CASE REPORT

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Challenges in the assessment and management of pain in non-verbal autistic children: a case report of an osteochondroma revealed by self-aggressive behavior

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ABSTRACT

Introduction: To highlight the challenges of identifying and managing pain in non-verbal autistic children through a case of osteochondroma revealed by self-aggressive behavior and to propose a structured diagnostic approach for these complex situations.

Case description: We report the case of a 14-year-old girl with severe non-verbal autism spectrum disorder who developed a progressive increase in self-aggressive behavior over 20 months. The evaluation required an in-depth functional behavioral analysis and an adapted clinical examination, complemented by a radiological work-up. Behavioral analysis revealed that the function of the behavioral problems pointed towards an organic origin: an increase in behavior when moving around, a protective limping when dodging with an exacerbation in the evening. The assessment identified a femoral osteochondroma. The introduction of an alternative communication system with analgesic treatment led to an 80% reduction in self-aggressive behavior. Our observation highlights the difficulties in identifying pain in non-verbal ASD patients and the importance of a structured diagnostic approach. The development of alternative communication appears to be a key element in the assessment and management of pain.

Conclusion: Pain management in non-verbal ASD patients requires particular vigilance and a systematic diagnostic approach. Early implementation of alternative communication tools is essential to optimize their assessment and treatment.

Keywords: *Autism spectrum disorder; Self-injurious behavior; Pain assessment; Nonverbal communication; Osteochondroma*.
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INTRODUCTION

Pain assessment and management in non-verbal patients with autism spectrum disorder represent significant clinical challenges, with potentially severe consequences when diagnosis is delayed. Recent epidemiological data indicate that 25 to 35% of individuals with autism spectrum disorder (ASD) are non-verbal or minimally verbal[1], creating a substantial population at risk for underdiagnosed and undertreated pain conditions.

The complexity of pain assessment in this population is multifactorial. First, the sensory processing particularities associated with ASD can mask traditional pain manifestations. Studies have shown that 45 to 65% of individuals with ASD demonstrate atypical pain responses[2], ranging from apparent hyposensitivity to paradoxical reactions. Second, the communication barriers inherent to non-verbal ASD significantly impede standard pain assessment methods, with research indicating that up to 70% of pain episodes may be missed in initial evaluations[3].

Orthopedic conditions, particularly osteochondromas, present a unique diagnostic challenge in this population. Recent studies report that ASD patients have a 20 to 25% prevalence of orthopedic comorbidities[4], significantly higher than in the general pediatric population. Osteochondromas, while being the most common benign bone tumors in adolescents (35% of benign bone tumors)[5], often go undiagnosed in non-verbal ASD patients due to atypical pain presentation.

The relationship between undiagnosed pain and self-injurious behavior (SIB) is particularly concerning. Studies indicate that 50 to 70% of severe SIB cases in ASD may be linked to underlying pain conditions.[6] The mortality risk associated with severe SIB is estimated at 0.7 to 2% in cases where the underlying cause remains untreated.[7] Additionally, chronic pain from undiagnosed osteochondromas can lead to permanent joint dysfunction (15-20% of cases), decreased mobility and participation in daily activities, increased risk of depression and anxiety, compromised educational and therapeutic interventions.

The diagnostic delay in identifying painful conditions in non-verbal ASD patients averages 15.2 months[8], significantly longer than in neurotypical populations. This delay can result in progressive worsening of behavioral

Table 1. Functional Analysis of Self-Injurious Behaviors (ABC Method)

Antecedents (A)	Behaviors (B)	Consequences (C)	Identified Pattern
Getting out of bed	Mild self-hitting	Avoidance of right-sided weight-bearing	More pronounced after prolonged rest
Prolonged walking	Intense head self-hitting	Activity cessation	Increase physical effort
Prolonged sitting	Moderate self-aggressive behavior	Position change	Positional discomfort
Climbing/descending stairs	Screaming and violent self-hitting	Stair avoidance	Mechanical pain
End of day	Increased self-aggression	Bed rest	Cumulative fatigue and pain
Dressing/undressing	Focused self-hitting	Limb protection	Pain on manipulation
Toileting/hygiene	Agitation and self-hitting	Right side avoidance	Pain on contact

symptoms, increased risk of complications, reduced treatment efficacy, heightened family distress,, and caregiver burden.

The case we present illustrates these challenges through the experience of a non-verbal adolescent with ASD whose osteochondroma was revealed through progressive worsening of self-aggressive behavior. This observation highlights the critical importance of systematic pain assessment and the potential value of alternative communication strategies in improving diagnostic accuracy and treatment outcomes in this vulnerable population.

CASE DESCRIPTION

A 14-year-old girl was referred to our department with a history of severe autism spectrum disorder diagnosed at age 3. Her condition was characterized by the complete absence of verbal language (DSM-5 severity level 3) and a history of well-controlled epilepsy on sodium valproate (30mg/kg/day), with no seizures for 7 years.

The current episode began in September 2021, following her older sister's death, with the onset of self-aggressive behaviors of initially moderate intensity (5-10 daily episodes). While the initial analysis suggested a psychological origin related to bereavement, an in-depth functional behavioral assessment revealed subtle but significant signs pointing towards an organic cause: a slight modification in gait pattern, initially imperceptible without structured observation, a progressive tendency to avoid certain positions, and intensification of self-aggressive behaviors following a specific temporal pattern.

The behavioral deterioration followed a significant progression over 20 months:

- Initial phase (September-December 2021): 5-10 daily episodes of moderate intensity
- Intermediate phase (January-May 2022): 10-15 episodes/day with increasing intensity
- Severe aggravation phase (June-October 2022): 15-20 episodes/day, high intensity, characterized by Major sleep disruption, intermittent food refusal, significant impact on daily activities, and Growing family exhaustion

Systematic functional behavioral analysis (ABC method) was crucial in identifying patterns suggesting an organic rather than purely behavioral origin:

The systematic use of the NCCPC-R (Non-Communicating Children's Pain Checklist-Revised)[9] during successive evaluations highlighted subtle but constant pain indicators: subtle changes in facial expression during certain movements (partial score 8/15), sleep modifications (partial score 6/12), and variations in usual vocalizations (partial score 7/12), resulting in a total score of 28/54, suggesting moderate to severe pain. The evolution of NCCPC-R scores clearly demonstrated pain progression:

Clinical examination, particularly challenging in this non-communicating patient, required a progressive approach over several weeks, establishing positive "pairing" in the parents' presence. This gradual approach revealed a subtle gait asymmetry and a palpable mass on the right distal femur's medial aspect, initially masked by behavioral opposition.

The initial neurological investigations included an EEG under mild sedation, showing no epileptiform activity and normal background rhythms, and a brain MRI revealing no structural abnormalities or signs of increased intracranial pressure.

Figure 1. Standard radiographs of the right knee, showing the characteristic metaphyseal osteocondensal pattern.

Further targeted examinations of the right lower limb were guided by functional assessment and NCCPC-R scores.

Complementary investigations showed normal complete blood count, C-reactive protein (3 mg/L), and slightly elevated alkaline phosphatase (250 U/L). Imaging studies performed under mild anxiolytic premedication included standard radiographs revealing a metaphyseal osteocondensal pattern (figure1). CT scan confirmed a 2.8cm sessile exostosis without complications (figure 2).

Following the diagnosis of osteochondroma, a comprehensive therapeutic strategy was implemented, including systematic analgesic treatment with paracetamol (15mg/kg/dose every 6 hours) and the introduction of an augmentative and alternative communication (AAC) system. The AAC implementation required a structured two-month learning period:

- Weeks 1-2: Familiarization with visual supports
- Weeks 3-4: Progressive introduction of pain pictograms
- Weeks 5-6: Learning of adapted visual analog scale
- Weeks 7-8: Integration of tools into daily routine

Figure 2. CT scan of the right knee showing femoral metaphyseal osteochondroma. The various sections (multiplanar reconstructions) show a 2.8cm sessile exostosis, well-circumscribed, with no signs of complications.

At three-month follow-up, significant improvement was observed with an 80% reduction in self-aggressive behavior (decreasing to 2-3 episodes/day), improved NCCPC-R scores (12/54), and successful use of AAC tools for pain communication. Sleep patterns and daily activity participation showed marked improvement, with stable osteochondroma size on follow-up imaging.

This case highlights three essential points: (1) the necessity of increased vigilance for subtle pain signs in non-communicating patients, (2) the importance of structured and repeated functional behavioral assessment, and (3) the crucial role of adequate time for effective implementation of an adapted AAC system.

The methodical approach in identifying and interpreting subtle signs, combined with the the systematic use of standardized assessment tools, enabled the diagnosis of osteochondroma despite atypical clinical presentation. The therapeutic success underscores the importance of rigorous functional evaluation and progressive implementation of alternative communication tools in non-verbal ASD patients presenting with behavioral changes.

DISCUSSION

Our clinical observation highlights the complexity of the etiological diagnosis of self-aggressive behaviors in non-verbal ASD patients. The 20-month diagnostic delay before identifying the osteochondroma raises fundamental questions about pain assessment in this population.

Recent studies by Courtemanche et al.[6] have shown that 50-70% of self-aggressive behaviors in ASD patients are linked to unidentified underlying pain, contrasting

Table 2. Clinical Parameters Evolution

Clinical Parameters	Initial Phase (09/2021-05/2022)	Worsening Phase (06/2022-10/2022)	Post-therapy Phase (12/2022-03/2023)
Self-aggressive behavior	5-10 episodes/day, moderate intensity	15-20 episodes/day, high intensity	2-3 episodes/day, low intensity
Locomotor signs	Subtle gait changes	Evident limping, right lower limb protection	Progressive normalization
Functional signs	Minimal	Evening exacerbation, pain on mobilization	Significant improvement
Functional impact	Mild	Impaired daily activities, sleep disorders	Return to usual activities

with the traditional approach that favored behavioral explanations. Our case confirms these findings by revealing that systematic functional analysis can identify specific patterns suggestive of an organic cause. This analytical approach aligns with Moore's work[8], which emphasizes the crucial importance of detailed temporal and contextual behavioral analysis.

Ruelle-Le Glaunec et al.[2] described a characteristic dissociation between pain perception and expression in ASD patients, with hyperreactivity to painful stimuli paradoxically associated with atypical behavioral manifestations. The systematic behavioral assessment, in our case, enriches this theory by demonstrating how these atypical manifestations can be identified through specific patterns: temporal correlation with physical activities, circadian evolution of behavioral manifestations, and characteristic postural avoidance behaviors.

The positive impact of post-diagnostic Alternative and Augmentative Communication (AAC) in our case aligns with Ely et al.[7] observations on problem behavior reduction. Our experience extends these findings by demonstrating that AAC effectiveness depends on early introduction and progressive adaptation to patient capabilities. This approach led to an 80% reduction in self-aggressive behaviors, significantly exceeding the 40-50% improvement rates previously reported in standard behavioral interventions[3].

The prevalence of orthopedic comorbidities in ASD patients (20-25%) reported by Muskens et al[4] underscores the need for increased vigilance. Our case illustrates how osteochondromas, while being the most common benign bone tumor in adolescents[5], can be diagnosed late without systematic pain assessment. This delay mainly affects non-verbal patients, who represent 25-35% of the ASD population[1], highlighting a significant healthcare disparity in pain recognition and management.

Our findings have significant implications for clinical practice. The successful integration of behavioral analysis with AAC tools demonstrates the potential for improving pain assessment in non-verbal ASD patients. This approach bridges the gap between pain perception and expression described in recent neurophysiological studies[2], offering a practical solution to a long-standing clinical challenge.

Several limitations must be considered in interpreting our results. First, being a single case study, the generalization of conclusions requires validation in a larger cohort. Second, the absence of standardized AAC tools from the beginning of care may have contributed to the diagnostic delay. Third, the impact of family bereavement on behavioral pain expression could not be fully evaluated. Fourth, the relative effectiveness of different AAC modalities in this specific context remains to be determined through comparative studies. Finally, the potential influence of concurrent medical treatments, particularly the long-term use of sodium valproate, on pain expression patterns warrants further investigation.

This case emphasizes the need for enhanced vigilance in pain assessment among non-verbal ASD patients. It suggests that a systematic diagnostic approach, combining detailed behavioral analysis with appropriate communication tools, can significantly improve outcomes. Our experience calls for further research into standardized assessment protocols and the development of specialized training programs for healthcare providers working with this vulnerable population.

CONCLUSION

Our experience highlights three essential points for clinical practice. Firstly, the need for increased vigilance regarding somatic causes in the face of worsening behavioral disorders. Secondly, the importance of a rigorous functional analysis to identify patterns suggestive of an organic origin. Thirdly, the crucial role of alternative communication tools in the expression and management of pain. The therapeutic success observed in this case demonstrates that a structured approach, combining appropriate somatic assessment and the early introduction of alternative communication tools, can significantly improve these patients' quality of life. This approach requires close collaboration between healthcare professionals and families, as well as specific training for healthcare teams.

DISCLOSURES

Authors Contributions

- LAARAJE Azzeddine (AL): Study conception, data collection, manuscript writing, clinical examination and follow-up
- RADI Abdelilah (RA): Data analysis, critical revision of the manuscript, behavioral assessment
- OURRAI Abdelhakim (OA): Clinical supervision, manuscript revision, therapeutic management
- HASSANI Amal (HA): Data collection, radiological analysis, literature review
- ABILKASSEM Rachid (AR): General supervision, final validation, coordination of multidisciplinary care.

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Conflict of Interest

The authors declare that they have no competing interests or financial relationships relevant to this article to disclose.

Consent for Publication

Written informed consent was obtained from the patient's parents for publication of this case report and accompanying images.

REFERENCES

1. Wodka, E. L., Mathy, P., Kalb, L. (2013). Predictors of phrase and fluent speech in children with autism and severe language delay. Pediatrics, 131(4):e1128-34. Available from: https://doi.org/10.1542/peds.2012-2221.
2. Ruelle-Le Glaunec, L., Inquimbert, P., Hugel, S., Schlichter, R., Bossu, J. L. (2021). Nociception, douleur et autisme. Med Sci (Paris), 37(2):141-151. Available from: https://doi.org/10.1051/medsci/2020280.
3. Dubois, A., Michelon, C., Rattaz, C., Zabalia, M., Baghdadli, A. (2017). Daily living pain assessment in children with autism: Exploratory study. Res Dev Disabil, 62:238-246. Available from: https://doi.org/10.1016/j.ridd.2017.01.003.
4. Muskens, J. B., Velders, F. P., Staal, W. G. (2017). Medical comorbidities in children and adolescents with autism spectrum disorders and attention deficit hyperactivity disorders: a systematic review. Eur Child Adolesc Psychiatry, 26(9):1093-1103. Available from: https://doi.org/10.1007/s00787-017-1020-0.

5. Garcia, S. A., Ng, V. Y., Iwamoto, M., Enomoto-Iwamoto, M. (2021). Osteochondroma Pathogenesis: Mouse Models and Mechanistic Insights into Interactions with Retinoid Signaling. Am J Pathol, 191(12):2042-2051. Available from: https://doi.org/10.1016/j.ajpath.2021.08.003.
6. Courtemanche, A. B., Black, W. R., Reese, R. M. (2016). The Relationship Between Pain, Self-Injury, and Other Problem Behaviors in Young Children With Autism and Other Developmental Disabilities. Am J Intellect Dev Disabil, 121(3):194-203. Available from: https://doi.org/10.1352/1944-7558-121.3.194.
7. Ely, E., Chen-Lim, M. L., Carpenter, K. M., Wallhauser, E., Friedlaender, E. (2016). Pain Assessment of Children with Autism Spectrum Disorders. J Dev Behav Pediatr, 37(1):53-61. Available from: https://doi.org/10.1097/DBP.0000000000000240.
8. Moore, D. J. (2015). Acute pain experience in individuals with autism spectrum disorders: a review. Autism, 19(4):387-99. Available from: https://doi.org/10.1177/1362361314527839.
9. Breau, L. M., McGrath, P. J., Camfield, C. S., Finley, G. A. (2002). Psychometric properties of the non-communicating children's pain checklist-revised. Pain, 99(1-2):349-57. Available from: https://doi.org/10.1016/s0304-3959(02)00179-3.