

Pediatric oncological spondylolisthesis: the contribution of Physical Medicine and Rehabilitation and Orthodontics. A narrative review

Andrea Scribante¹, Matteo Pellegrini^{2,3}, Martina Ghizzoni¹, Maria Gloria Nardi¹, Barbara Rocca⁴, Marco Monticone⁵

¹Section of Dentistry, Department of Clinical, Surgical, Diagnostic and Pediatric Sciences, University of Pavia, Pavia, Italy

²Department of Biomedical, Surgical and Dental Sciences, University of Milan, Milan, Italy

³Maxillo-Facial Surgery and Dental Unit, Fondazione IRCCS Ca' Granda Ospedale Maggiore Policlinico, Milan, Italy

⁴Department of Clinical Psychology, International Institute of Behavioral Medicines, Sevilla, Spain

⁵Department of Medical Sciences and Public Health, University of Cagliari, Cagliari, Italy

Abstract

Different forms of spondylolisthesis (SP) have been described. This pathological condition is often due to a birth defect in the lumbar area, but it can also be caused by a sudden injury resulting from an acute trauma. In athletic children and adolescents, SP may represent a stress fracture triggered by overuse. SP can also be consequent to cancer, although this clinical condition is rare. The first manifestation of SP is pain. Neoplasm-related SP may also induce physical impairment and postural alterations of the spine, such as flatback and hyperkyphosis. These changes may also affect the entire spine, including the cervical region (i.e., forward neck) and the head (i.e., forward head). A prompt assessment of children with oncological SP is crucial and involves clinical and radiological evaluations. Multiple healthcare professionals are involved in the management of children with oncological SP, including spinal specialists, pediatricians, oncologists, and psychologists. Orthodontists may also contribute to cervical posture evaluation. The authors of this review aim to provide a comprehensive overview of the topic, particularly focusing on the clinical

assessment of children with oncological SP and the possibilities for rehabilitative treatment.

Keywords

Dentistry, orthodontics, pain, pediatric oncology, rehabilitation, spondylolisthesis.

Corresponding author

Maria Gloria Nardi, Section of Dentistry, Department of Clinical, Surgical, Diagnostic and Pediatric Sciences, University of Pavia, Pavia, Italy; email: mariagloria.nardi01@universitadipavia.it.

How to cite

Scribante A, Pellegrini M, Ghizzoni M, Nardi MG, Rocca B, Monticone M. Pediatric oncological spondylolisthesis: the contribution of Physical Medicine and Rehabilitation and Orthodontics. A narrative review. *J Pediatr Neonat Individual Med.* 2023;12(2):e120218. doi: 10.7363/120218.

Introduction

There are 5 common types of spondylolisthesis (SP): congenital, isthmic, degenerative, traumatic and pathologic [1]. Congenital, traumatic and isthmic types are commonly found in pediatric patients, while degenerative and pathologic types may occur in both developing and older individuals [1]. In small children, SP is often due to a birth defect in the lumbar area or a sudden injury caused by acute trauma [1]. In athletic children and adolescents, SP may result from a stress fracture triggered by overuse [2].

Pathologic SP in children is anatomically characterized by a slip resulting from weakness within the bones [1, 3]. This weakness leads to the destruction of the posterior portion of the vertebrae, including the neural arch that surrounds the spinal cord [1].

Pediatric oncological SP can also cause physical impairment and postural alterations of the spine, such as hyperkyphosis or swayback [4]. These changes can also affect the posture of the entire spine, including the upper part of the column and the head [5].

The primary manifestation of SP is pain, which should never be underestimated by clinicians, particularly when dealing with pediatric patients [1]. Low back pain (LBP) is relatively common in children and is often attributed to inactivity and poor posture [6]. However, persistent LBP,

especially when accompanied by tingling, weakness, or numbness in both legs, should be considered a red flag – a clinical condition requiring rapid and thorough evaluation before starting any treatment [4].

A prompt assessment of children with LBP and suspected SP is necessary and involves appropriate clinical and radiological evaluations [1]. If an oncological disease is detected, further assessments are required, and surgery becomes unavoidable [7].

Various healthcare professionals, each contributing their skills and expertise, are involved in the treatment process, taking care of children with oncological SP [8].

The authors of this review aim to provide a comprehensive overview of the topic, particularly focusing on the clinical assessment of children with oncological SP and the possibilities for post-surgical rehabilitative treatment.

Materials and methods

Focused questions

What approach is taken in the clinical assessment of children with a pathological SP due to cancer? What conservative treatments are beneficial for treating these young individuals?

Eligibility criteria

The analysis of the studies was conducted based on the following specific inclusion criteria.

1. Study design: randomized clinical trials, observational and cohort studies, and case reports.
2. Participants: children with a history of pathological SP.
3. Interventions: conservative treatments concerning young people with pathological SP due to neoplasms.
4. Outcome: the role of clinical evaluation of kids with a pathological SP due to cancer. Studies that investigated conservative treatments in aid of these individuals were considered.

Only investigations that accomplished all these inclusion criteria entered the review. Furthermore, the following exclusion criteria were applied.

1. Abstracts of papers published in non-English languages.
2. Duplicate manuscripts.

3. Irrelevant studies, such as those with aims unrelated to the research questions, those analyzing different supplementary treatments, or those where the full-text content did not align with the abstract.
4. *Ex-vivo* or experimental animal researches.
5. Analyses without Ethics Committee authorization.
6. Meta-analysis, systematic reviews, or narrative reviews.

Search strategy

The PICO model (Population, Intervention, Comparison, Outcome) was referred to carry on this review. An extensive literature search was carried out by considering Scopus and PubMed (MEDLINE) electronic databases. Abstracts of papers evaluating the clinical assessments of children with SP due to cancer were identified and analyzed. Abstracts of studies on conservative treatments useful for treating these young individuals were investigated as well.

Research

The following medical subject heading (MeSH) terms were used in the search: spondylolisthesis, cancer, pain, pediatrics, and spine. The Scopus and PubMed (MEDLINE) databases were electronically searched, targeting manuscripts published between 2000 and 2023, and restricted to ages ranging from 11 to 19 years old. The latest search was conducted on July 5, 2023. Two reviewers, who were trainees as concerns this study analyses, performed the search and resolved disagreements and discrepancies through consensus and consultation with two extra reviewers. Titles and abstracts of the initially identified papers were carefully reviewed, and non-relevant studies were eliminated. The significant articles were then extensively surveyed to exclude any similar studies that met the inclusion criteria. For the extraction of relevant results, the full texts of the included studies were read, and all the findings were recorded.

Results

The primary search identified 140 articles based on MeSH terms, published from 2000 to 2023. Following those, the research has been restricted to randomized clinical trials, observational and cohort studies and case reports concerning human

studies; thus, 14 articles were screened and evaluated for eligibility.

Discussion

Clinical approach

A clinical approach should include a comprehensive anamnesis (i.e., the young individual history), a thorough physical examination, and a satisfactory radiological assessment.

As for the first evaluative point, information on familiar (for instance, by asking: “Did the suspect condition also occur in the parents or relatives?”), physiological (i.e., pregnancy, delivery, psychomotor development, and menarche), past (for instance, by inquiring: “Are there any clinical states or events that may have caused the suspect condition, such as traumas?”), and recent (for instance, by asking: “Has anything recently occurred that caused a spinal pain?”) history is mandatory.

As for the second assessing point, clinicians have to carefully evaluate the lumbar spine with special attention to [9]:

- i. lateral inspection, by observing any abnormal postural deviation (e.g., hyperkyphosis, sway-back, or hyperlordosis);
- ii. joint mobility, by evaluating the range of motion of the lumbar tract as the extension, lateral flexion, and forward bending;
- iii. evaluation of muscle strength, by assessing the main extensor, flexors and latero-flexors of the lumbar spine according to the Medical Research Council Scale for Muscle Strength, which ranges from 5 (normal strength) to 0 (no visible contraction);
- iv. palpation of the muscles, by feeling any abnormal contracture of the main lumbar muscles located in the back;
- v. peripheral neurological examination, by examining superficial and deep reflexes, as well as superficial, deep and composed sensitivity;
- vi. balance, by assessing the capacity to maintain static and dynamic equilibrium;
- vii. walking, by evaluating the ability of a young person to ambulate normally, and by pointing out any serious deviations from the standard schemes of gait.

An in-depth examination of the rest of the column and the head is further advised, to point out any additional postural deviations, such as

the forward neck and head, their joint mobility (also including the temporomandibular joint), and any muscle contracture in the neck or the head.

A widely known self-administered outcome measure, the Scoliosis Research Society-22 Patient Questionnaire (SRS-22), is advised to assess the health-related quality of life (HrQoL) of young people with spinal disorders [10]. It is a region-specific instrument and includes 5 domains (i.e., function, pain, mental health, self-image, and management satisfaction/dissatisfaction with treatment). The total score for each domain ranges from 1 to 5, with 5 being the best. Further refinements with this outcome measure were proposed [11]. The SRS-22 patient questionnaire is also available in the native language of the authors of this study, and further investigations also in oncological populations with spinal problems would be interesting [12].

Concerning the third evaluative point, a radiological examination is the best way to diagnose SP. In detail, a standing lateral X-ray allows us to see the vertebra that has slipped off the spinal column (usually L5-S1, then L4-L5, with multiple sites retrieved in blue cases) [13]. When there is a suspect of malignancy, more advanced evaluations are recommended, such as computed tomography and magnetic resonance imaging [13]. A full standing X-ray and posterior-anterior and latero-lateral views allow a complete spine assessment to exclude additional deformities (i.e., scoliosis), and a skull X-ray is advised by expert physicians [14]. More invasive radiological examinations of the full spine and the head are recommended only when there is a suspicion of bone or visceral metastases in these tracts of the locomotor system [13, 14].

Aside from the physical assessment, psychological disturbances evaluations are also recommended to rapidly catch mood disorders and maladaptive thoughts that are expected when a pain in the back occurs [15]. These feelings are, of course, greatly worsened when an oncological diagnosis adds up to pain [15]. Measures of mood disorders, catastrophizing, and fear avoidance were discussed by the authors of this study, but further analyses are advised to investigate their application and evaluation within young Italian individuals [16-18].

Tab. 1 shows symptoms and signs of pediatric oncological SP.

Table 1. Symptoms and signs of pediatric oncological spondylolisthesis (SP).

Pain	Intractable pain irrespective of motions and stances (also during night)
	Pain in the lower back
	Pain in the lower limbs
Pain-associated warning clues (red flags)	Fever
	Chills
	Unintended weight loss
Physical impairment	Difficulty with physical functioning
	Reduced mobility
	Lowered dexterity
	Lessened strength
	Thigh hamstrings
Postural alteration	Forward head
	Hyperkyphosis
	Flat back
	Swayback

Oncological treatments

Once a diagnosis of SP due to cancer in young people is achieved, surgery and radiotherapy (RT) are often mandatory, as the above condition may result in high morbidity and a high rate of lower limb paralysis [19]. Surgery usually encompasses posterior-only fusions, anterior lumbar interbody fusions, and combined anterior and posterior fusions procedures [19]. However, it is still being discussed which is the best way to merge spinal surgery and RT to increase the benefits and abate the risks, as well as what are the optimal timing, course, and dose of RT, especially in children [19].

Post-oncological treatments

When an oncological treatment (both surgical and radiotherapeutic) takes place, conservative treatments – including both orthoses and exercise-based therapy – should be adopted, as well as psychological therapies, as a comprehensive way to take care of the young patients with cancer-related SP. These treatment options are detailed below.

1. *Orthoses.* They are devices designed to improve biomechanical function, encourage proper lumbar joint alignment, and protect the spine. The Boston and the Cheneau braces are the most used orthoses as concerns SP and are molded to hold the operated tract of the spine in a straighter and unrotated position, by putting pressure on the outer edge of the curve. These braces limit

excessive motion at the operated segment, helping control pain and lessening damage to the joints, nerves, and muscles [20].

2. *Exercise therapy.* It includes active spinal mobilization aimed at slowly improving the spinal and head joints' mobility, improving spinal deep muscles and head muscles awareness by means of specific techniques for strengthening these muscles, and segmentary stretching involving the lower limb (with special attention to thigh hamstrings) and back muscles. Postural control by active self-correction is also advised by enhancing the motor control of the head, the spine, and the pelvis: concerning the head, exercises aim at adjusting the forward posture of the cranium; regarding the spine, specific movements tailored to children to counteract hyperkyphosis, flatback or swayback are mainly recommended [21]. Children also have to perform balance and walking exercises and be trained in how to change position (e.g., when lying in bed, sitting, or standing). Ergonomic advice is also important to help with the modification of usual daily activities (e.g., avoiding crossing legs, taking brief walks, and switching positions often) [21]. Types of physical exercises for SP are shown in **Tab. 2**.
3. *Psychological therapy.* It consists of cognitive-behavioral techniques that enable to improve mood disorders (i.e., anxiety and depression), catastrophizing, and the fear of movement by guaranteeing gradual reactions to illness behaviors. Young people are encouraged to view their delicate problems as positively as possible.

Table 2. Types of physical exercises^a for the head, spine and limbs for spondylolisthesis (SP), after oncological treatments.

Type	Aim
Mobilization (except the site of SP)	Joint mobility
Strengthening	Muscle strength
Stretching	Elasticity and muscle tone
Postural control	Neuromotor control
Balance	Static and dynamic stability
	Prevention of falls
Walking	Cardiovascular fitness
Ergonomic modifications of usual daily activities	Activities of daily living at home and outside

SP: spondylolisthesis.

^aNote: the exercises are to be tailored and adapted to the young people, and progressively implemented by adopting pacing and graded exposure techniques.

The correct awareness of the problem helps relearning and reacting to frightening thoughts, by increasing their level of activity by means of graded exposure to the situations they may identify as dangerous. The reacquisition of adaptive coping strategies is also targeted by means of communication, motivation, and sharing of the goals to be reached during common activities [22].

A similar program was already undertaken in adults operated for SP, and further evidence in this population should be provided [23].

Multidisciplinary care team

Several healthcare professionals are engaged: those who are directly involved in the clinical management, such as the spinal specialists, as well as the pediatrics, the oncologists, and the psychologists.

The spinal physicians include the spinal surgeons (both orthopedics and neurosurgeons), who surgically treat the children with an SP, and the physiatrists, who are commonly engaged in evaluating postural changes of the column and in delivering the conservative treatment (e.g., with orthoses and planning the exercise program).

The physiotherapists put into practice the indication received by spinal specialists and provide young people with individualized exercises.

Children with SP are also referred to two other essential physicians: oncologists (also including the radio-oncologists), who are part of the care up to post-treatment, and pediatrics, who provide medical care for children and their families.

The clinical psychologist is the health professional who performs psychological evaluations and talk therapy to help young people to learn and better cope with their life, problems, and other people (both peers and adults).

Orthodontists are commonly involved in the cervical posture assessment of young people with SP due to cancer, by evaluating the postural alterations related to teeth and jaw irregularities and temporomandibular disorders (TMDs). Cervical spine posture can be assessed through teleradiographs and cephalometric analysis, which are routinely performed by orthodontists [24].

Limits of the study

This review presents some limits. It does not allow focused comparisons between studies. The matching

between some variables could be problematic and might vary depending on the patient. Moreover, the search method could have been too specific for a scoping inquiry. More studies are needed to deeply analyze the evaluation of postural assets in young people with SP because of a tumor.

Conclusions

This overview reported the most frequent medical assessments of young people with cancer-related SP. Proper clinical and radiological evaluations are needed. Evidence-based multidisciplinary rehabilitation approaches, which primarily include braces, exercise, and cognitive-behavioral therapy, should be provided by the treatment. A multidisciplinary team including spinal specialists, pediatrics, oncologists, psychologists, and orthodontists is necessary to take care of these patients.

Research agenda

- Add evidence of self-administered outcome measures, such as the SRS-22 patient questionnaire, for the assessment of HrQoL in young people with SP due to cancer.
- Improve evidence, possibly through randomized controlled trials, of post-surgical multidisciplinary rehabilitation that incorporates braces, exercise, and cognitive-behavioral therapy for young people with SP due to neoplasms. Adequate follow-ups are expected.
- Add evidence, possibly through randomized controlled trials, to the role of orthodontists as part of multidisciplinary teams, concerning the postural evaluation of the head.

Acknowledgments

- The adolescents, who are a source of optimism and worry.
- The healthcare personnel involved in oncological rehabilitation who, despite their almost always busy days, take charge kindly of children with cancer.
- The orthopedics, neurosurgeons, physiatrists, physiotherapists, psychologists, oncologists, pediatrics, and orthodontics (and the oral surgeons as well) who, despite several clinical activities to carry on every day, should never forget the privilege of caring for young people with cancer.

Data availability statement

Upon request to the corresponding author, the data are available for use.

Declaration of interest

The Authors declare that there is no conflict of interest. Funding: this research received no external funding.

References

1. Lonstein JE. Spondylolisthesis in children. Cause, natural history, and management. *Spine (Phila Pa 1976)*. 1999;24(24):2640-8.
2. Herman MJ, Pizzutillo PD, Cavalier R. Spondylolysis and spondylolisthesis in the child and adolescent athlete. *Orthop Clin North Am*. 2003;34(3):461-7.
3. Logroscino G, Mazza O, Aulisa G, Pitta L, Pola E, Aulisa L. Spondylolysis and spondylolisthesis in the pediatric and adolescent population. *Childs Nerv Syst*. 2001;17(11):644-55.
4. Frosch M, Mauritz MD, Bielack S, Blödt S, Dirksen U, Dobe M, Geiger F, Häfner R, Höfel L, Hübner-Möhler B, von Kalle T, Lawrenz B, Leutner A, Mecher F, Mladenov K, Norda H, Stahlschmidt L, Steinborn M, Stücker R, Trauzeddel R, Trollmann R, Wager J, Zernikow B. Etiology, Risk Factors, and Diagnosis of Back Pain in Children and Adolescents: Evidence- and Consensus-Based Interdisciplinary Recommendations. *Children (Basel)*. 2022;9(2):192.
5. Bayattork M, Sköld MB, Sundstrup E, Andersen LL. Exercise interventions to improve postural malalignments in head, neck, and trunk among adolescents, adults, and older people: systematic review of randomized controlled trials. *J Exerc Rehabil*. 2020;16(1):36-48.
6. Bernstein RM, Cozen H. Evaluation of back pain in children and adolescents. *Am Fam Physician*. 2007;76(11):1669-76.
7. Murphy KP, Sanders C, Rabatin AE. Evaluation and Treatment of the Child with Acute Back Pain. *Pediatr Clin North Am*. 2023;70(3):545-74.
8. Klassen A, Gulati S, Dix D. Health care providers' perspectives about working with parents of children with cancer: a qualitative study. *J Pediatr Oncol Nurs*. 2012;29(2):92-7.
9. Hamill J, Knutzen KM, Derrick T. *Biomechanical Basis of Human Movement*. Philadelphia: Wolters Kluwer, 2014, p. 484.
10. Asher MA, Min Lai S, Burton DC. Further development and validation of the Scoliosis Research Society (SRS) outcomes instrument. *Spine (Phila Pa 1976)*. 2000;25(18):2381-6.
11. Mannion AF, Elfering A, Fekete TF, Harding IJ, Monticone M, Obid P, Niemeyer T, Liljenqvist U, Boss A, Zimmermann L, Vila-Casademunt A, Sánchez Pérez-Grueso FJ, Pizones J, Pellisé F, Richner-Wunderlin S, Kleinstück FS, Obeid I, Boissiere L, Alanay A, Bagó J. Shorter and sweeter: the 16-item version of the SRS questionnaire shows better structural validity than the 20-item version in young patients with spinal deformity. *Spine Deform*. 2022;10(5):1055-62.
12. Monticone M, Baiardi P, Calabrò D, Calabrò F, Foti C. Development of the Italian version of the revised Scoliosis Research Society-22 Patient Questionnaire, SRS-22r-I: cross-

- cultural adaptation, factor analysis, reliability, and validity. *Spine (Phila Pa 1976)*. 2010;35(24):E1412-7.
13. Weiser DA, Kaste SC, Siegel MJ, Adamson PC. Imaging in childhood cancer: a Society for Pediatric Radiology and Children's Oncology Group Joint Task Force report. *Pediatr Blood Cancer*. 2013;60(8):1253-60.
 14. Wood JR, Pedersen RC, Rooks VJ. Neuroimaging for the Primary Care Provider: A Review of Modalities, Indications, and Pitfalls. *Pediatr Clin North Am*. 2021;68(4):715-25.
 15. Kearney J, Bartell AS, Pao M. Psychiatric issues in pediatric oncology: Diagnosis and management. In: Abrams AN, Muriel AC, Wiener L (Eds.). *Pediatric psychosocial oncology: Textbook for multidisciplinary care*. Cham: Springer International Publishing, 2016.
 16. Costantini M, Musso M, Viterbori P, Bonci F, Del Mastro L, Garrone O, Venturini M, Morasso G. Detecting psychological distress in cancer patients: validity of the Italian version of the Hospital Anxiety and Depression Scale. *Support Care Cancer*. 1999;7(3):121-7.
 17. Monticone M, Baiardi P, Ferrari S, Foti C, Mugnai R, Pillastrini P, Rocca B, Vanti C. Development of the Italian version of the Pain Catastrophising Scale (PCS-I): cross-cultural adaptation, factor analysis, reliability, validity and sensitivity to change. *Qual Life Res*. 2012;21(6):1045-50.
 18. Monticone M, Giorgi I, Baiardi P, Barbieri M, Rocca B, Bonezzi C. Development of the Italian version of the Tampa Scale of Kinesiophobia (TSK-I): cross-cultural adaptation, factor analysis, reliability, and validity. *Spine (Phila Pa 1976)*. 2010;35(12):1241-6.
 19. Bian C, Chen N, Li XL, Zhou XG, Lin H, Jiang LB, Liu WM, Chen Q, Dong J. Surgery Combined with Radiotherapy to Treat Spinal Tumors: A Review of Published Reports. *Orthop Surg*. 2016;8(2):97-104.
 20. Gheitasi M, Bayattork M, Kolar MK. Adding corrective exercises along with bracing for postural hyperkyphosis among adolescents: A randomized controlled trial. *PM R*. 2023;15(7):872-80.
 21. Zecirovic A, Bjelica B, Pajovic L, Aksovic N. Postural status and kyphosis in school-age children. *Int J Acad Health Med Res*. 2021;5(11):90-7.
 22. Salley CG, Catarozoli C. Cognitive behavioral therapy in pediatric oncology: Flexible application of core principles. In: Friedberg RD, Paternostro JK (Eds.). *Handbook of Cognitive Behavioral Therapy for Pediatric Medical Conditions*. Cham: Springer Nature Switzerland, 2019.
 23. Monticone M, Ferrante S, Teli M, Rocca B, Foti C, Lovi A, Brayda Bruno M. Management of catastrophising and kinesiophobia improves rehabilitation after fusion for lumbar spondylolisthesis and stenosis. A randomised controlled trial. *Eur Spine J*. 2014;23(1):87-95.
 24. Armijo-Olivo S, Jara X, Castillo N, Alfonso L, Schilling A, Valenzuela E, Frugone R, Magee D. A comparison of the head and cervical posture between the self-balanced position and the Frankfurt method. *J Oral Rehabil*. 2006;33(3):194-201.