle pulling, nutritional, endocrinal, teratogenic and infectious agents (6). Blackwood (8) and Moffett (9) examined human fetuses postulated the developmental causes of BMC in TMJs. They suggested that persistence of a vascularized fibrous septum in the condylar cartilage or rupture of some of the blood vessels in the septum in some part of life during development can lead to impairment of the condylar ossification and lead bifidity of the condyle. The studies of Poswillo and Walker on rhesus monkeys supported trauma as the etiology for BMC (6). Gundlach et al. experiments on rats with teratogenic agents points BMC as a form of embryopathy resulted by a effect of a teratogenic agent and misdirection of muscle fibres influencing condylar bone formation (6).

The morphology of BMC varies from a shallow groove to two distinct condylar heads with separate necks. The orientation of the two heads may be in the mediolateral or anteroposterior directions. Szentpetery et al. postulated that AP BMC are likely to be post-traumatic whereas ML BMC could be developmental whereas studies by Wang et al. and Yao et al., had demonstrated that a fracture of the mandibular condyle could result in ML as well as AP BMC (1, 6).

Among the reported cases of BMC, only fourteen patients had associated TMJA (6). The first report of BMC with TMJA was by Stadnicki (2) in 1971 and subsequently by To (3), Daniels and Ali (4) and Sales MAO (5). The largest case series was published by Rehman TA et al. (6) consisting of 10 cases and last report was of two such cases by Gulati A et al. (7) The details of first four cases are given in Table 6.

Reported cases of BMC and TMJA in the Indian studies are summarized in Tables 1 and 2. The prevalence of TMJA and BMC in lower age at the present patient cohort ($n = 12$) could be due to referral bias – the increased number of cases of TMJA reported to this exclusive craniofacial center. The higher female incidence in this facility further indicates that females seek more and early treatment. The lower MO, further emphasis these findings. The significant *P*-values in Table 4 proceeds to emphasis this view.

As CT and MRI are evolving as the mainstay modalities in the evaluation of TMJA, non-inclusion of other plain radiographs was beyond clinical applications. However, this has been accepted as a standard method of practice and has been used before (6).

The CT studies were analyzed after patients had already developed TMJA. The first report of a 3-year-old male patient with evidence of TMJA at 14 years (2), still remains the evidence of the progression. The present cohort also shows three cases below the age of 6 years. This report probably reports the youngest person with BMC and TMJ ankylosis in a 3-year-old female with unilateral BMC and TMJA in the same side. However, these bifid condyles were discovered after the development of ankylosis. Hence, it is still not possible to conclude whether the bifid condyles were present in these patients before TMJA or how the presence of bifidity contributed to development of TMJA.

These cases were treated by two accepted modalities of treatment of TMJA – GA with CC graft or temporalis flap. The choice rested on the age and development of TMJ in the individual. Though each technique has their own advantage and disadvantage, the significant mean difference between MO was $27.8 \pm 8.38$ mm ($P = 0.000$) indicates that GA with temporalis is better technique. As a child with TMJA often does not comply with much needed physiotherapy or co-operate during clinical examination, a lower MO in these cases is expected.

As this study is the first of its kind to compare the results of surgical correction of BMC and TMJA, the results of these surgeries would not be compared to any existing literature. In conclusion, the current series helps to elucidate the various patterns of TMJA and the CT morphology of bifid condyles in cases of BMC with associated TMJA in Indian population. Further studies with imaging documented performed at earlier stages of trauma and subsequent development of the BMC could compare the relative evolution of the double heads and ankylosis. Such studies would shed more light on the role of bifidity of the condyle in progressing into TMJ ankylosis. This case series would also reveal the effectiveness of treatment of such as gap arthroplasty with either costochondral graft or temporalis flap.

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