

Benign joint hypermobility syndrome among children with inguinal hernia

Masoud Nazem, Peyman Mottaghi¹, Alireza Hoseini², Hesam-al-din Khodadadi²

Department of Surgery, ²General Physician, Al-Zahra Hospital, Isfahan University of Medical Sciences, ¹Department of Medicine, Noor Hospital, Isfahan University of Medical Sciences, Isfahan, Iran

Background: Benign joint hypermobility syndrome (BJHS) is a disorder due to laxity of supporting connective tissue of joints. Inguinal hernia is also proposed due to weak supporting tissue that may be a clinical presentation of a more widespread problem of connective tissue. **Materials and Methods:** In a cross-sectional study, prevalence of benign hypermobility joint syndrome (BHJS) was assessed among 100 children aged 2-12 year admitted with inguinal hernia during 2010-2011. **Results:** BJHS (Beighton score ≥ 4) were detected in most of children (92%) with inguinal hernias. **Conclusion:** BHJS amongst this population was substantially greater than reported prevalence in healthy children and due to subsequent clinical significances; it is worthy to screen such patients for BHJS.

Key words: Inguinal hernia, joint hypermobility, pediatric

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INTRODUCTION

Pediatric inguinal hernia is one of most common causes of referring to pediatric surgeons and is the most common type of hernia in children.^[1] There is always a risk for bowel incarceration and secondary complications such as atrophy of testicles; thus, early diagnosis of these hernias is important in the pediatric age group.

Benign hypermobility joint syndrome (BHJS) as described by Ansell and Kirk is a complex of musculoskeletal symptoms caused by having hypermobile joints, is a relatively common disease in children.^[2] Recent studies suggest that this clinical syndrome not only includes symptoms related to joints and musculoskeletal system, but also visceral symptoms. Patients with BHJS are more prone than others to suffer from visceral problems, which can be a result of weak supporting structures and tissues in the body.^[3] These problems include association between hiatal hernia and hypermobile joints have been previously reported.^[4]

The theory that the infirmity of collagen in ligaments and weakness of supporting tissues could be a factor for inguinal hernias,^[5] we decided to assess the prevalence of inguinal hernia and BHJS in a referral university hospital.

MATERIALS AND METHODS

This cross-sectional study was conducted from February 2010 to November 2011 in pediatric surgical wards of Isfahan University of Medical Sciences. One hundred children with diagnosis of inguinal hernia by pediatric surgeons, whom consecutively admitted in a university hospital, were enrolled in the study. The inclusion criteria were: children between 2 and 12 years old with inguinal hernia whom not affected by acquired rheumatologic and hereditary connective tissue diseases (Ehlers-Danlos syndrome and Marfan syndrome.) causing joint disorders with participation consent. Exclusion criteria were: patients with inherited metabolic diseases that lead to weakness of joints or severe physical disability and inguinal hernias resulting from intra-abdominal complications such as ascites, secondary to tumors, trauma or previous surgery in this area.

The children were evaluated for joint hypermobility by pediatricians and a trained general practitioner by using the Beighton scoring system to determine the presence of generalized joint hypermobility.^[6] The patient regarded to have hypermobility if both had to agree that the child is hypermobile according to score of $\geq 4/9$ for joint hypermobility. Standard tests of significance (Chi-square test, $P < 0.05$ -significant) were applied.

Address for correspondence: Address for correspondence: Dr. Peyman Mottaghi, Isfahan University of Medical Sciences, Noor Hospital, Isfahan, Iran. E mail: motaghi@med.mui.ac.ir

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RESULTS

In this study, average age of patients with inguinal hernia was 4.3 ± 2.6 years and 77% of children with inguinal hernia were male and 23% were female. BHJS was diagnosed in most of children with inguinal hernia (93%) and the average Beighton score in the involved patients was 6.2 ± 2 . In patients with inguinal hernia there were no significant differences in prevalence of BHJS between girls and boys ($P = 0.82$).

About half of involved children were younger than 4 years (49%) and half of them were 4 or more than 4 years (51%). Prevalence of benign joint hypermobility (Beighton score) in the groups of children with inguinal hernia showed in Table 1.

DISCUSSION

There are some reports that hernias in children may have a direct relationship with connective tissue disorders. [5-9] Valayannopoulos *et al.* reported that abnormalities of collagen in ligaments causing weakness of supportive tissues could be a risk-factor for the creation of inguinal hernias, theoretically.^[10]

In a study, about the relationship between BHJS in adult patients with inguinal hernia, Pans *et al.* reported that the prevalence of inguinal hernia was between 25% and 16% in patients with BHJS and the control group, respectively. This difference was not significant.^[5]

In another study in 2005 at Ankara University, Seçkin *et al.* investigated the prevalence of BHJS in high schools and concomitant diseases. No difference in the incidence of inguinal hernia was found in these patients.^[11]

Robinson *et al.* in a study in 2005 showed a high incidence of inguinal hernias in patients with Shprintzen-Goldberg syndrome (a heritable disorder of connective tissue) compare to the normal population.^[12]

Our results showed that the average Beighton Score in children with inguinal hernia is significantly higher than reported scores in studies about normal population. These data indicating that the prevalence of BHJS (Beighton Score ≥ 4) in children with inguinal hernia is significantly higher than normal children. Our results were similar with the study of pans *et al.* in adults. In this study, inguinal hernia had a higher incidence in patients with connective tissue

Table 1: Prevalence of joint hypermobility (Beighton score) in subgroups of patients depending on sex

Beighton score Groups	Cases* (%)
Boys	
<4	6 (7.8)
≥ 4	71 (92.2)
Girls	
<4	1 (4.3)
≥ 4	22 (95.7)

*Children with inguinal hernia

disease.^[5] These findings suggest that due to long-term complication of BHJS, it could be worthy to screen children with inguinal hernia for this syndrome.

Although few studies similar to our research has been carried out, but the conflicting results, which is inconsistent with the results of our study, suggests a need to conduct more studies with a larger population in different parts of the world with various races.

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