Early detection of idiopathic scoliosis – analysis of three screening models

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Abstract

Introduction: The prevalence of lateral curvatures of the spine ranges from 0.3% to 15.3% in the general population. The aim of the study was to develop and compare three different screening tests for idiopathic scoliosis (IS) with respect to their effectiveness and costs.
Material and methods: The Delphi method was used to assess the efficacy of each screening algorithm in detecting IS in the population. An economic analysis was also performed.
Results: Diagnostic Algorithm 1 for IS comprised a screening examination performed by nurses and a general practitioner (GP) with verification by specialists. The unit cost of carrying out diagnostic work-up for IS in Algorithm 1 was €94 per child. The second algorithm involved the use of the moiré computer method, followed by verification by a specialist. The lower unit cost of €86 per child of diagnostic work-up according to Algorithm 2 was due to fewer stages compared to Algorithm 1. The highest effectiveness with the highest costs were found for the third algorithm, with only one stage, a specialist's consultation (cost €153 per child).
Conclusions: The number of stages in an algorithm does not correlate positively with its efficacy or cost. The recommended scheme is Algorithm 3, where children are examined by rehabilitation specialists or a physiotherapist using a scoliometer and an inclinometer. The use of the apparently most expensive scheme (Algorithm 3) should result in lowering the costs of treatment of established idiopathic scoliosis and, in the long term, prove to be the most cost-effective solution for the health care system.

Key words: idiopathic scoliosis, screening, scoliosis prevention, spinal deformity.

AMS

Introduction

The prevalence of abnormal lateral spine curvatures in the general population ranges from 0.3% to 15.3%, with idiopathic scoliosis (IS) accounting for about 90% of all spinal deformities – up to 13.77% of the general population [1–4].

Idiopathic scoliosis can result in serious organic deformities of the musculoskeletal and respiratory systems. They cause pulmonary dysfunction with a decrease in forced expiratory volume and vital capacity that may lead to cardio-respiratory failure and, in severe cases, even premature sudden death [5–9]. Furthermore, other less serious complications of IS are encountered, such as chronic back pain, low self-esteem, reduced quality of life and mild mental disturbances or central coordination disorder (CCD) [10, 11].

Significant variations in the reported prevalence of detected IS cases result from the use of different methods of assessment, incompatible IS classifications, as well as inconsistencies within the populations under investigation, including sample size calculations (i.e. the number of children who have an X-ray or magnetic resonance imaging (MRI) examination in order to identify scoliosis). In Europe, IS is estimated to occur in 1% to 14% of children aged between 6 and 16 years [12]. Epidemiological studies have demonstrated that IS is significantly more frequent in girls than boys, reaching a ratio of 4 : 1 [13]. It also has been shown that intensive rehabilitation can significantly improve the patient's condition, with a considerable reduction of abnormal spinal curvatures [14]. Current data suggest that IS detection in Poland among children and teenagers is not very effective, as only 30% of all cases are identified [15]. It is obvious that more effective screening tests for idiopathic scoliosis should be performed to significantly increase the IS detection rate at an early stage. Using this diagnostic approach, one ought to be also aware of the environmental background of idiopathic scoliosis [16].

The present study was stimulated by the high prevalence of scoliosis in children aged 6–16 years, the high cost of untreated scolioses to society, and the lack of a uniform effective system for monitoring scoliosis.

Accordingly, it is advisable to try to find an accurate diagnostic method that would allow for the identification of as many children at risk of complications of IS as possible at an early stage of the disease. The diagnostic efficacy of such a method ought to be balanced against the financial cost it brings for the health care system.

Considering the above, we attempted to develop the most effective and least expensive approach to population screening to detect idiopathic scoliosis in children between the ages of 6 and 16 years.

Material and methods

The Delphi procedure was employed as a standard method used for long-term prediction of the efficacy of diagnostic and therapeutic procedures and the associated costs in health services [17–20].

We invited 5 Polish experts in medical rehabilitation or physiotherapy to take part in the study. The inclusion criteria comprised: specialisation in medical rehabilitation or physiotherapy, working in these specialties for at least 15 years, active daily practice in the treatment of idiopathic scoliosis, having treated at least 100 children with idiopathic scoliosis a year, having authored 5 or more publications on idiopathic scoliosis, and possessing a Ph.D. degree or a higher academic degree.

In the first round (Round 1 – Figure 1) of the Delphi assessment, based on both clinical expertise and knowledge, the experts proposed three possible algorithms of diagnosis and treatment of IS. Discussions about all details regarding those algorithms continued until all experts accepted them, with no critical comments or queries remaining unresolved.

In stage 1 of Algorithm 1, community nurses conduct examinations of posture in posterior, lateral and anterior views. The examination should proceed from the feet to the head. Symmetry of the feet, crurotalar articulations, pelvis, knee joints, waistlines, scapulas, shoulders and head position should be evaluated. If a postural asymmetry is noted, the nurse should qualify the child for the next stage of examination with a suspicion of idiopathic scoliosis. It is not verified if this asymmetry corresponds to a central axis (spinal column) asymmetry. Especially in younger children (age < 14 years old) trunk asymmetry does not significantly correlate with scoliosis, and further examinations are needed [21, 22]. At the second stage, children suspected of having scoliosis are clinically examined by a general practitioner
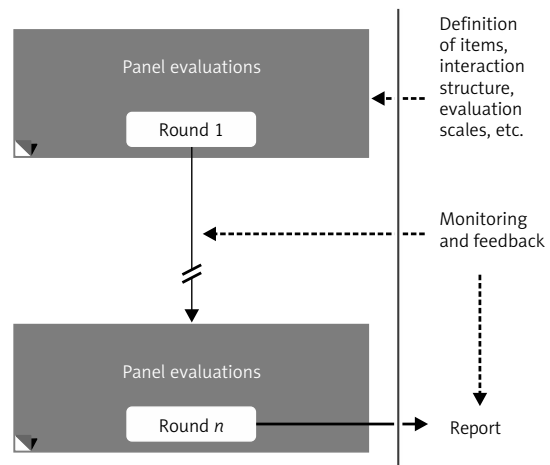

Figure 1. Stages of the Delphi method

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(GP). The GP examines the active movements of flexion and extension as well as lateral flexion of the spine in the cervical, thoracic and lumbar segment. This is followed by the forward bending test for the cervical to lumbar spine in the standing and sitting position. The position of spinal processes and presence of a costal hump are evaluated. The aim of the functional examination is to distinguish between faulty posture and actual idiopathic scoliosis. The detection of an asymmetry during the postural examination with the presence of scoliotic arcs and sometimes also a costal hump in the forward bending test of the cervical to lumbar spine in the standing and sitting position is tantamount to a diagnosis of idiopathic scoliosis. Patients with confirmed spinal deformities are subsequently referred to the 3rd stage, which is conducted at a specialized rehabilitation outpatient clinic where the history is taken and a physical examination is conducted with special focus on individual phases (visual inspection, active motion, passive motion, palpation, and functional and orthopaedic tests). The physical examination mainly employs a scoliometer and an inclinometer. If necessary, the final diagnosis is established by means of X-ray examination and other instrumental methods if necessary for very precise diagnosis, e.g. static and dynamic postural evaluation, surface electromyography (EMG) examination, or magnetic resonance imaging. The static postural evaluation serves to measure body sway and changes of the projection of the centre of gravity in a free standing position. It is used to diagnose disturbances of transfer of the force of gravity in patients with IS. The static posture evaluation uses a platform to record ground reaction forces in a static standing position. The dynamic posture evaluation uses a platform to record ground reaction forces as well as body sway during walking and the centre of gravity during jumps and different activities. Superficial EMG, which is helpful in examining spinal muscle work, is used in the detailed work-up of IS. The technology of complex analysis of movement can be applied with passive markers that reflect emitted infra red (IR) waves as well as ground reaction platforms connected to EMG recorders and video cameras. The examination of complex movement allows three-dimensional analysis of skeleton and muscle movement [23, 24]. In the case of a positive result, the child is referred for the 4th stage, that is outpatient (patients are not hospitalized) or inpatient rehabilitation (patients are hospitalized in more difficult and more advanced cases of scoliosis).

In Algorithm 2, the first stage consists of a surface topography examination of body posture, using a moiré-based method [25]. The examination is performed by a trained physical therapist for the entire study population. An examination with the Moiré technique is a computer examination of human body surface topography with the use of moiré patterns. The moiré technique is spatial photogrammetry (phototopography), used in population screening [26, 27]. Children with a suspicion of idiopathic scoliosis are subsequently referred for stage '2' at a specialized rehabilitation outpatient clinic in order to exclude or confirm IS by X-ray examination. In the case of a positive result, the child is referred for the 3rd stage, that is outpatient or inpatient rehabilitation.

Algorithm 3, in stage '1', involves a diagnostic examination performed at school [28]. The examination is carried out by a physician specializing in rehabilitation or orthopaedics supported by a team of physiotherapists and a complete set of diagnostic equipment (such as an inclinometer or scoliometer). In the case of a positive result, the child is referred for the 2nd stage of Algorithm 3, that is outpatient or inpatient rehabilitation.

In the second round (Round n – Figure 1), based on their clinical expertise and review of literature data, the experts were asked independently what percentage of IS cases may be detected at each stage of testing in each algorithm (i.e. subjective estimation of true positive (TP)) cases in the population per 10,000 children aged 6–16 years [29, 30]. Population risk of IS is 13.77%, so the experts could expect 1377 children with IS [1, 2]. The second round (Figure 1) was repeated several times until full agreement was achieved. The values were then subjected to a basic mathematical analysis (Table I).

Finally, an economic analysis was carried out (Table I). The costs of each programme were calculated on the basis of medical service rates (re-calculated from the Polish national currency to Euro) followed at the time by the National Health Fund in Poland [31].

Results

Diagnostic Algorithm 1 for IS comprised a screening examination performed by nurses (stage 1: population under investigation $n = 10,000$; children at risk of scoliosis in that population, at the level of IS risk $ISR_\% = 13.77\%$, $n = 1377$; TP rate is 34%, $n = 468$), an examination by a GP (stage 2: population under investigation $n = 468$, TP rate is 58%, $n = 271$), and verification by specialists (stage 3: population under investigation $n = 271$; TP rate is 90%, $n = 244$). The unit cost of carrying out diagnostic work-up for IS in Algorithm 1 for TP was €94 per child. Since, as a result of the low TP rate in stages 1 and 2, a rather small group of children was examined, the total cost of Algorithm 1 for the health care system was €22,956.5 (Table I).

The second algorithm involved the use of the moiré computer method (stage 1: population un-

Table I. Number of detected cases and detection unit costs for the population ($n = 10,000$) and estimated percentage of children at risk of scoliosis in the general population, at the level of IS risk $ISR_{\%} = 13.77\%$; $n = 1377$

Screening stages	Single test cost [Euro]	Population under investigation, n	True positives (%)	True positives, n	Total examination cost [Euro]	Case at IS risk/unit cost [Euro]
Algorithm 1:						
Stage 1	1.75	10,000	34	468	17,500	37
Stage 2	5.0	468	58	271	2,340	9
Stage 3	11.5	271	90	244	3,116.5	13
Stage 4				244		
Total				244	22,956.5	94
Algorithm 2:						
Stage 1	5.2	10,000	57	785	52,000	66
Stage 2	11.5	785	90	707	9,027.5	13
Stage 3				707		
Total				707	61,027.5	86
Algorithm 3:						
Stage 1	20.0	10,000	95	1308	200,000	153
Stage 2				1308		
Total				1308	200,000	153

der investigation $n = 10,000$; children at risk of scoliosis in that population, at the level of IS risk $ISR_{\%} = 13.77\%$, $n = 1377$; TP rate is 57%, $n = 785$), followed by verification by a specialist (stage 2: TP is 90% $n = 707$). The higher efficacy of Algorithm 2 (compared to algorithm 1) was due to the higher rate of TP results in stage 1. Algorithm 2 involved a larger group of children in stage 2. The lower unit cost of €86 of diagnostic work-up according to Algorithm 2 was due to fewer stages compared to Algorithm 1. The examinations involved a much larger group of children owing to a lower TP rate per stage. The total cost to the health care system of diagnostic work-up according to Algorithm 2 was €61,027.5 (Table I).

The highest effectiveness was achieved with the third algorithm (TP rate = 95%, $n = 1308$; €200,000). The unit cost of diagnostic work-up according to Algorithm 3 was the highest, at €153. The cost of the specialist examination was €20. As Algorithm 3 for diagnostic work-up of IS involves just one stage, it should be administered to the entire paediatric population. The calculations presented in this paper were carried out for a population of 10,000 children, resulting in an overall cost to the health care system of €200,000 (Table I).

Discussion

The algorithms developed by the participating experts were subsequently evaluated by the same experts with regard to their efficacy in diagnosing IS in terms of TP rates. The different TP values assigned were the result of detailed examination procedures at individual stages of each algorithm.

The widespread use of the diagnostic and therapeutic method that meets the expected efficacy targets on the population scale has to be relatively cost-effective, medically efficacious and must embrace all estimated cases of IS. The Delphi method was employed to choose the model of diagnostic work-up in idiopathic scoliosis that would be most appropriate of all commonly known algorithms of diagnosis and therapy. The selection of a populationally effective method for use in children is not based on medical efficacy as the sole criterion, but also, or perhaps most of all, accounts for the costs of implementing a given algorithm. The use of the Delphi method in this case is essential so that the least expensive, but still highly effective, method can be chosen by relying on the expertise of academic authorities who are also practitioners.

Algorithm 1 was less precise because examinations were carried out by a nurse. They did not involve imaging investigations or specialized objective investigations. Nurses are not experienced in diagnosing idiopathic scolioses; therefore, examinations carried out by nurses may be associated with a low percentage of TP results. While severe idiopathic scoliosis is readily identifiable in

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such examinations, early IS, producing hardly any symptoms, may escape diagnosis [32].

The results of the analysis performed for the purpose of this work allow the conclusion that the most cost-effective method for detection of idiopathic scoliosis is that described by Algorithm 2. The lowest unit cost of administering Algorithm 2 stems from a lower number of stages than in Algorithm 1. This lowest unit cost (€86) will generate much higher overall costs for the health care system as the number of children to be examined in Stage 2 is going to be much higher.

Algorithm 3 is the most effective and quickest approach. It is also, but only seemingly, the most expensive to implement. The number of cases of IS detected with this algorithm (TP results) is very high, corresponding to epidemiological estimates of the prevalence of IS in the population of children aged 6–16 years. With Algorithm 3, the number of children qualified for treatment is 1,308 out of every 10,000, compared to 244 with Algorithm 1 and 707 with Algorithm 2. Thus, while the initial costs of implementation of Algorithm 3 are the highest, this solution becomes more economical later on as established scolioses do not develop and so it is not necessary to treat their sequelae. Looking at the costs generated by each algorithm, it should be pointed out that Algorithm 2 is associated with the use of very expensive diagnostic equipment the cost of which exceeds that involved in the implementation of Algorithm 3.

The fundamental assumption underlying the use of a diagnostic method in IS is that the percentage of TP results should match the prevalence rate of IS in the entire population of children aged 6–16 years. Reliance on any other diagnostic approach that fails to detect every possible case of IS in the entire population is unethical. The initially higher cost of implementing a method producing a high percentage of TP diagnoses is also economically feasible. Undetected cases of IS will eventually generate much higher costs in health care to treat established IS, including the cost of treating back pain in the shorter and longer term, cost of surgery and, in extreme cases, the cost to society associated with inability to work.

The Delphi method was applied so that the participating experts, who are authorities in the field, could estimate the values of the TP parameters, which determined the percentage of spinal deformities detected at particular stages of the early testing algorithms. The costs of each programme were calculated on the basis of medical service rates (re-calculated from the Polish national currency to Euro – Table I), which were at the time followed by the National Health Fund in Poland [30]. Also, financial simulations were carried out for individual projects.

An economic analysis was performed for the three algorithms of screening tests (Figure 2). The results concerning individual algorithms of screening tests for a general population (GP) of 10,000 children, with the assumed maximum percentage of IS risk cases being GP_{SR} =13.77% [1–3], are shown in Table I.

In other words, the application of cost-effective methods at the initial stage of screening tests is of paramount importance. Moreover, it should be stressed here that for the simplest examination programme (Algorithm 1), a significant number of children being at risk of scoliosis are excluded from further work-up due to the first stage findings. As a result, these children fail to gain access to appropriate treatment. The price estimates of the screening in the school setting conducted by a nurse and other examinations were based on official Polish medical service prices.

It is well known [33] that untreated scoliosis may to some extent affect patients' social functions, including marriage, child-bearing and professional development. Thus, when such comparative analyses are performed, one should also consider the social value of potential health improvement (when more precise methods for IS de-
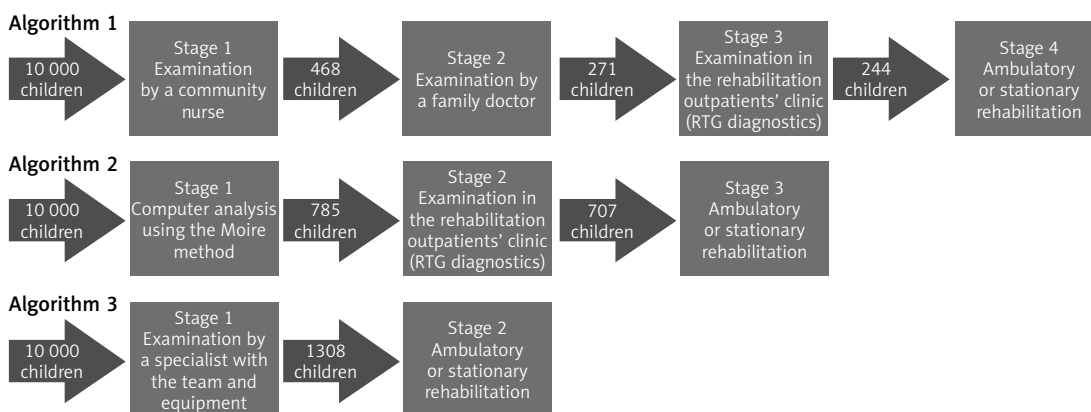

Figure 2. Three diagnostic screening algorithms for scoliosis. The presumed number of children involved at each stage is given

tection are used). The above could be expressed in a measurable way (preferably as a financial simulation, e.g. by applying the quality-adjusted life year (QALY) or quality of life (QOL) of scoliosis indicator) [34, 35].

This issue should be investigated further, with a special focus on determination of the actual IS risk level for children and teenagers, possibilities of IS prevention (halting its progression), especially at the society level (determination of the QALY indicator can serve this latter purpose), possible assessment of long-term social benefits resulting from the use of a cost-effective IS prevention method and thorough verification of qualitative parameters obtained from the Delphi method, in order to increase the precision of determination of the percentage of children at IS risk.

In conclusion, the number of stages in an algorithm does not correlate positively with its efficacy or cost. The recommended scheme is Algorithm 3, where children are examined by rehabilitation specialists or a physiotherapist using a scoliometer and an inclinometer. The use of the apparently most expensive scheme (Algorithm 3) should result in lowering the costs of treatment of established idiopathic scoliosis and, in the long term, prove to be the most cost-effective solution for the health care system.

Acknowledgments

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