

Assessment of Early Juvenil Idiopathic Scoliosis: Clinical Trial for the Evaluation of A School-Based Screening Protocol

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Abstract

This study aims to evaluate the effectiveness of clinical tests currently in use for the diagnosis of JIS, through a survey, carried out in secondary schools in the province of Palermo, in order to standardize a school screening protocol that could be extended to the whole regional territory. The adolescents underwent a specialist psychiatric examination in their classrooms; moreover three screening tests have been administered: Adams test, Bunnel Inclinator and Plumb line test. In case of single positivity to one of the three tests, the adolescent underwent to radiography of the whole column. Then we examined the sensitivity and diagnostic specificity of the Adams or inclinometer test, respectively: 56.3% and 92.7%. On the other hand, the positivity to at least one between inclinometer or plumb line sensitivity was higher than the specificity: 91.3% versus 80.8%. The same happened for the positivity to at least one test between Adams index or plumb line test, which showed a sensitivity of 95.2% compared to a specificity of 81.5%. Finally, taking into consideration patients with triple test positivity, a clear increase in specificity compared to sensitivity was demonstrated, reaching 99.7%. Performing a single test cannot be considered a sufficient tool aiming to detect potential scoliotic subjects. To conclude, an effective screening program can reduce health care expenditure related to specialist medical examinations as well as unnecessary instrumental exams such as X-rays.

Background Section

Juvenile Idiopathic Scoliosis (JIS) is a complex structural deformity of the spinal column on the three planes of space; on the frontal plane a lateral bending movement occurs, as well as an alteration of the curves on the sagittal plane, most often causing a curve inversion; on the axial plane a rotational movement still occurs [1–3].

Classically, JIS does not recognize a well-known cause and probably not even a singular cause. Nevertheless, considering the etiopathogenetic mechanisms, vertebral deformity, caused by JIS, can be defined as a sign of a complex syndrome with a multifactorial etiology [4–5]. Possible etiological factors such as female sex, familiarity, first born being, genetic background, biomechanical or neurological disorders, aberrant hormones functioning such as growth hormone and melatonin, body schema disorders are mentioned in several studies. Behind this etiological heterogeneity, clear scenarios of autosomal dominant or autosomal recessive transmission are described together with a multifactorial heredity background [6].

According to recent literature, the prevalence rate varies from 0.47–5.2%, though it is commonly accepted 2–3% occurrence of the disease in the general population, with a female to male ratio of 4:1 [7].

The most common classification identifies congenital and acquired scoliosis. Moreover, acquired scoliosis is classified into idiopathic acquired scoliosis (80% of cases) and acquires scoliosis secondary to neurological diseases, connective system diseases, etc. Other criteria that need to be considered are chronological criterion that enable to divide three types of scoliosis based on the age at the diagnosis

(Infant 0–2 years, Youth 3–9 years, Adolescent 9–18 years); radiological criterion which, according to Cobb's Angles, classifies six severity degrees (mild < 15°, mild-moderate 16–24°, moderate 25–34°, moderate-severe 35–44°, severe 45–59°, very severe > 60°); lastly topographic criterion identifies five types of scoliosis based on the siting of the curves (cervical C6-C7, cervical-dorsal C7-T1, dorsal T1-T12, dorsal-lumbar T12-L1; lumbar L1-L5) [8].

According to Society On Scoliosis Orthopaedic and Rehabilitation Treatment (SOSORT), a Cobb's Angle greater than or equal to 10° radiographic degrees defines scoliosis. Above 30° Cobb's Angle degrees the probability of anatomical damage increases significantly [8].

JIS may occur at any time of growth development. However, Tanner demonstrates that growth spurt does not proceed with a linear pattern; actually there are two periods of rapid growth, from birth to three years and during the pubertal crisis. This latter occurs at two different times in both sexes.

In fact, in females, the pubertal crisis coincides with or precedes the beginning of the development of the breast and pubic hair (about 11 years old), with a peak around the age of 12 and a term at the age of 14. The presence of menarche indicates a reduced risk in scoliosis progression. Conversely, in males it appears about two years after the appearance of pubic hair, with a peak around 16 years of age.

An early diagnosis correlates with less invasive conservative treatments, preventing surgery and avoiding respiratory, psychological and social complications that characterize the later stages of the disease [9].

Nowadays in Sicily, no early diagnosis program throughout a school-based screening process for JIS is validated.

This study aims to evaluate the effectiveness of clinical tests currently in use for the diagnosis of JIS, through a survey, carried out in secondary schools in the province of Palermo, in order to standardize a school screening protocol that could be extended to the whole regional territory.

Methods

This Quasi-experimental clinical trial was conducted by the Rehabilitation and Epidemiology and Preventive Medicine Hospital Units University of Palermo.

Between February 2019 and January 2020 preadolescents, that differ from urban area and socio-cultural status and attending eight first-grade secondary schools of the Province of Palermo were recruited.

Experimenters went to the schools and provided detailed information and behavioral norms for effective prevention of JIS.

Subsequently, the adolescents, whose parents had signed their informed consent, underwent a specialist physiatric examination in their classrooms; moreover three screening tests have been administered: Adams test, Bunnel Inclinator and Plumb line test.

Adams test: flex the patient's torso forward, with the head bent, arms relaxed and lower limbs extended: in this way, the torso is highlighted and the torso must be measured at the point where there is the greatest difference in height between the two sides. [10]

Bunnel's Inclinator: uses an inclinometer (scoliometer) to measure the tines and determine the angle of rotation of the trunk. [10]

Plumb line test: patient standing in correct posture with a straight gaze, position the tangent line until it reaches the intergluteal fold in order to obtain a true reference vertical for measurements. From the plumb line, once immobile, it is possible to calculate the distance up to the three reference points C7-D12-L3. The values considered normal for kyphosis are given by the sum of the arrow of C7 and L3 up to 90 mm, and at L3 level up to 55 mm. Values above 90 mm and 55 mm should lead to a specialist investigation according to the latest scientific ISICO screening protocol [10], however, there is no unanimous consensus.

In case of single positivity to one of the three screening tests, the adolescent underwent to further instrumental diagnostic investigation: radiography of the whole column, under load, in two projections (A/P and L/L) with measurement of Cobb's Angle and Risser grade. JIS was defined as a curve with 10° or more and rotation of the vertebral body, according to the Scoliosis Research Society (SRS) and the International Scientific Society on Scoliosis SOSORT (Society On Scoliosis Orthopaedic and Rehabilitation Treatment). Patients with confirmed diagnosis of radiographic scoliosis were then given an aesthetic evaluation through TRACE (Trunk Aesthetic Clinical Evaluation) developed by ISICO (Istituto Scientifico Italiano Colonna Vertebrale) [11].

The parameters taken into consideration by our study for the evaluation of the efficacy of the diagnostic tests examined were [Table I]:

- Sensitivity: = $a/(a + c)$, i.e. the probability that a patient testing positive;
- Specificity = $d/(b + d)$, i.e. the probability of a healthy person testing negative;

Table I

TEST	Sick	Healthy	TOT
Positives	a (true positives)	b (fake positives)	a+b
Negatives	c (fake negatives)	d (true negatives)	c+d
TOT	a+c	b+d	a+b+c+d

- Positive likelihood ratio (LR+) i.e. the ratio between (sensitivity) / (1 - specificity), i.e. $LR+ = [a / (a + c)] / [b/(d + b)]$

Specifically:

- LR + is the ratio between the probability that a patient is positive and the probability that a healthy person is positive. In other words, in the case of a positive test result, LR + is times more likely that the subject is sick than healthy.

- LR- is the ratio between the probability that a patient is negative and the probability that a healthy person is negative. In other words, in the case of a negative test result, LR- is times more likely that the subject is ill than healthy (Table II):

Table II

LR+	LR-	Diagnosis Result
>10	<0,1	conclusive
5-10	0,1-0,2	moderately useful
2-5	0,2-0,5	Sometimes useful
1-2	0,5-1	rarely useful
1	1	not useful

Results

428 subjects aged between 11 and 14 years (average age 11.87) were examined, including 228 males (53.3%) and 200 females (46.7%).

186 (43.46%) tested positive for at least one Adams, plumb line and inclinometer test. Of these, 20 subjects (10.8%) were positive in all three tests.

13 subjects (7%) were positive only at Adams test, 46 (24.7%) at both Adams test and inclinometer; 2 subjects (1.1%) were positive to both Adams test and plumb line; 2 at both inclinometer and plumb line (1.1%); 93 subjects (50%) were positive at plumb line only, 10 (5.4%) were positive at inclinometer only.

Associating the variables, a total of 81 adolescents (43.5%) were positive for the Adams test, 78 (41.9%) for the inclinometer and 117 (62.9%) for the plumb line (Fig. 1).

Interestingly, after the radiographic study, we observed that, among the 186 patients tested positive to at least one screening test, 126 of them (66.7%) had an angle of Cobb greater than 10° and therefore considered affected by JIS; moreover 46 (24.7%) adolescents had an angle greater than 8 and less than 10.

Analyzing and cross-referencing the data described above, we experienced that the Adams test sensitivity and specificity were of 50.8% and 94.4% respectively with a positive predictive value of 79%, representing,

among the three tests performed, the one with the highest specificity, sensitivity and positive predictive value.

Bunnel's test showed a sensitivity of 46%, a specificity of 93.4% and a positive predictive value of 74.4%.

Finally, the plumb line test showed the lowest results in terms of sensitivity (61.1%), specificity (86.8%) and positive predictive value (65.8%). [Table III].

Then we examined the sensitivity and diagnostic specificity of the group of positive subjects to at least one Adams or inclinometer test, respectively: 56.3% and 92.7%.

On the other hand, the positivity to at least one between inclinometer or plumb line sensitivity was higher than the specificity: 91.3% versus 80.8%.

The same happened for the positivity to at least one test between Adams index or plumb line test, which showed a sensitivity of 95.2% compared to a specificity of 81.5%. [Table IV-V].

Finally, taking into consideration patients with triple test positivity, a clear increase in specificity compared to sensitivity was demonstrated, reaching 99.7% [Table VI].

Table n°III

	Sensibility (%)	Specificity (%)	Positive predictive value (%)	Negative predictive value (%)	LR+ (n)	LR- (n)
Adams Index	50,8	94,4	79,0	82,1	9,02	0,52
Inclinometer	46,0	93,4	74,4	80,6	6,95	0,58
Plumb Line	61,1	86,8	65,8	84,2	4,61	0,45

Table n° IV

	Sensibility (%)	Specificity (%)	Positive predictive value (%)	Negative predictive value (%)	LR+ (n)	LR- (n)
Adams Index or Inclinometer	56,3	92,7	76,3	83,6	7,74	0,47
Inclinometer or Plumb Line	91,3	80,8	66,5	95,7	4,75	0,11
Adams Index or Plumb Line	95,2	81,5	68,2	97,6	5,14	0,06

Table n°V

	Sensibility (%)	Specificity (%)	Positive predictive value (%)	Negative predictive value (%)	LR+ (n)	LR- (n)
Adams Index and Inclinometer	40,5	95,0	77,3	79,3	8,15	0,63
Inclinometer and Plumb Line	15,9	99,3	90,9	73,9	23,97	0,85
Adams Index and Plumb Line	16,7	99,7	95,5	74,1	50,33	0,84

Table n°VI

	Sensibility (%)	Specificity (%)	Positive predictive value (%)	Negative predictive value (%)	LR+ (n)	LR- (n)
Positive at least one test	100,0	80,1	67,7	100,0	5,03	-
Positive to all three test	15,1	99,7	95,0	73,8	45,54	0,85

Declarations

The study was approved by the Ethical Committee Palermo I of the University Hospital of Palermo (5/2019 of May 22th 2019) with the frame of rules specified by both the Helsinki declaration (World Medical Association, 559th WMA General Assembly, Seoul, Republic of Korea, October 2008) and its subsequent amendments, and the principles of Good Practice.

All the authors of the paper “Assessment of early Juvenil Idiopathic Scoliosis: clinical trial for the evaluation of a school-based screening protocol” declare that they are not in a situation of conflict of interest, agree for publication and give to data and material. Don't have funding.

Ethics approval and consent to participate

The protocol for the study was approved by the Ethics Committee Palermo 1, University of Palermo. Each author certifies that all investigations were conducted in conformity with ethical principles. All participants provided informed consent before their participation in the study and written were obtained from all participants.

Consent for publication

The authors have received written consent from participants to publish individual patient data.

Availability of data and materials

Not applicable.

Competing interests

The undersigned, DS, corresponding author, and all the authors of the paper “Assessment of early Juvenil Idiopathic Scoliosis: clinical trial for the evaluation of a school-based screening protocol” declare that they are not in a situation of conflict of interest.

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Authors' contributions

All authors contributed equally to the manuscript and read approved the final version of the manuscript.

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Conclusions

Basing on the prevalence result in the Province of Palermo, implementing a school-based screening program and standardizing a protocol for the early diagnosis of JIS is needed. Performing a single test chosen from Adams test, inclinometer and plumb line test, cannot be considered a sufficient tool aiming to detect potential scoliotic subjects, due to the low sensitivity of each test. In fact, analyzing the high LR value of Adams test and inclinometer test, these tests administered individually may fail to intercept potentially ill subjects. So, they can be singularly considered useful but not conclusive diagnostic tests. Conversely the single plumb line test is not indicated both in diagnosing and screening process, having a lower sensitivity, specificity and positive predictive value.

Notably the association between Adams test/plumb line or inclinometer test/plumb line significantly increases the sensitivity while maintaining high specificity. The extremely high LR and positive predictive value obtained from these combinations, may suggest the use of both matches as a screening tool in order to identify JIS at an early stage.

The institution of a team of specialists focusing on screening programs, extended to the whole regional territory, for JIS is needed, besides considering the easy execution and high reproducibility of the three screening tests.

The identification of the most valid associations (Adams test/plumb line or inclinometer/plumb line) can provide an early diagnosis and may shed light to a full acknowledgement of the natural history of Idiopathic Scoliosis. Moreover, an early diagnosis may lead to a decrease in complications related to mistreated JIS and a consequent reduction of their social consequences.

To conclude, an effective screening program can reduce health care expenditure related to specialist medical examinations as well as unnecessary instrumental exams such as X-rays.

Abbreviations

JIS: Juvenile Idiopathic Scoliosis; SOSORT: Society On Scoliosis Orthopaedic and Rehabilitation Treatment; ISICO: Istituto Scientifico Italiano Colonna Vertebrale; AP: Antero-posterior; L-L: Latero-lateral; SRS: Scoliosis Research Society; TRACE: Trunk Aesthetic Clinical Evaluation; LR: likelihood ratio; WMA: World Medical Association;

References

1. Stagnara P [1985] Les déformations du rachis. Paris: Masson

2. Perdrille R [1979] La scoliose: son étude tridimensionnelle. Paris: Maloine
3. Selleri U, Negrini S [1999] La riabilitazione del paziente affetto da scoliosi idiopatica. Trattato di Medicina Riabilitativa, Medicina Fisica e Riabilitazione. Napoli: Idelson Gnocchi: 1123-47
4. Nachemson A, Sahlstrand T [1977] Etiologic factors in adolescent idiopathic scoliosis. Spine 2:176-84
5. Moen KY, Nachemson AL [1999] Treatment of Scoliosis: An Historical Perspective. Spine 24(24):2570-5
6. L. Aulisa et al [2008] La familiarità nella scoliosi idiopatica. G.I.O.T. 34 (suppl. 2):S00-S00
7. Konieczny M.R., Senyurt H., Krauspe R [2013] Epidemiology of adolescent idiopathic scoliosis. J. Child. Orthop. 7(1):3–9. doi: 10.1007/s11832-012-0457-4
8. Negrini S, Donzelli S, Aulisa AG, Czaprowski D, Schreiber S, de Mauroy et all. [2018 Jan 10] SOSORT guidelines: Orthopaedic and rehabilitation treatment of idiopathic scoliosis during growth. [Scoliosis Spinal Disord.](#)13:3. doi: 10.1186/s13013-017-0145-8
9. Fusco C, Donzelli S, Lusini M, Salvatore M, Zaina F, Negrini S [2014 Aug 18] Low rate of surgery in juvenile idiopathic scoliosis treated with a complete and tailored conservative approach: end-growth results from a retrospective cohort. [Scoliosis](#): 9-12. doi: 10.1186/1748-7161-9-12.
10. Protocollo Scientifico Screening ISICO Rev.10 [2018 Mar 3]
11. [Zaina F](#), [Negrini S](#), [Atanasio S](#) [2009 Jan 20] TRACE (Trunk Aesthetic Clinical Evaluation), a routine clinical tool to evaluate aesthetics in scoliosis patients: development from the Aesthetic Index (AI) and repeatability. [Scoliosis](#) 4:3. doi: 10.1186/1748-7161-4-3.

Figures

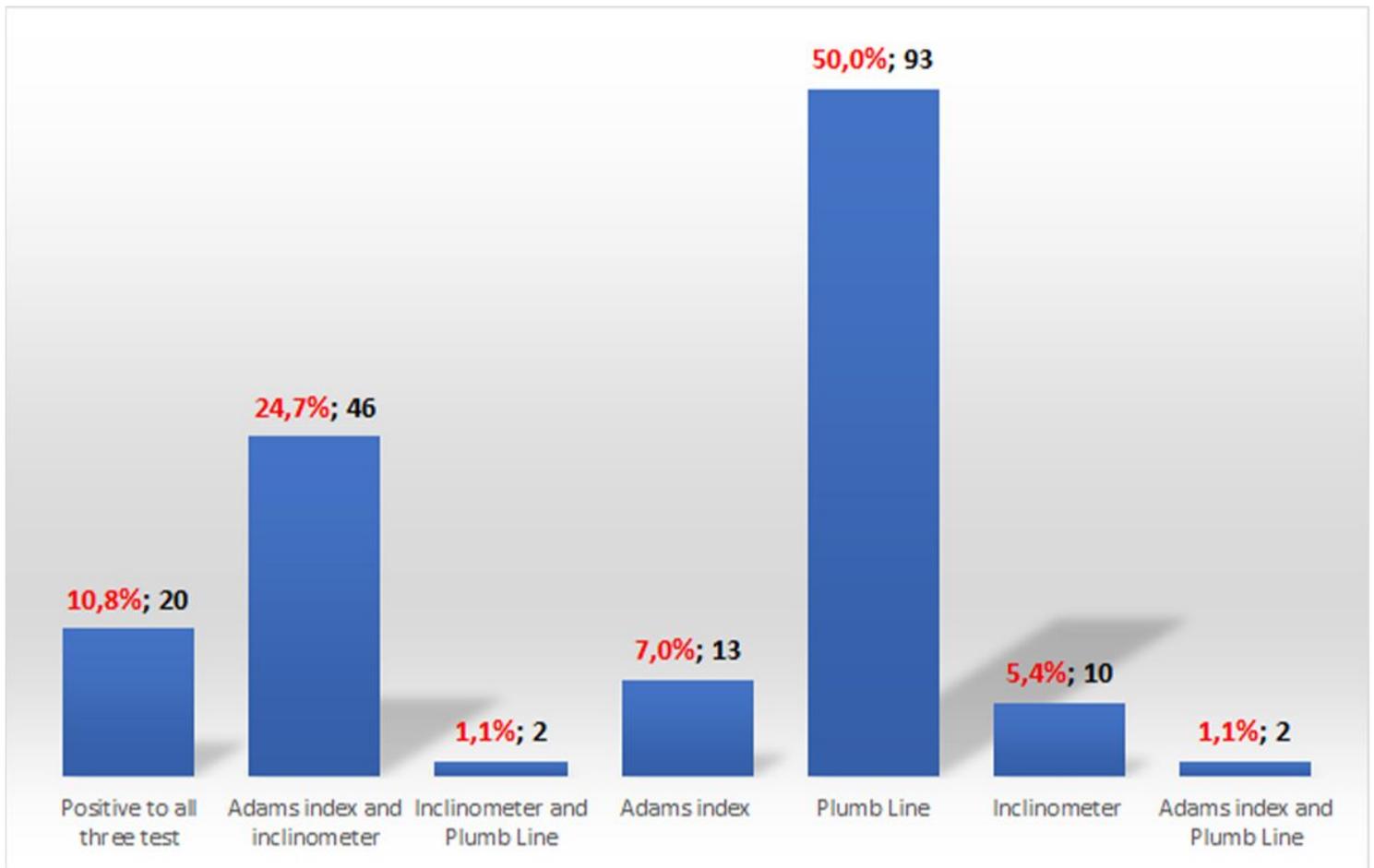


Figure 1

Figure 1