“Temporomandibular hypermobility as a sequelae of generalized joint hypermobility in temporomandibular disorders: a pilot study”

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**Abstract**

**Background.** Various studies have found increased association between occurrence of Generalized joint hypermobility (GJH) and signs and symptoms of temporomandibular disorders (TMDs), suggesting that the former may represent a risk factor in the development of TMDs. The purpose of this study is to investigate the correlation between temporomandibular joint (TMJ) hypermobility and GJH, clinically as well as radiographically.

**Materials and methods.** 60 participants, between age range of 18-35 years, equally divided into study and control groups, were evaluated for the presence of TMDs based on the RDC/TMD criteria. Radiographic examination was done using panoramic view, TMJ open and close tomographic views. Systemic joint hypermobility was assessed by using Beighton criteria.

**Results:** Based on the Beighton’s score for GJH, 53.3% of study patients were distinctly hypermobile, 20% were moderately hypermobile and 26.7% had no hypermobility. In the control group, only 3.3% exhibited distinct hypermobility, 10% had moderate hypermobility and 86.7% showed no hypermobility. There was statistically significant correlation (p=0.001) between the TMD group and individuals with severe GJH (high Beighton score). 73.3% of the study participants demonstrated radiographically hypermobile TMJ as well as GJH. The results of spearman correlation indicated a high correlation between radiographically hypermobile TMJ and GJH.

**Introduction:**- Temporomandibular disorders (TMDs) are a heterogeneous group of craniomandibular disorders involving the joint, musculature and associated structures (Okeson, 2005 and Romero-Reyes, 2014), and characterized by a triad of clinical signs: joint or/muscle pain, restricted or altered path of mandibular motion and joint sounds (Manfredini, 2011; Guarda-Nardini, 2012; Singh, 2014). Other symptoms may include headache, dizziness, earache, toothache and neuralgia (Guarda-Nardini, 2012). TMDs cannot be attributed to a single cause. Although, both pathophysiological (musculoskeletal injuries, malocclusion, loss of teeth, connective tissue laxity and systemic diseases) and psychosocial factors (emotional stress, depression, anxiety and sleep disturbances) (Wieckiewicz, 2014 and Perrini, 1997) have been proposed, the exact etiology of this intricate entity remains un-deciphered (Greenberg, 2008).
Generalized joint hypermobility (GJH) syndrome was first described by Kirk et al, in 1967, as laxity of joints associated with musculoskeletal complaints excluding any defined rheumatological disorders (Kirk, 1967; Adair, 1993; Westling, 1992). The syndrome does not include inherited connective tissue or bone disorders, such as Ehlers-Danlos syndrome, Marfan syndrome, osteogenesis imperfecta, and others (Adair, 1993). Range of motion of joints is controlled by numerous factors, including biochemical structural changes of collagen and elastin, causing a loss of resistance to traction, laxity and increased joint mobility (Pasinato, 2011). Various studies have found increased association between occurrence of GJH and signs and symptoms of TMJ disorders, suggesting that the former may represent a risk factor in the development of TMDs (Perrini, 1993; Adair, 1993; Westling, 1992). The present study was designed to investigate the correlation between TMJ hypermobility and GJH clinically as well as radiographically, and to assess symptoms of TMDs independently.

Material and methods:-
A total of 60 participants, between age range of 18-35 years, equally divided into study and control groups, were evaluated in the current study. Individuals with symptoms of TMJ dysfunction and who had undergone treatment for the same previously were included in the study group while the individuals with no symptoms and prior history of any treatment for TMD formed the control group. Individuals with known systemic bone diseases, any syndrome, history of trauma to maxillo-facial region and those undergoing steroid treatment were excluded from the investigation. Athletes, gymnasts and dancers were also excluded.

The study protocol was reviewed and approved by the Institutional ethics committee of People’s University, Bhopal and written consent was taken from all the participants. All the participants were thoroughly examined and brief history was taken. A questionnaire was used for documenting the absence or presence of pain in jaws, TMJ noise, altered jaw movements and position based on RDC/TMD, which is standard for the systematized diagnosis of TMDs. Panoramic radiographs were made to evaluate the joint morphology and gross bony changes in the condyle. TMJ open and close tomographic views were made to assess the relationship of the condyle with the fossa in open and close mouth position. Systemic joint hypermobility was assessed by using Beighton joint hypermobility test (score 0 to 9) (figures 1 and 2) (Beighton, 1973). Data was subjected to Chi-square test to compare the frequencies between the groups, Mann-Whitney (U Test) test to compare the mandibular range of motion between the groups, One-way (ANOVA) test to find significant differences between groups and Fisher’s Exact and the Spearman tests to correlate mandibular movement, range of motion and GJH scores.

Results:-
Of the total 60 participants, 30 each in the study and control group, 14 (46.7%) were males and 16 (53.3%) females in the former group while the latter group comprised of 2:1 male-to-female ratio. Difference between the two groups, based on gender, was statistically non-significant (p=0.100). Mean age in the TMD group was 30.5 years and that in the control group was 29.8 years. The distribution of signs and symptoms, among the study and control groups, is shown in figure 3. 80% (n=24) of study subjects and none of the controls elicited pain, which was extremely statistically significant (p=0.001). Joint sounds were also found to be highly statistically significant when compared between the two groups.

The mean value of active and passive range of mandibular opening among TMD patients was 49.95±2.35 and 52.25±2.84. In the control group, the active and passive mouth opening was found to be 43.55±0.83 and 46.25±0.99, which was highly statistically significant (p<0.001) when compared with the study group (table 1, figure 4). On categorizing the participants on the basis of Beighton’s score for GJH, 53.3% (n=16) study patients were distinctly hypermobile, 20% (n=6) were moderately hypermobile and 26.7% (n=8) had no hypermobility. In the control group, only 3.3% (n=1) exhibited distinct hypermobility, 10% (n=3) had moderate hypermobility and 86.7% (n=26) showed no hypermobility (table 2, figure 5). There was statistically significant correlation (p=0.001) between the TMD group and individuals with severe GJH (high Beighton score).

Clinical assessment yielded 27 (90%) patients with hypermobile temporomandibular joint in the study group and none in the control group, while radiographic examination (figure 6) (condylar position beyond the articular eminence on maximum mouth opening in TMJ tomographic open view) showed that 83.3% (n=25) patients in the study group and 3.3% (n=10) controls had TMJ hypermobility. The result when compared was found to be significant (p=0.04). 73.3% (n=22) of the study participants demonstrated radiographically hypermobile TMJ as well as GJH. The results of spearman correlation indicated a high correlation between radiographically hypermobile TMJ and GJH (table 3, figure 7).
Discussion:
Temporal mandibular disorders are the musculoskeletal conditions known for their chronicity. Previous literature has been suggestive of higher risk of TMDs in females (Wieckiewicz, 2014; Ebrahimi, 2011; Casanova-Rosado, 2006; Oliveira, 2006; Bagis, 2012; Chauhan, 2013; Alnesary, 2012; Minghelli, 2014; Tecco, 2011; Machado 2009). The present study also exhibited the same. Factors responsible for such predilection include hormonal influences, stoic nature of males and higher pain sensitivity in females (Wieckiewicz, 2014; Chalkoo, 2014; Kishimoto, 2007; Warren, 2001; Wang, 2008; Nekora-Azak, 2004; Macfarlane, 2002).

Pain and joint sounds as the most common manifestations of TMJ dysfunction were revealed in the current study. This is in agreement with studies by Ryalat et al and Cooper and Kleinberg who found pain in 45.6% and 96.1% in the study population, respectively (Ryalat, 2009 and Cooper, 2007). Prevalence of TMJ sounds reported by Bagis et al was 39.6% (Bagis, 2012). Elfving et al recorded joint sounds in 56% of the TMD patients and 36% of the matched controls (Elfving, 2002). Temporomandibular joint sounds may relate to a pathological condition (disc derangement, muscular incoordination or arthritic changes) or may be a variation of the normal anatomy (Westling, 1992 and Elfving, 2002). Joint noises represent alteration in the regular smooth movement of the disc and condyle, predisposing the joint to inflammation and subsequently pain.

Higher range of mandibular motion in patients with high GJH scores was found, which was comparable to the findings of Pasinato et al and Hirsch et al (Pasinato, 2011 and Hirsch, 2008). Kalaykova et al found condylar position beyond the articular eminence in TMJ hypermobility patients which is in corroborate with our study (Kalaykova, 2006). Haghigaht et al in his study observed condylar position beyond the articular eminence on maximum mouth opening in TMJ hypermobility cases (Haghigaht, 2014).

Numerous studies have analyzed association between GJH and dysfunctions of the temporomandibular joint, and have found a higher incidence of GJH among individuals with signs and symptoms of TMD (Pasinato, 2011 and Hirsch, 2008). Likewise, the current investigation established similar result. Westling et al found that joint laxity accounts for 3% to 25% of the TMD population (Westling, 1992). The hypothesis of a causal relationship between the two entities, especially regarding the displacement of the articular disc, seems to be biologically possible because constitutional hypermobility can affect all joints, including the TMJ (Hirsch, 2008).

Figure 1. Apposition of the thumb to the flexor aspect of the forearm.
Figure 2. Forward flexion with the hands flat on floor and knees extended.

Figure 3. Distribution of signs and symptoms of TMDs among study and control groups

Table 1. Level of range of mandibular opening

| Mandibular ROM | Study group | Control group | U Test |
|---------------|-------------|---------------|-------|
|               | Mean        | SD            | Mean  | SD     | p   |
| Active opening| 49.95       | 2.35          | 43.55 | 0.83   | 0.00|
| Passive opening| 52.55     | 2.84          | 46.25 | 0.99   | 0.00|
Table 2. Generalized joint hypermobility on the basis of Beighton score between study and control group

|                | Study group N (%) | Control group N (%) | P value |
|----------------|-------------------|---------------------|---------|
| No GJH         | 08 (26.7)         | 26 (86.7)           | 0.012   |
| Moderate GJH   | 06 (20)           | 03 (10)             | 0.35    |
| Severe GJH     | 16 (53.3)         | 01 (3.3)            | 0.001   |
Figure 6. TMJ open and close tomographic view showing the relationship of the condyle with the glenoid fossa in open and close mouth position.

Table 3. Spearman Correlation between radiographically hypermobile TMJ and GJH among study and control group

|                  | TMJ Hyper mobility (Tomographic view) | GJH |
|------------------|--------------------------------------|-----|
| Study group      | 25                                    | 22  |
| Control group    | 10                                    | 04  |

Figure 7. Spearman Correlation between radiographically hypermobile TMJ and GJH among study and control group

The authors declare that there is no conflict of interests regarding the publication of this paper.
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