Familial back shape in adolescent scoliosis

A photogrammetric population study

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A stereophotogrammetric technique (ISIS scanning) was used to assess the back shape of children with adolescent idiopathic scoliosis (index cases), their unaffected relatives, normal adults and children, and children with small idiopathic scoliotic curves (Cobb's angle < 30°) to detect any familial trends that may predispose to scoliosis. The analysis revealed a characteristic three-dimensional scoliotic back shape; no differences in unaffected relatives as regards sex or a positive family history of scoliosis; similar back shapes in unaffected parents and normal adults, both of whom differ from index cases and children with small curves. Unaffected siblings had a sagittal profile between those of normal children and children with small curves. This may represent an inherited tendency to scoliosis.

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Wynne-Davies (1968) and Czeizel (1978) have noted that there appears to be a strong genetic susceptibility within families with scoliosis. By examining such families, it may be possible to identify factors responsible for the development or progression of the deformity.

Recent studies in the biomechanics of idiopathic scoliosis have emphasized the three-dimensional nature of the deformity—lateral curvature associated with rotation and lordosis, any of which may be relevant etiologically. Both Archer and Dickson (1985) and Smith and Dickson (1987) suggested that the genetic component predisposing to scoliosis may be a relative flattening of the lateral profile.

Traditional screening methods are based on a clinical evaluation—the forward bend test—to assess the presence of any trunk asymmetry caused by vertebral and rib rotation. It is unlikely that rather large curves would be missed in this way, but some rather small curves may escape detection. These curves could be detected radiographically if the attendant risk of radiation did not prohibit its use in screening. Surface topographic measurements—e.g., the automated stereophotogrammetric technique (ISIS; Turner-Smith 1988, Turner-Smith et al. 1988)—are not so restricted. In addition, ISIS provides a three-dimensional analysis of back shape.

In this study, ISIS scanning was used to assess the back shape of adolescent idiopathic scoliotic children, as well as that of their affected and unaffected relatives to detect any familial trends in back shape. ISIS measurements of normal back shape have previously been reported (Carr et al. 1991), and these data were used for comparison. If an abnormality of back shape were the underlying cause of scoliosis, then, it might be expected to be more pronounced in the families of children with idiopathic scoliosis.

Materials and methods

The ISIS system

The commercially available ISIS system comprises a scanner, special computer console, and plotter. The scanner contains a stationary television camera producing a plane of light that is deflected by a mirror for scanning down the patient's back. The line formed as the light falls on the skin surface is measured by the computer to acquire the three-dimensional back surface shape photogrammetrically. The patient stands against a bar at the level of the pelvis, and black self-adhesive markers are placed over eight to 10 spinous processes from the vertebra prominens downwards and over the posterior iliac spines. These indicate the line of the spinous processes, and permit adjustments to be made for the patient's posture. The scan takes approximately 1.5 seconds; and within a few minutes, the computed back-shape analysis is plotted for the physician. A detailed description of the technique and of the various parameters is given by Turner-Smith et al. (1988), and a typical scan is shown in Figure 1.

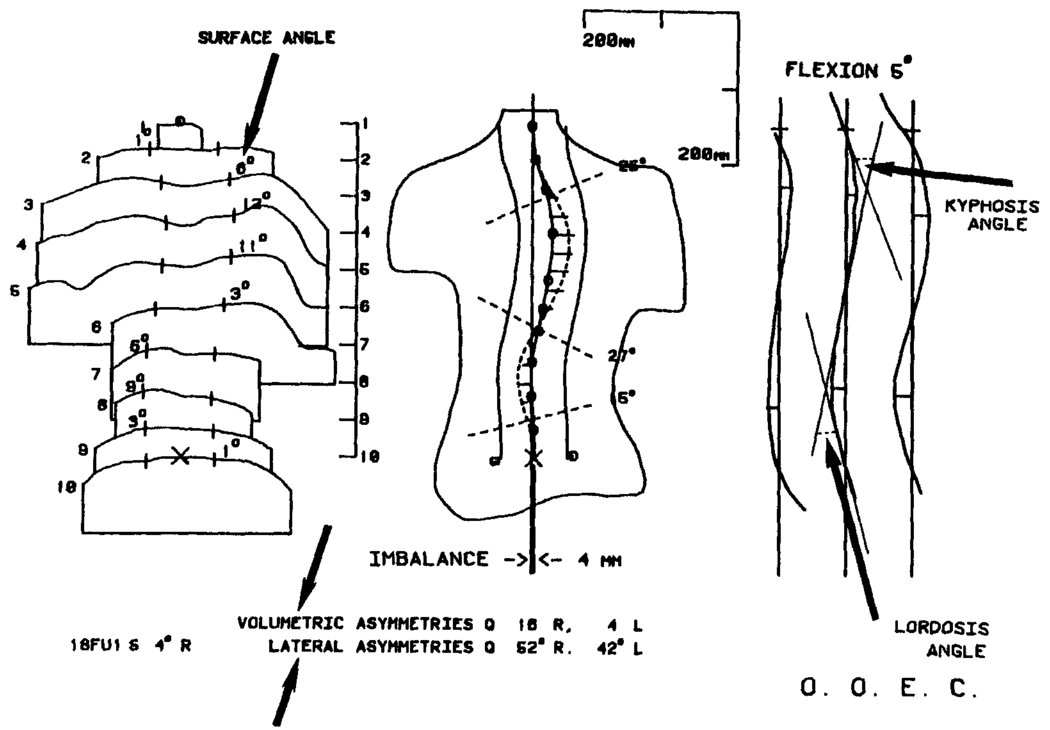

Figure 1. Typical ISIS scan. The arrows indicate parameters used in the study: viz., lateral asymmetry, volumetric asymmetry, surface angle, kyphosis, and lordosis.

The lateral asymmetry may be considered the surface analogue of the Cobb angle, and represents the lateral element of cosmetic deformity. Both the surface angle and the volumetric asymmetry quantify any asymmetry of the back surface in the transverse plane and provide an estimate of the effects of vertebral rotation in a scoliotic spine. At some ISIS levels, the right side of the back will be more prominent than the left, and vice versa, so that the routine scan will produce two values for the volumetric asymmetry: one to the right and the other to the left. The surface angle represents the slope of the back surface at a particular level (ISIS analysis divides the back into 10 evenly spaced levels between the vertebra prominens and the posterior iliac spines).

In the present study, standard ISIS analysis was modified, following Carr et al. (1989), so that kyphosis and lordosis were measured as angles rather than millimeters distant from a flat back reference plane. Back shape was characterized by the measurement of specific surface parameters in each of the three orthogonal reference planes (Figure 1): i) coronal plane—lateral asymmetry, ii) sagittal plane—kyphosis and lordosis angles, and iii) transverse plane—volumetric asymmetry and maximum surface angle.

The subjects

Oxfordshire children initially attending the scoliosis clinic at the Nuffield Orthopedic Centre with diagnosed late-onset idiopathic scoliosis over an 18-month period between July 1986 and December 1987 were asked to consent, with their families, to inclusion in the study. Forty-six children (index cases) were eligible, and of these 40 (9 boys and 31 girls) were included. Six children and families were excluded for the following reasons: 3 were adopted or orphaned, and family details were unknown; 1 refused to participate; and 2 moved away from the area.

The age, sex, age at onset of spinal deformity (by history), curve site, side, and size were noted in the clinic, and all the children underwent a stereophotogrammetric ISIS scan (Turner-Smith 1988).

The first-degree relatives of these children comprised a) 37 out of 40 mothers—of the remaining 3, 1 had died and 2 were separated from the family and were not traceable; b) 35 out of 40 fathers—of the remaining 5, 2 had died, 1 was working abroad, and 2 had separated from the family; c) 23 out of 26 sisters—2 of the remaining 3 were living away or were not available for examination and 1 refused; d) 15 out of 17 brothers—the 2 who were excluded were living abroad.

These first-degree relatives were examined clinically using the forward bend test for any evidence of spinal deformity. They were also examined using ISIS.

The criteria for suspecting a scoliosis were the presence of any one of the following: i) a positive forward bend test, ii) a lateral asymmetry of greater than 10°, iii) a maximum skin surface angle of greater than 7°.

The 21 relatives who fulfilled one or more of the above criteria all underwent radiography, having given their prior consent. Of these, 8 were found to have an idiopathic scoliosis of Cobb's angle greater than 10°.

The group of normal subjects is described in detail elsewhere (Carr et al. 1991). It consisted of 266 normal schoolchildren aged 10–16 years and 72 normal adults aged 21–59 years. There were equal numbers of males and females. All of them were scanned by ISIS and assessed clinically by means of the forward bend test.

In addition, 50 children attending the scoliosis clinic with idiopathic curves between 11° and 30° were included to ensure that ISIS can discriminate small scoliotic curves.

Table 1. back shapes of the seven different groups of subjects. ISIS parameters, mean *SD*

Lateral asymmetry		Volumetric asymmetry		Maximum surface angle	Kyphosis angle[a]	Lordosis angle
right	left	right	left	all right		
Group 1. Scoliotic children, index cases (n 40; C 41° *15°*)						
41	26	23	3.3	14	23	27
16	*19*	*15*	*5.2*	*5.4*	*13*	*8.9*
Group 2. Unaffected parents (n 76)						
11	7.5	7.2	1.7	5.6	38	34
7.0	*7.0*	*7.1*	*3.5*	*2.3*	*7.0*	*9.8*
Group 3. Unaffected siblings (n 39)						
9.9	6.1	8.5	0.8	6.0	33	29
6.5	*6.2*	*6.5*	*2.1*	*2.5*	*5.7*	*9.0*
Group 4. Normal children (n 271)						
11	7.4	8.2	1.4	5.8	37	34
5.8	*6.8*	*7.4*	*3.2*	*2.3*	*7.3*	*8.3*
Group 5. Normal adults (n 72)						
12	7.3	8.8	1.2	6.1	43	33
6.2	*6.8*	*6.3*	*2.6*	*2.4*	*6.8*	*12*
Group 6. Scoliotic children, 11°–20° (n 26; C 14° *2.6°*)						
20	16	12	1.5	9.6	28	29
6.4	*11*	*7.7*	*1.9*	*3.6*	*5.7*	*9.8*
Group 7. Scoliotic children, 21°–30° (n 24; C 24° *10°*)						
29	15	18	2.2	12	28	27
11	*15*	*11*	*4.5*	*2.7*	*8.6*	*10*

[a] C Cobb's angle.

Results

The scoliotic children—index cases

The back shape of the scoliotic children demonstrated the typical features of lateral curvature, rotation, and relative flattening of the lateral profile (Table 1). Linear regression analysis yielded a correlation coefficient of 0.82 ($P < 0.01$) between lateral asymmetry and Cobb's angle. On the basis of a Student's *t*-test, no differences were found between the characteristic parameters of back shape in the male and female scoliotic children except for the kyphosis angle, which was greater in the boys ($P < 0.05$).

The index children were also divided into two groups according to whether or not there was a positive family history of scoliosis. Maximum skin surface angles were found to be greater in those children with a positive family history (Student's *t*-test, $P < 0.05$). There were no other differences between either radiographic or ISIS parameters of back shape in the two groups.

The relatives

Compared with the 8 affected relatives (whose back shapes were found to be similar to that of the index children), the back shapes of the unaffected family members showed considerable reductions in both lateral and transverse asymmetry (Table 1). Some of the unaffected family members had high values for lateral asymmetry, maximum surface angle, or volumetric asymmetry; but they were subsequently found to be radiographically normal. Apart from the kyphosis angles, which were larger in the parents ($P < 0.001$), there were no differences between the back shapes of the unaffected parents and siblings.

When the unaffected family members were divided according to sex, no significant differences were found between the mothers and fathers, or between the brothers and sisters. If the families were

Table 2. Significant differences at the 5% level in ISIS parameters between different groups of subjects. Groups

Lateral asymmetry		Volumetric asymmetry		Maximum surface angle	Kyphosis angle	Lordosis angle
right	left	right	left	all right		
Group 1. Scoliotic children						
2–7	2–7	2–6	–	2–6	2–5	2–4
Group 2. Unaffected parents						
1,6,7	1,7	1,6,7	–	1,6,7	1,4,6,7	–
Group 3. Unaffected siblings						
1,6,7	1,6.7	1,6,7	–	1,6,7	1,2,5	–
Group 4. Normal children						
1,6,7	1,6,7	1,6,7	–	1,6,7	1,5,6,7	–
Group 5. Normal adults						
1,6,7	1,7	1,6,7	–	1,6,7	1,3,4,6,7	–
Group 6. Scoliotic children, 11°–20°						
1–5	1,3,4,7	1–5	–	1–5	2,4,5	–
Group 7. Scoliotic children, 21°–30°						
1–5	2–6	2–5	–	2–5	2,4,5	–

subdivided into those who had only 1 affected member and those with more than 1 affected member, then, no difference was detected for any of the ISIS parameters. Comparisons were made between parents and siblings independently.

Small curves

This group of 50 children was divided into two parts according to the Cobb angle, 11°–20° and 21°–30°. All the children had right thoracic curves (Table 1). There was a difference between the right-sided lateral asymmetry in the two groups ($P < 0.01$).

Normal subjects

The adult males and females had larger kyphosis angles than all the other groups of the same sex ($P < 0.05$); and among the children, the girls had larger lordosis angles ($P < 0.001$) and left-sided volumetric asymmetry ($P < 0.01$) than the boys (Table 1). Apart from these things, there were no differences between the groups, neither according to age nor according to sex.

The back shapes of the different groups were compared using an analysis of variance and Scheffe's test with a significance level of 5 percent. The results are summarized in Table 2.

To determine if the unaffected relatives showed any trend of back shape that may predispose them to develop scoliosis, they were compared with the index children, normal children and adults, and children with small scoliotic curves (Tables 1 and 2). There were no differences between the unaffected parents and normal adults, or between unaffected siblings and normal children in the coronal and transverse planes. However, the back shapes of the index scoliotic children and the children with small curves were different from all the other groups, with significant lateral curvature and transverse asymmetry. In the sagittal plane the unaffected parents and normal children were different from the index cases and from the children with small curves. The index scoliotic children had a much flatter sagittal profile, with smaller kyphosis angles. The unaffected siblings had sagittal profiles that were midway between those of the normal children and the children with small curves, so that they did not differ from either of these groups.

Discussion

The back shape of the index children as measured by ISIS showed clearly the three characteristic components of the curve in all three reference planes, whether relative flattening of the sagittal profile, vertebral rotation (evidenced as the maximum skin surface angle/volumetric asymmetry), or lateral curvature.

The ISIS system has been shown to be a reliable and reproducible noninvasive method of assessing back shape (Fidler et al. 1984). It has allowed us to safely undertake a novel study of the back shape of unaffected family members.

For the unaffected family members, no differences were observed between the parents and siblings except for kyphosis angle, which was greater in the adults. Also, the right-sided volumetric asymmetry was greater than that on the left, suggesting that the right side of the back was more prominent. Similar observations were made regarding the back shapes of normal children and adults (Carr et al. 1991). Other methods of assessing back shape have also noted a right-sided asymmetry (Burwell et al. 1983), which may be caused by increased prominence of the paraspinal musculature rather than any skeletal asymmetry.

No difference was observed between parents or siblings with a positive family history of scoliosis and those without. This does not seem to conform with the belief that certain families, who might be expected to have flatter backs than normal, possess a strong genetic susceptibility to the disorder. However, it may well be that the relatives with flatter sagittal profiles have already developed a scoliosis with associated lateral curvature and rotation.

It is interesting that the unaffected siblings had sagittal profiles that were midway between those of the normal children and the children with small curves, so that they did not differ from either of these groups. The siblings of affected families are considered to be more important because they may be expected to have a genetic makeup that is similar to their scoliotic brother or sister. This may represent an inherited tendency in back shape.

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