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Is scoliosis a source of pain?

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Abstract

Purpose: Pain in scoliosis is definitely a hot topic with growing popularity. The literature remains very controversial, with a pain prevalence ranging from 23% to 90%, but this can be explained by the great heterogeneity of the numerous series. The aim of this review was to report results from the literature regarding pain in relation to scoliosis regardless of the etiology.

Methods: A bibliographic search in Medline and Google database from 2003 to March 2023 was performed. Relevant literature was analyzed, summarized, and discussed based on authors' experience. A 1-year prospective series of adolescent idiopathic scoliosis patients was also included to compare with the existing literature.

Results: A total of 126 adolescent idiopathic scoliosis patients were included, with a mean preoperative Cobb angle of 64.5° (range, 45°–112°). Reported pain prevalence was 34.1%. Pain and no-pain groups were very different in their self-reported experience, with a very low mean visual analogue scale score of 0.5 ($\pm$ 0.6) in the no pain group, while visual analogue scale averaged 5.6 ($\pm$ 1.2) in the pain group ($p < 0.001$). No significant difference was found between groups regarding the most relevant demographic and radiological parameters.

Conclusion: Evidence-based literature on "scoliosis as a source of pain" remains ambiguous. There seems to be a consensus on the lack of direct relationship between deformity magnitude and back pain intensity. A comprehensive evaluation of the patient is therefore necessary before any treatment, including medical history, clinical examination, and relevant imaging for any child with scoliosis and back pain.

Level of evidence: Level VI

Keywords: Idiopathic scoliosis, pain assessment tool, non-idiopathic scoliosis, pain

Part I: adolescent idiopathic scoliosis

Introduction

Adolescent idiopathic scoliosis (AIS) is the most common spinal condition that pediatric orthopedic surgeons have to deal with. There is currently a dogma that AIS is not painful, and that if patients complain about their back, further investigations should be performed to eliminate spinal tumor or cord anomaly (4%–10%). However, in our current practice, pain is often the main symptom leading to AIS diagnosis, and many patients and/or caregivers describe back pain as a main motivation for surgery. In addition, a majority of patients become asymptomatic after posterior fusion.

The original distinction between functional and organic pain has now become meaningless, and several specialists from different horizons have concentrated their efforts to better understand and apprehender it.[1] In scoliosis patients, muscle dysfunction, concave disk pressure, asymmetrical facet joint strains, and high body mass index (BMI) are the main mechanical parameters incriminated, but the analysis of the role of anxiety and self-perception has recently gained popularity.[2] The literature remains controversial to date and not sufficiently clinically relevant for the

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pediatric surgeons who have to manage these patients. In addition, detecting painful patients is for sure a matter of interest, as pain during adolescence has been correlated with pain during adulthood, with professional and economic impact.[3–5] The goal of this article was therefore to summarize the existing evidence on pain in AIS, help identify some predictive factors that should be searched to optimize perioperative management, and report our recent experience with a 1-year prospective series.

Literature review

Materials and methods. A bibliographic search in Medline and Google database from 2003 to March 2023 was performed. The keywords included "adolescent idiopathic scoliosis" and "pain," used individually or in combination. Relevant literature was analyzed, summarized, and discussed based on the author's experience.

Analysis

Pain prevalence in AIS. Pain is definitely a hot topic with growing popularity, with 839 studies found in AIS in the last two decades, and half of them published in the last 5 years. The literature remains very controversial, with a pain prevalence ranging from 23% to 90%, but this can be explained by the great heterogeneity (age, Cobb, pain evaluation method) of the numerous series. The first observation is that pain needs to be better defined, located, and followed over time to be objectively reported.

Based on a wide systematic review of the literature (PubMed, CINAHL, and CENTRAL), Théroux et al.[6] tried in 2017 to estimate the prevalence of low back pain in adolescents followed for idiopathic scoliosis. Their electronic search strategies yielded 1811 unique studies, out of which only 2 fulfilled the eligible criteria. In addition, these two papers were considered at high risk of bias, so the authors concluded that there was insufficient evidence to confidently estimate the prevalence of pain in AIS patients. The study published in 1997 by the Texas Scottish Rite Hospital has for long been considered the reference paper on the topic.[7] In a large series of 2442 adolescents with Cobb angles ranging from 10° to 122°, Ramirez et al. found a pain prevalence of 23%, which was not considered different from that observed in the general population. No correlation was found with curve type and/or magnitude, but the incidence was greater in older adolescents (Risser 2 or more, and >15 years old). This article is still a milestone, but its main limitations were the absence of a control group and the poor quality of pain assessment (definition, intensity). Sato et al. later reported another extensive epidemiological study in 43,630 pupils in Japan and found a comparable rate of pain in AIS (27.5%), although only 51 patients were affected by scoliosis.[8] They also addressed the previous limitations and found an increased risk (odds ratio: 2.29) in case of idiopathic scoliosis compared to controls. Pain location and intensity were evaluated, and one of their surprising results was that 22.6% of the students reported such a high pain level that they had to miss school during the past year. In addition, they showed for the first time that the most frequent locations of pain in AIS were the upper and middle right back (periscapular zone and near thoracic rib hump).

The first pitfall of the literature is that pain definition often varies among the numerous studies. This issue was resolved in adults with spinal deformity, as Mannion et al.[9] showed that a pain level greater than 3 on a numerical rating scale (NRS) was considered clinically relevant (so-called "nonacceptable patient symptom state"). This is therefore the threshold that should also be used in adolescents. With the same definition, Matamalas et al.[10] recently found a 37.1% pain prevalence in a group of 195 AIS and 77 young adults with a Cobb angle >30°. The pain group (PG) showed a significantly higher curve magnitude and age, and more frequent family history of nonspecific back pain. SRS-22 pain and mental health scores were lower in the PG, but function and self-image were not affected. Another interesting conclusion was that the PG showed higher levels of kinesiophobia (fear of movement, assessed by the Tampa scale), anxiety, depression, absenteeism from school, and impact on social environment. However, the same team also emphasized that the exact correlation between kinesiophobia and pain intensity and disability still required further investigation in adolescents.[11]

Pain in AIS surgical candidates. Another way to assess pain level in AIS with less bias is to focus on a subgroup of patients. Smorgick et al.[12] first studied 70 candidates for surgery, excluding AIS patients with moderate and mild curves, in which the exact relation between pain and the spinal deformity can be questioned. Using a visual analogue scale (VAS), they found that 71% of the patients reported some kind of back pain, with 48% of them grading their pain greater than or equal to 5 on the VAS. Older age at diagnosis, no use of brace, and rigid lumbar curve were the only three parameters correlated with pain scores. Unfortunately, NRS and VAS are not parallel scales and assess different aspects of pain, so no direct comparison with previous papers can be performed.[13] In a more recent study on 124 AIS candidates for posterior fusion in Canada, Teles et al.[14] observed a very high pain prevalence, with 90% of patients describing some back pain over the last 6 months (and 85.8% over the last 30 days!), and a 25% medication rate. They also investigated the psychological component of the symptom and found that pain catastrophizing, a poorer self-reported state of mental health, a decreased thoracic kyphosis, and a greater pelvic asymmetry were the four independent risk factors for pain in AIS. Initially defined by Sullivan et al.[15] as an exaggerated negative mindset toward actual or anticipated pain, characterized by magnification, rumination, and helplessness,

Table 1. Comparison between demographic and radiological parameters among the 126 AIS patients' candidates for posterior fusion.

	Pain group (n = 43)	No pain group (n = 83)	p-value
Age (years)	14.9 ± 1.6	15.3 ± 1.1	0.34
BMI (kg/m^2)	20.3 ± 3.4	20.9 ± 3.2	0.63
Main Cobb angle (°)	62 ± 15	65 ± 17	0.41
Coronal tilt (mm)	16 ± 9	17 ± 11	0.79
Maximal thoracic kyphosis (°)	35 ± 13	34 ± 11	0.86
Maximal lumbar lordosis (°)	58 ± 11	64 ± 9	0.14
Pelvic incidence (°)	45 ± 8	50 ± 12	0.34
Sacral slope (°)	39 ± 7	38 ± 6	0.51
SVA (mm)	12 ± 8	13 ± 9	0.69

AIS: adolescent idiopathic scoliosis; BMI: body mass index; SVA: sagittal vertical axis.

"catastrophizing" was relatively ignored by the spine community until recently, but its role in AIS is probably non-negligible.[16] As a matter of fact, the catastrophizing level appeared to be highly correlated with pain intensity (assessed by NRS) in the series of Teles et al., but its role remains controversial in the pediatric population as mentioned by Chidambaran et al.[14,17]

Pain assessment tool. Based on the recent literature, there is now sufficient evidence that pain-level assessment should be included in the latest best practice guidelines before any spinal surgery and that interventions such as cognitive behavioral therapy should be considered in the pain management strategy. The problem is that until now pain doctors had rarely been actively involved in the multidisciplinary approach of AIS. In 2015, Simons et al.[18] presented the pediatric pain screening tool (PPST), modified from the STarT Back Screening Tool in adults, which allows providers to quickly and effectively identify the medium to high-risk youth who will benefit from access to more comprehensive treatments.[19] This 9-item self-report questionnaire helps target during follow-up of the physical (function, pain, sleep quality) and psychosocial (anxiety, depression, catastrophizing) factors that may maintain chronic pain and that can be addressed early in order to improve recovery rates. Narayanasamy et al.[20] recently confirmed the predictive accuracy of PPST for chronic postsurgical pain (CPSP) in a multicentric pediatric study of AIS candidates for surgery, and the cutoff score >2 to define high-risk patients. However, patients with low PPST scores <2 still had a 10%–30% risk of CPSP, emphasizing the complexity of the preoperative evaluation.

Prospective series

Materials and methods. All AIS patients considered for posterior fusion between January 2021 and December 2021 were prospectively included. A 1-day preoperative imaging check-up was performed 4–6 months prior to surgery, and all patients were also evaluated by a single pain doctor using a visual analog scale (VAS) and the SRS-22 score. Data were retrospectively analyzed, and the main demographic (age, BMI) and radiological parameters were compared between painful patients, defined as VAS > 3, and non-painful patients (VAS = 3 or less) using XLSTAT 2022.4 (Addinsoft, Paris, France).

Results. A total of 126 AIS patients were included, with a mean preoperative Cobb angle of 64.5° (range, 45°–112°). Age at evaluation averaged 15.1 years old (± 1.5), and the reported pain prevalence was 34.1%. Interestingly, pain and no-pain groups were very different in their self-reported experience, with a very low mean VAS score of 0.5 (± 0.6) in the no pain group, while VAS averaged 5.6 (± 1.2) in the pain group ($p < 0.001$). No significant difference was found between groups regarding the most relevant demographic and radiological parameters (Table 1). However, results were significantly lower in the pain group regarding global SRS-22 scores (3.4 vs 3.9, $p < 0.001$), but also in the function (3.8 vs 4.2, $p < 0.001$), self-image (2.9 vs 3.6, $p < 0.001$), and mental health domains (3.6 vs 4.0, $p < 0.001$). No significant difference was found in the satisfaction domain (3.2 vs 3.3, $p = 0.22$).

Discussion

Results of the current 1-year perspective series confirm the high prevalence of back pain in AIS patients. They also emphasize the need for a multidisciplinary global and comprehensive approach of pain-level assessment in all adolescents' candidates for surgery, in order to optimize the perioperative management. This measure should be systematic and adopted as early as possible in the patient's journey to improve postoperative outcomes.

The weak correlation between pain scores and curve magnitudes proves again that pain cannot be limited to an organic symptom related to the spinal deformity (Figure 1). Psychosocial parameters play a major role in pain genesis and its chronicity, as shown by the lower SRS-22 scores found in the self-image and mental health domains,

Figure 1. Different curve patterns associated with painful adolescent idiopathic scoliosis (AIS).

and they should be detected and treated before surgery. The new workflow proposed to our AIS patients is therefore a combination of VAS, SRS-22, Peds-QL, and PPST preoperatively (minimum 6 months prior to surgery).[21] In case of VAS > 3 or PPST > 2, patients are now referred and followed by the pain department, using a mobile application, and the date for surgery can sometimes be postponed if necessary (as it can be for a bad skin status). Whether or not this new adjustment will help improve postoperative functional outcomes and the rate of chronic back pain still need to be further studied.

Part II: non-idiopathic scoliosis

Introduction

Non-idiopathic scoliosis does not represent any ICD-10 (International Classification of Diseases, 10th Revision) nosology unity. It is a general term covering a wide spectrum of pathologies comprising (1) a basic disease and (2) a spine deformity developing within a frame of the disease. Non-idiopathic cases are reported to represent about 20% of all scoliotic deformities. Specific conditions exist, such as scoliosis in myelomeningocele, combining congenital, neuromuscular, and postural (prolonged sitting position) causes, or dysplastic type scoliosis in neurofibromatosis combining bony intrinsic deformation and neuromuscular impairment. From a morphological point of view, non-idiopathic scoliosis can develop as a 3D torsional curve pattern similar to AIS; however, other curve patterns can be present, such as a long thoracolumbar curve combined with pelvic obliquity in neuromuscular scoliosis. Back pain can occur in any type of non-idiopathic scoliosis (Table 2). Usually, it does not reveal the major leading symptom, except for reactive scoliosis where pain is the essential cause of the deformity. For most of the non-idiopathic scoliosis types, the trunk deformity itself represents the major symptom and sign, while the back pain remains a secondary complaint.

Literature review

Congenital scoliosis results from bony malformations, mainly the vertebrae embryogenetic defects of segmentation or formation. The rationale of the management of congenital scoliosis consists of the identification of highly progressive curve patterns (e.g. unilateral unsegmented bar with contralateral hemivertebra) and early preventive surgical treatment.[22] Congenital scoliosis is not reported to be associated with back pain in children. Few case reports published on the coexistence of back pain and congenital scoliosis underline the atypical nature of such a combination (unusual curve pattern, unusual pain type, accidental diagnosis in an apparently healthy individual).[23–25]

Neuromuscular (NM) scoliosis prevalence greatly varies among the different pathologies, from 25% in cerebral palsy, 50% in poliomyelitis, 60% in myelodysplasia, 65% in spinal muscular atrophy (SMA), up to 80% in Friedreich ataxia and 90% in Duchenne muscular dystrophy.[26] The rationale of the management of NM scoliosis consists primarily in preserving or restoring the gross motor functions of sitting, standing, or walking.[27] Back pain management remains an important secondary aim, requiring surgical curve correction in selected cases. Correcting the pelvic obliquity can be an important step to achieve mechanical pain release, especially in cerebral

Table 2. Simplified overview on back pain in non-idiopathic scoliosis in children.

Scoliosis type	Curve pattern	Underlying disease	Main clinical problem	Back pain prevalence	Is the deformity a source of pain?	Specific issues
Congenital	Frontal > sagittal	Formation and/or segmentation defect	Risk of curve progression	Very low	No	Early surgery if progressive
Neuromuscular	Single long curve with pelvic obliquity	Upper or lower motoneuron lesion	Loss of gross motor functions	Moderate to high	Probably	Functional over morphological approach
Syndromic	3D double or triple curvature	Genetically defined syndrome	Similar to idiopathic	Low to moderate	Undefined	Underlying disease
Reactive	Single curve, no rotation	Tumor/infection/disk disease	Misdiagnosis	High	No	Cure of the cause needed
Postural	Single frontal curve	Weak trunk muscles, lower limbs discrepancy	Psychological burden if misdiagnosed	Low to moderate	No	Insufficient spine stability, lifestyle
Specific types						
Myelomeningocele	Any type (scoliosis, kyphosis, lordosis)	Spinal dysraphism, vertebral malformations, lower motoneuron lesion	Severe disability	Low	Rare	Neurological and psychological burden
Neurofibromatosis (NF 1)	Angular curve, thoracic kyphoscoliosis	Hereditary developmental defect	Neurological risk	Moderate	Possible	Surgery

palsy patients.[28] On the other hand, due to a high perioperative morbidity, the surgical correction of NM scoliosis may also expose the patient to postoperative back pain in case of protruding implants, uncontrolled spasticity, or late infection. Recent achievements in medical and genetic SMA cures will probably dramatically improve both scoliosis prevalence and back pain in SMA.[29] A distinct subgroup of NM scoliosis is represented by the apparently idiopathic curves revealing intracanalar magnetic resonance imaging (MRI) abnormalities (Chiari malformation, syringomyelia, tethered spinal cord). Such findings modify the diagnosis from idiopathic to neurogenic spinal deformity. These patients can express atypical complaints around the spine, such as back pain, leg pain, numbness, tremor, or weakness.[30] Surgical treatment of central nervous system pathology may be beneficial for both the symptoms and curve progression rate.[31]

Syndromic scoliosis represents a heterogeneous group, associated with specific orthopedic issues typical for the underlying disease. The subgroup of connective tissue disorders (Ehlers-Danlos, Willy-Prader, Larsen) presents generalized joint hypermobility. The back pain can accompany the spinal deformity and is usually attributed to insufficient soft tissue stability around the vertebral column. The patients reveal slim, weak, and hypermobile while the spinal curvatures tend to become stiff. The back pain is musculoskeletal in type, diffused, of mechanical origin, and alleviated with rest. Depending on curve and pain severity, stabilizing physiotherapy, orthosis, or spinal instrumentation with fusion can be effective in the management of back pain.

Reactive scoliosis relates to a spinal curvature which is secondary to the paravertebral muscles' spasm, caused by a local spine pathology (dura irritation by tumoral, inflammatory, infectious, or mechanical stimuli). Scoliosis itself does not represent the source of back pain. Lumbar disk disease, benign bony tumor (osteoid osteoma), or discitis/spondylomyelitis are examples of the diagnosis. The scoliosis onset is usually rapid, noted by the family. The pattern is predominantly a single frontal curvature without significant rotation. On clinical examination, an unusual curve pattern and spine stiffness are typical (Figure 2). Back pain of moderate to severe intensity may be a leading symptom, and the pain location is usually precisely shown by the child. The symptoms are typically not relieved with bed rest or exercises; however, anti-inflammatory medication is effective. Reactive scoliosis requires an etiological approach (investigation type) and adequate treatment which can be surgical. Once the intraspinal pathology is cured (disk or tumor resection, infection management), pain is usually relieved, and the secondary spinal curvature tends to diminish. However, delayed diagnosis may lead to curve structuralization.[32]

Postural or functional scoliosis represents another category, associated with a mild non-structural spine curvature developing because of various spinal (weak

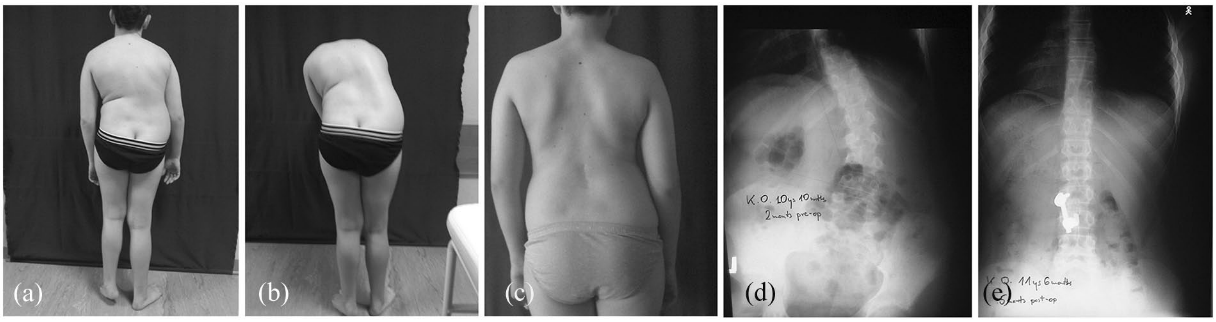

Figure 2. A 11-year-old boy with a 6-month history of stiff and painful lumbar spine presenting a right-sided thoracolumbar curvature with no rotation or compensatory curve (a–c), initially managed as "scoliosis." Left sublaminar L2 osteoid osteoma of 1.5 cm diameter was identified on scintigraphy and CT scans, and was uneventfully resected; the left lamina reconstruction using local bone graft stabilized by a left L2 pedicular screw connected to sublaminar hook placed on the same L2 vertebra was performed with the aim of avoiding fusion of any articulation. Surgery followed by a 3-month cast immobilization resulted in resolved pain and scoliosis, restoring a full range of motion at a 5-month follow-up (d and e).

postural muscles) or distant (pelvic obliquity due to lower limbs discrepancy) causes.[33] The trunk asymmetry is easily noted in standing position; however, in opposition to idiopathic scoliosis, minimal or no rotation is detected on forward bending using a scoliometer. Radiographs may be misleading because they demonstrate a lateral spine inclination, often more than 10° Cobb, which triggers an erroneous diagnosis of structural scoliosis.[34] Postural scoliosis is mainly related to weak postural muscles and a sedentary lifestyle, and this is why it often generates moderate mechanical back pain. Management with "core stability" exercises, such as transverse abdominis strengthening, combined with recreational sport, is capable of fully resolving back pain and improving spinal alignment.

Type 1 neurofibromatosis (von Recklinghausen disease) may be separately discussed to mention an atypical pain pattern or tenderness occurring on thorax palpation or percussion. It is provoked by rib head displacement into the vertebral canal through the enlarged intervertebral foramen and was previously reported as a "painful rib hump."[35] Surgical treatment consists in resecting the protruding rib head and neck, associated with instrumented spinal fusion (Figure 3).

Discussion

Back pain remains an imprecise term that should be better defined with a more precise description of the location, onset, frequency, intensity, and nature. Postural spine malalignments as well as structural scoliotic curves may provoke moderate, diffused, myofascial back pain. However, the majority of children or adolescents with scoliosis do not suffer from back pain if regular physical activity is provided. Limited evidence of increased scoliosis prevalence under intensive training mobilizing the vertebral column (rhythmic gymnasts, ballet dancers) has been published. However, a combination of physical activity with generalized joint hypermobility and delayed puberty was a prerequisite observed in this particular group.[36]

The cause-effect relationship between deformity and pain is not certain nor direct in any type of scoliosis.[37] Due to the diversity of curve patterns, a mechanism of pain generation is difficult to propose. Articular overloading, muscular hypertension, lack of spine stability due to soft tissue quality, or other mechanisms can be discussed but need to be further studied and confirmed.

In conclusion, evidence-based literature on "scoliosis as a source of pain" remains ambiguous. There seems to be a consensus on the lack of direct relationship between deformity magnitude and back pain intensity. A comprehensive evaluation of the patient is therefore necessary before any treatment, including medical history, clinical examination, and relevant imaging for any child with scoliosis and back pain. If non-idiopathic scoliosis is suspected, a detailed investigation looking for the underlying disease should be performed. However, back pain is rarely a major clinical problem to be managed in non-idiopathic patients. On the other hand, in the absence of an evident non-idiopathic etiology, any painful "idiopathic" scoliosis should be considered as a "red flag" and trigger a diagnostic pathway. Back pain is frequently reported in AIS but remains poorly understood. Psychosocial factors clearly play a significant role in the genesis and tolerance of the symptom, and they should be properly addressed if surgery is considered. Best Practice Guidelines should now consider pain management as a crucial preoperative measure to optimize functional outcomes and patients' satisfaction.

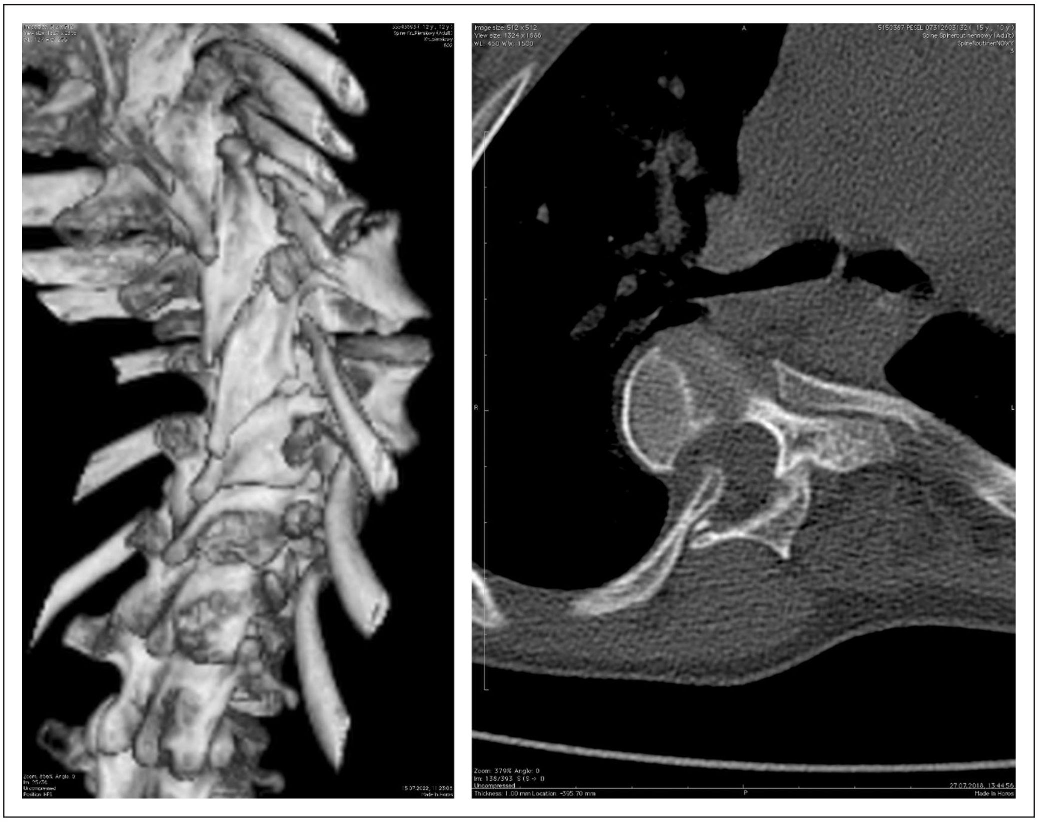

Figure 3. Painful rib head protrusion in a NF1 patient.

Author contributions

B.I. contributed to conception and design. A-L. S. and M.S. contributed to administrative support. B.I. and T.K. contributed to provision of study material and patients. A-L. S. and M.S. contributed to collection and assembly of data. B.I. and T.K. contributed to data analysis and interpretation. All authors contributed to manuscript writing and final approval of the manuscript.

Declaration of conflicting interests

The author(s) declared the following potential conflicts of interest with respect to the research, authorship, and/or publication of this article: Professor B.I. is a consultant for Implanet and Medtronics and perceives royalties from ZimmerBiomet.

Ethical statement

The study was conducted in accordance with the Declaration of Helsinki and was approved by the Ethics Committee from Robert Debré Hospital (Comité d'évaluation de l'éthique des projets de recherche de Robert Debré, CEER-RD) (no 2022-621ter) on 18 July 2023, with an exemption from informed consent. No specific consent is needed for statistical analyses of aggregated de-identified data. For this study, the raw data were first extracted from HIS, and patients' identities, including names, screening IDs, patient IDs, and mobile phone numbers, were de-identified.

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Availability of data and materials

The datasets generated during and/or analyzed during the current study are available from the corresponding author on reasonable request.

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