Joint mobility and motor development

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SUMMARY   The association of joint hypermobility and motor development was sequentially investigated in 715 infants from the ages of 8 to 14 months. Seven joints were evaluated for mobility, and each infant underwent a physical and neurological examination. Parents were given a Denver Developmental Parents' Questionnaire. All subjects with a general developmental delay, systemic illness or syndrome were excluded. The infants were classified as having normal or delayed motor development with normal or delayed joint mobility. They were re-examined six months later. Multivariate statistical techniques was used for categorical analysis, and three joints were found to be significantly associated with motor delay at the first examination—hip abduction, elbow hyperextension, and foot dorsiflexion. Of the 715 infants, 126 had joint hypermobility and of these 38 (30·2%) had motor delay. Sixty four of 589 (10·9%) with normal joints had delayed motor development. Six months later 23 out of 35 of the group with joint hypermobility and 42 out of 53 of the group with normal joints had normal motor function.

   Joint hypermobility is associated with an increased incidence of motor delay in infancy. Over the ensuing six months most of the subjects will catch up. These findings, indicating a favourable prognosis, have implications regarding clinical assessment and parental counselling.

Paediatricians frequently encounter infants presenting with delayed gross motor development. The evaluation of such cases entails comprehensive paediatric and neurodevelopmental examination, and when cerebral palsy, mental retardation, and other neuromuscular aetiologies have been excluded, a group of slow motor developers of unknown aetiology and prognosis remains. The rather dramatic presentation of an infant who does not attain sitting, crawling, or walking at the same time as his peers often engenders anxiety in the parents. It has been our experience that a number of these infants have hypermobile joints and that they ultimately seem to catch up in motor development.

   Previous reports suggest that hypermobile joints can be the result of either increased laxity of the ligaments and joint capsule[1] or muscle hypotonia.[2] Owing to the inherent difficulties of clinically quantifying degrees of muscle hypotonia we studied the problem by assessing joint mobility and did not attempt to differentiate between the two possible causes. Hypotonia associated with slow motor development has been described,[2][3] and one study has investigated the association of joint hypermobility and motor development.[4] No prospective, controlled blind studies, however, have been performed.

   The two main purposes of this study were as follows: (a) To investigate whether hypermobile joints were in fact associated with an increased incidence of motor delay, and, if so, whether specific joints were more often affected. (b) In the event of a positive association, to study these infants prospectively in order to elucidate their subsequent motor development and joint mobility as compared with controls.

Patients and methods

Altogether 729 infants from the ages of 8 to 14 months were recruited from 10 well baby clinics and seven day care centres in the Haifa area. Staff at the well baby clinics had previously screened the infants and infants with the following conditions were excluded from the cohort: those with acute or chronic illnesses; those who had recently been in hospital; those suffering from diagnosed neurodevelopmental delay or metabolic disorders; those who had had low birth weight including preterm infants; and those having defined syndromes. An additional

12 infants were excluded by the examining paediatrician: three because of cerebral palsy, six because of general developmental delay, two because of significant cardiac abnormality associated with congestive cardiac failure, and one because of thalassaemia major. The original study sample comprised 717 subjects of who 365 were boys and 352 girls. This study population presented a cross section of the ethnic and socioeconomic characteristics of the Haifa community.

The study consisted of two stages:

Stage I. (1) Information concerning the developmental state of each infant was obtained from the records of the well baby clinic where the results of periodic standard assessments by the nurses using the Stycar Sequence to ascertain the infant's development state were rated.[5] (2) The Denver Developmental Parents' Questionnaire was completed by the parents.[6] This questionnaire is based on the Denver Development Screening Test (DDST), which unlike the above procedure has been standardised for the Israeli population and was used to determine the present motor developmental state of the child.[7] It was completed before the joint mobility assessment. For the purposes of this study motor delay was defined as a failure to attain the motor skills achieved by 75% of the Israeli population of the same age.[7] (3) A paediatrician performed a complete physical and neurological examination, with particular emphasis on the assessment of muscle strength and the presence of normal tendon reflexes. Clinical evidence of connective tissue disorders was also sought. (4) The mobility of seven joints was assessed, five of which were those investigated in other studies (table 1).[1] Hip abduction and shoulder adduction (scarf sign) were added. The paediatrician performing the joint assessment was unaware of the motor developmental state of the infant as reported by the parents and staff of the well baby clinic. (5) Information on demographic data, family history of joint hypermobility, rheumatic diseases, and motor development was obtained

Table 1 *Assessment of joint laxity*

1	Thumb—passive apposition to the flexor aspect of the forearm
2	Fingers—passive hyperextension so that they lie parallel with the extensor aspect of the forearm
3	Elbow—passive hyperextension to 10° or more
4	Knee—passive hyperextension to 10° or more
5	Ankle—passive dorsiflexion of foot score (1) for proximal contact and score (2) for complete contact with tibial volar aspect
6	Shoulder—passive adduction with elbow crossing chest midline (scarf sign)
7	Hip—passive abduction to 180°

by a questionnaire designed for the purpose of this study. The information was gathered from the parents who were unaware of the joint assessment findings in their offspring.

Interobserver reliability for the joint mobility assessment between the study paediatrician and another paediatrician and reproducibility were established before the initiation of the study. Thirty infants of the same age range as the study population were examined, with a resulting reliability of 90% and reproducibility of 96%. This procedure was repeated during the study on a random sample of 40 infants from the total cohort, and resulted in a reliability of 0·86.

Stage II. Six months later a random representative sample of the cohort was re-examined for joint mobility, and motor achievement was assessed by the DDST.[7]

Statistical analysis. As all joint mobility variables are categorical (most of them dichotomous and one trichotomous), a classical linear discriminant analysis could not be applied. A regression model was therefore used in which the dependent variable was the log odds ratio on the gross motor development variable. The independent variables were the joints included in the assessment, which were modelled as factors.[8] The statistical package SAS (*SAS Users guide statistics 1982*) was used for computer analysis purposes. The FUNCAT procedure was utilised to model the functions of categorical variables.

Results

Out of the 715 subjects on which complete data was available 525 (73·4%) had normal joints and normal motor development, and 102 (14·3%) with and without hypermobility presented gross motor delay. Eighty eight infants with hypermobile joints had normal gross motor development. To explain the delay various combinations of main effects and interaction were tried from the seven joint mobility variables. In the final model each of the included effects were highly significant. The three joints that best correlated with gross motor developmental delay at stage I were: (a) foot dorsiflexion ($\chi^2=13·38$, p=0·003); (b) hip abduction ($\chi^2=0·53$, p=0·002); (c) elbow extension ($\chi^2=11·25$, p=0·008). If all of the three joints were hypermobile the odds of developmental motor delay were 10·11 (that is, 10 times more than the odds from motor delay in the total cohort). If none of the three joints were affected the odds were 0·43 (compared with the total cohort). The relative odds of developmental motor delay were therefore 23 (10·11/0·43).

One hundred and twenty six subjects showed hypermobility in one or more of these three joints in various combinations. Of these 38 (30·2%) presented gross motor delay as opposed to 64 (10·9%) if none of the three joints were hypermobile (table 2).

Six months later (stage II) a stratified random sample of the cohort was re-examined. Four diagnostic categories (see table 3) were selected as follows: (a) gross motor delay and hypermobility— 35 infants (92% of the original group); (b) gross motor delay and normal joints—53 (83% of the original group); (c) normal motor development and normal joints—90 randomly selected (every fifth infant from each clinic) from the 525 subjects who comprised the original group; (d) normal motor development and hypermobile joints—25 randomly selected (every third infant from each clinic) out of the 88 who comprised the original group. Four subjects and two subjects from groups (c) and (d) respectively were excluded from the analysis because of missing data.

Out of the 58 infants with joint hypermobility (comprising both normal and delayed motor development), 44 (75·9%) lost their hypermobility. Out of the 35 who had hypermobility and delayed development, 24 (68·6%) infants lost their hypermobility and 20 (83·3%) of these caught up in motor development. Out of all the 11 whose joints remained hypermobile only six (54·5%) caught up.

(This difference did not reach a significant level but may indicate a trend.)

Over the six month interval between stage I and II the prevalence of gross motor delay in the groups with hypermobile and normal joints remained constant (tables 2 and 3). Factors such as sex, ethnic origin, socioeconomic status, joint hypermobility, or rheumatic diseases in other family members were not found to be significantly associated with the hypermobility in the cohort subjects. Two significant factors associated with joint hypermobility were bottom shuffling and young age (table 4).

Although the reliability of parental recollections

Table 4 *The association of other factors with hypermobility and their significance*

Factor	χ^2	p Value
Age	26·5	0·0002
Sex	0·004	NS
Ethnic origin	1·85	NS
Socioeconimic status	6·69	NS
Bottom shuffling	6·98	0·0082
Family history		
Age of maternal walking	28·75	0·0071
Age of paternal walking	21·30	0·019
Age of siblings walking	18·36	0·049
Hypermobility in parents	0·73	NS
Hypermobility in siblings	0·12	NS
Rheumatic diseases	0·312	NS

Table 2 *Classification of subjects into four clinical categories at stage I*

	Group with hypermobile joints No (%)	Group without hypermobile joints No (%)	Total No (%)
Gross motor development			
Delayed	38 (30·2)	64 (10·9)	102 (14·3)
Normal	88 (69·8)	525 (89·1)	613 (85·7)
Total	126 (17·6)	589 (82·4)	715 (100)

Group with hypermobile joints *v* group without hypermobile joints: $\chi^2=31\cdot59$, p=0·0001.

Table 3 *Classification of subjects into four clinical categories at stage II*

	Group with hypermobile joints No (%)	Group without hypermobile joints No (%)	Total No (%)
Gross motor development			
Delayed	7 (30·4)	18 (10·3)	25 (12·7)
Normal	16 (69·6)	156 (89·7)	172 (87·3)
Total	23 (100)	174 (100)	197 (100)

Group with hypermobile joints *v* group without hypermobile joints: $\chi^2=7\cdot4$, p=0·0065.

was not established, 223 fathers (39 of the group with hypermobility and 184 in the normal group) and 308 mothers (76 in the group with hypermobility and 332 in the normal group) were able to supply information regarding the age they attained walking. A significant association with their children's joint hypermobility was shown (table 4). A significant association was found between the sibling's walking age and the subject's joint hypermobility (table 4).

Discussion

The association between hypermobile joints and motor development in infancy has not been comprehensively evaluated. It is complicated by the fact that norms for the very young age group do not exist, and studies on the subject use criteria derived from older children. In this investigation the problem was circumvented by using age controlled subjects.

The results of this study confirm that in infants from the ages of 8 to 14 months, hypermobile joints are associated with a significantly increased incidence of motor delay. Specific joints were more often associated with the delay, namely excessive foot dorsiflexion, hip abduction, and elbow hyperextension (see definition). Furthermore, these three joints showed a synergistic rather than a additive effect.

Over the ensuing six months, 44 out of 58 (75·9%) of the infants with hypermobile joints lost their excessive mobility, 20 out of 24 (83·3%) of the infants with delayed development whose joints had become normal had caught up in motor development, whereas only six out of 11 (54·5%) of the infants with persistent joint hypermobility had achieved normal motor function. While these data did not reach significance, it may suggest that a biological relation does exist between hypermobile joints and motor development, and longer follow up may well have shown a larger percentage of motor catch up.

The benign nature of the phenomenon of hypermobile joints and motor development was suggested in the only study specifically studying this problem.[4] This study found that a motor catch up occurred by the age of 3 years, while our investigation suggested that motor development became normal earlier.

Other investigations dealing with floppy infants and benign congenital hypotonia might have included a similar population to ours.[9][10] These studies, however, were differently designed in that they were descriptive, uncontrolled, and based on a broad clinical group who had been primarily referred for motor delay. The interesting association of bottom shuffling observed with hypotonia was also noted in our subjects.[3] Unlike other studies we were unable to show a significant association of joint mobility with variables such as sex and ethnic origin.[11] An increased incidence of rheumatic diseases in other family members was not found.[12]

The implications of these results for clinical practice are that infants with a developmental profile showing motor delay as a sole manifestation, normal muscle strength and tendon reflexes, and increased joint mobility do constitute a clinical entity. This entity is associated with a favourable prognosis. Medical and neurophysiological investigations are not indicated and with appropriate counselling much anxiety and expense can be spared. It must be stressed that general clinical examination to exclude systemic disease is essential.

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