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Pain Expression in Children Suffering from Autism

التعبير عن الألم عند الأطفال المصابين باضطراب طيف التوحد

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Abstract:

Pain expressed by children with autism spectrum disorders (ASD) is now left aside because of the fact that only little research has investigated this topic. The objective of this research is to identify how do children with ASD express pain. Concerning the materials and methods, we conducted a cross-sectional study on 12 children with ASD in the internal medicine department at MAALEM Lahcen hospital in Ghazaouet. All subjects underwent venipuncture. Reactivity to pain was assessed by a pain assessment grid inspired by GED DI and CFCS. We measured the intensity of ASD in children using the CARS (E, Schoppler, 1971), and their developmental level using the Vineland-I (VABS, E, Doll & S, Sparrow). Results showed that the responses of children with ASD to painful stimulation were distributed as follows: Facial responses 49 percent, verbal responses 32 percent, and behavioral responses 19 percent. To conclude, pain expression in children with ASD is most of the time hypo-reactive in response to significant child deficiency in terms of communication and interaction.

Keywords: *Autism Spectrum Disorders; Children; Pain; Expression; Assessment.*

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ملخص :

إن فرضية عدم حساسية الاطفال المصابين باضطراب طيف التوحد اتجاه الألم تم تفنيدها حاليا من خلال البحوث الاخيرة. الهدف من هذه الدراسة هو التعرف على كيف يعبر الأطفال المصابون باضطراب طيف التوحد عن

الألم . المواد والأساليب: أجرينا دراسة على 12 طفل مصاب باضطراب طيف التوحد تمت متابعتهم في قسم الطب

الداخلي بمستشفى معلم لحسن بالغزوات. خضع جميع الأطفال لبزل الوريد، وتم تقييم التفاعل مع الألم من خلال استمارة لتقييم الألم مستوحاة من GED- DIو CFCS .قمنا بقياس شدة التوحد لدى الأطفال بإستخدام(CARS(E,Schppler,1971. ومستوى تطورهم بإستخدام Doll ,E ,Vineland_I(VABS) ,Sparrow

تم توزيع استجابات الأطفال المصابين بالتوحد على النحو التالي التالي:تعابير الوجه 49٪ ، استجابات لفظية 32٪ ، استجابات سلوكية 19٪. خاتمة: التعبير عن الألم عند الأطفال المصابين بالتوحد غالبًا ما يكون رد فعل ناقصًا استجابة لنقص كبير لدى هؤلاء الاطفال من حيث التواصل و التفاعل.

الكلمات المفتاحية : اضطراب طيف التوحد؛ الأطفال؛ الألم؛ التعبير؛ التقييم.

1. Introduction

Contrary to popular belief spread 30 years ago, children with autism spectrum disorders (ASD) experience pain in the same way as typical neuro subjects. However, they express pain differently from typical neuro children due to their neurodevelopmental specificity.

The fifth edition of the Diagnostic and Statistical Manual of Mental Disorders, DSM-5, classifies autism pectrum disorder as neurodevelopmental disorders.it is characterized by a persistent deficiency in social communication and social interactions in differents contexts, restricted, repetitive patterns of behavior, interests or activities. The last criterion includes “Hyper or hypo reactivity to sensory stimuli or un usual interest in sensory elements of the environment (e.g. Indifference to pain ……)” (American Psychiatric Association, 2013)

The prevalence of these sensory peculiarities is estimated at 90% of the population of ASD . They include all sensory modalities and can be hyper-sensitivities, hypo-sensitivities or, paradoxical sensitivities (Sahnoun & Saravane, 2015).

The Autism and Developmental Disabilities Monitoring (ADDM) program of the Centers for Disease Control and Prevention (CDC) estimates the prevalence of ASD in 2014 at 16.8 / 1000 in 8-year-old children (Baio, Wiggins, &

Christensen, 2018). Boys seem to be more affected than girls (ratio of 4 to 5 boys to 1 girl, according to studies (Looms, Hull, & Mandy, 2017).

The intellectual abilities of about 44% of children with ASD are considered as being average to above average. There is no direct cause for ASD, and most researchers asserted that it is probably caused by acombination of genetic and environmental factors (Guinchat, 2014). Autism spectrum disorders (ASD) have a multi factorial origin, with a strong implication of genetic factors (Looms, Hull, & Mandy, 2017).

Pain is defined by the International Association for the Study of Pain as "An unpleasant sensory and emotional experience associated with, or resembling that associated with actual or potential tissue damage". However, since this definition emphasizes self-assessment and an individual's ability to communicate verbally, the IASP clarified this definition by recognizing that "the inability to communicate verbally does not in any way negate the possibility that an individual experiences pain and needs appropriate treatment to feel relieved" (IASP). It is a complex phenomenon that involves different components: nociceptive, cognitive-behavioral, motivo-affective and, sensory discriminative.

Children with ASD are likely to have comorbidities, common sources of pain in these children, which are primarily: (Courtemanche & Black, 2016)

- Abdominal pain: severals tudies have shown a high prevalence of gastro-intestinal symptoms in childre nwith ASD when compared to children without ASD, with an odds ratio of 4.5 (Mc Elhanson, Mc Cracken, Karpen, & Sharp, 2014). The most common symptoms are constipation, vomiting, aerocolia, food intolerance, regurgitation, gastroesophageal reflux disease, gastritis or, esophagitis.

-Dental or gingival pain related to abscesses, cavities or, tartar.

- ENT pain due to medium or external chronic otitis ;

- Headaches and facial pain that may be related to an ENT or ophthalmological pathology.

- Period pains or premenstrual syndrome.

- Pain related to epileptic seizures, mostly partial and atypical. The prevalence of seizure disorders in the population of children with ASD is higher than in the general population. Spence and Schneider (Spence & Schneider, 2009) report a rate between 7 and 46% (Saravane & Mytych, 2017).

Accurate pain assessment is necessary to provide an appropriate intervention (Courtemanche & Black, 2016). Till now, few studies have been conducted on pain expression in this population.

Some researchers have highlighted the factor of hypo responsiveness to pain. Thus, several behavioral studies have suggested that children with ASD are associated with a reduced pain sensitivity, which is consistent in clinical observations (Allely, 2013).

The evidence supporting this reduction in pain response in children with ASD can be found in a series of case studies (Wing 1976, 1996), as well as the self-report of adults with high-level ASD (Minshew and Hobson 2008), and parent reports (Klintwall et al. 2011; Militerni et al. 2000) .(Garcia-Villamisar, Moore, & Garcia-Martinez, 2019).

Tordjman et al (Tordjman, Anderson, Botbol, &al., 2009) show a decrease in the behavioral reactivity of children with ASD compared to a control group during venipuncture. Likewise, Pernon and Rattaz (Pernon&Rattaz, 2003) found that children with ASD exhibit a decrease in facial reactivity during painful stimulation compared to normal children. (Spence & Schneider, 2009).

To other researchers, hyperreactivity is the factor that is linked to the singular expression of pain in children with ASD.

However, in others tudies using venipuncture procedures, people with ASD appear to show a greater behavioral and physiological response, which suggests that pain may be felt more intensely (Nader et al. 2004; Rattaz et al. 2013 ; Tordjman et al. 2009) . (Spence & Schneider, 2009).

Moreover, experimental pain induction procedures have suggested that people with ASD have the same or lower threshold for pain (Bird et al. 2010; Cascio et al. 2008; Duerden et al. 2015). (Garcia-Villamisar, Moore, & Garcia-Martinez, 2019) .

2. Explanatory Models:

Many explanatory models have been evoked to explain the difference between the feeling and expression of pain in people with ASD. They are three:

-The approach that favors neuro-chemical hypotheses, which, particularly, implies the existence of a dysfunction of the opioid system or the serotoninergic system in children with ASD (S, Tordjman; Antoine, C, 1999).

-Developmental approach that takes into consideration the specific socio-communicative capacities of children with ASD: the existence of a link between

the quality of pain expression and children's level of development,(Pernon & Rattaz, 2003) .

-The absence of a possible social referencing, a possible adapted communication, and the difficulties faced in regulating the expression of negative emotions may explain the atypical expressions of pain (Nader, Oberlander, Chambers, & Craig, 2004), (Dubois, Michelonc, Rattaz, Zabalia, & Baghdadli, 2017).

Integrated models are also proposed, by taking into consideration the different hypotheses involving intra-personal (sensory alterations, self-mutilations, speech disorder) and environmental factors (the observer's ability to understand the message) concerning the feeling and expression of pain (Nader, Oberlander, Chambers, & Craig, 2004).

3.-Evaluation of pain in children with ASD

The disruptions in non-verbal communication and social interaction seen in children with ASD regarding all aspects of life; interfere with the ability of this population to effectively use traditional pain assessment tools (Ely, Chen-Lim, Carpenter, Wallhauser and Friedlaender, 2016). To children with ASD, who are unable to accurately verbalize and describetheir pain, assessing the pain experience requires behavioral assessment tools (Dubois, Rattaz, Pry, & Baghdadi, 2010).

No tool has been validated to be specifically used to assess pain in children with ASD. Nevertheless, somescales have been validated for children unable to communicate verbally and suffer from cognitive impairments: the pain checklist for non-communicative children (NCCPC; Breau, McGrath, Camfield, Rosmus &Finley, 2002) and the Face, Legs, Activity, Cry, ConsolabilityScaleRevise (FLACC-R; Malviya, Voepel-Lewis, Burke, Merkel & Tait, 2006) which included children with ASD (between 12% and 16%) in validation studies. Only the NCCPC has been validated in English (Breau et al., 2000,2001; Breau, Finley, and al., 2002; Breau, McGrath, and al., 2002; Massaro et al., 2014; Voepel-Lewis et al., 2008) and in French (Pain assessment grid – Intellectual Disability; GED-DI; Zabalia and al., 2011). This scale is validated to assess pain in a post-operative context. The English version is also reliable to assess every day life pain when it comes to people with severe cognitive impairment (Dubois, Rattaz, Pry, & Baghdadi, 2010).

4.- Experimental

The current investigation is a monocentric, cross-sectional descriptive study that included 12 children with ASD, and carried out at the level of the internal

medicine department of MAALAM LAHCEN hospital from January 2018 to January 2019.

The sample of children with ASD under study included ten boys and two girls whose chronological ages are between 5 and 9 years old.

The diagnosis of ASD was made using the international classification of diseases (ICD 10) according to the criteria of the "Diagnostic and Statistical Manual of Mental Disorders" in itsfifth version (DSM V). ADIR (Autism Diagnostic Interview-Revised) and the ADOS scale (Autism Diagnostic Observation Schedule).

The severity of ASD was assessed using the Childhood Autism Rating Scale (CARS), and cognitive profile assessment was performed using measurement of children's developmental level using the Vineland-I (VABS, E, Doll& S, Sparrow). This scale is a hetero-assessment that measures adaptive behaviors. Faced with the heterogeneity of cognitive performance in children with ASD and the interpretability of an IQ (intelligence quotient) or QA (adaptive quotient) score (Pernon&Rattaz, 2003).

All the children were subjected to the same nociceptive situation, a venipuncture. The blood sample was taken by two experienced nurses, in the presence of the parents.

An explanation of the purpose of the study was given to the families of the participants before the beginning of the study. An agreement was provided by tutors of all the participants. Each participant was filmed all along with the venipuncture procedure.

Research data is collected using a pain assessment grid of the behavioral reactions of children with ASD. (developed from the Pain – Intellectual Disability Assessment Grid (GED-DI) GED-DI and the CFCS (Child Facial coding system) This grid is subdivided into three areas (20 items): facial reactions (item 1 to 6), vocal/verbal (item 7 to 12) and motor (item 13 to 20). All behaviors are rated in presence-absence. The absence of reactive behavior is noted 0, the presence of one or more behaviors is noted 1.(Figure 1)

Figure N° 1: Pain assessment grid for children with autism spectrum disorders

Pain assessment grid for children with autism spectrum disorder

Name:

Date :

cotation instructions

0 = non observed
1 = observed

	ITEMS	Cotation
1	Deepened naso-labial furrows	
2	Eye squeeze	
3	A furrowed brow	
4	Turning down of mouth	
5	Clenching or grinding teeth, chewing or thrusting tongue out	
6	A change in eyes, including:squinching of eyes, eyes opened wide, eyes frowning	
7	Crying	
8	shouting	
9	Moaning, whining, whimpering	
10	A specific sound or word for pain (e.g., a word, cry or type of laugh	
11	Screaming/yelling	
12	visible tears	
13	Agitated	
14	Sttif, Rigid	
15	Tighten up, Tense	
16	Hetero-aggressiveness	
17	Flinching or moving the body part away, being sensitive to touch	
18	Protecting, favoring or guarding part of the body that hurts	
19	Moving the body in a specific way to show pain	
20	Try to grab the syringe and the nurse's hand	

5.-Discussion and analysis of results

Table 1,- Study population

	Age in month	Gender	Intensity of trouble	Developpemental level
1	63	Male	30	151
2	92	Female	26	274
3	100	Male	28	201
4	89	Male	30	244
5	89	Male	25	132
6	119	Male	30	158
7	83	Male	26	229
8	115	Female	36	97
9	98	Male	28	158
10	69	Male	28	186
11	63	Male	33	122
12	64	Male	28	172

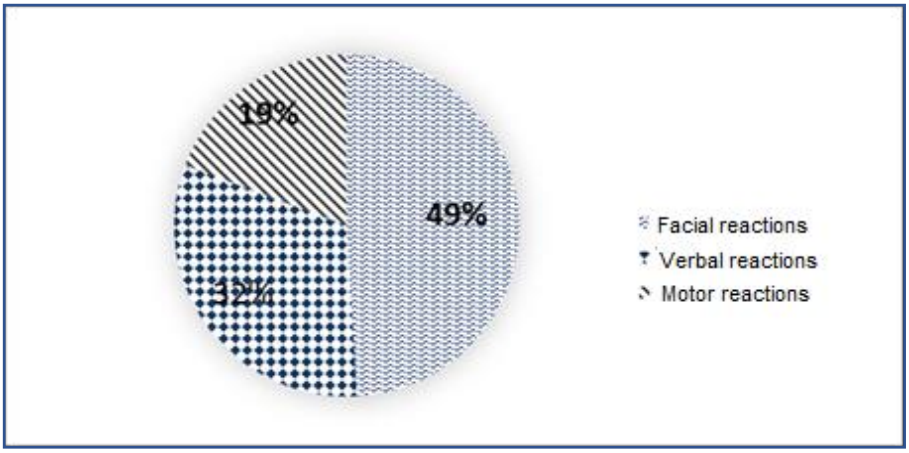
The average age of the children was 7 years and 3 months. It is observed that 83.33% of the children were male.

According to the CARS scale, the children had mild to moderate autism and severe autism, and according to Vineland, intellectual disability ranged from mild to moderate to severe (Table 1).

Table 2,-Results of the coding of the expressed reactions

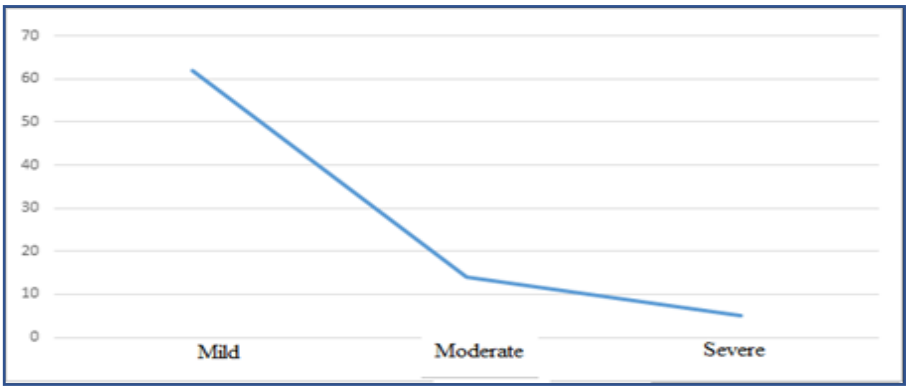
	Facial reactions	Vocal reactions	Motor réactions
Observer 1	39	27	17
Observer 2	51	33	20
Total	90	60	37

Figure N° 2: Corrected graphical representation of the observations



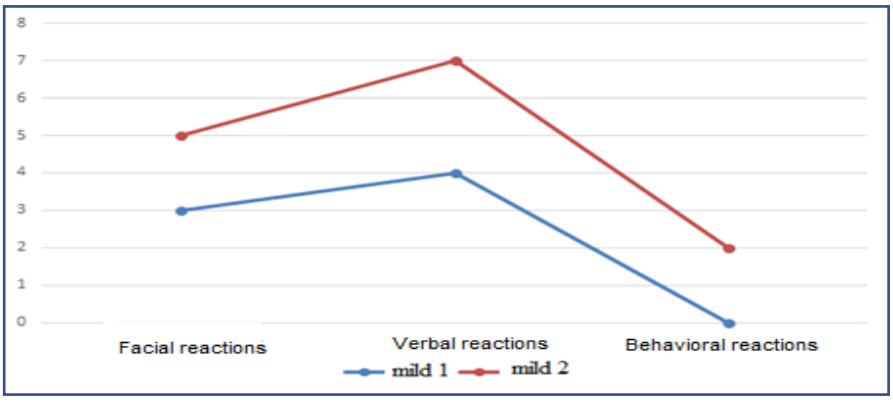
-19% of children with ASD reacted to the venipuncture with motor reactions, 32% reacted with vocal productions, half of the children (49%) presented facial expressions with a high correlational concordance index: ρ equalsat 0,98 (Figure 2, Table 2).

Figure N° 3: Expression of pain according to the intensity of the disorder



The assessment of pain in children by the grid showed that children with mild autism express their feelings of pain more during painful stimulation (Figure 3).

Figure N° 4: The inter-individual variability in children with autism with identical characteristics



There is inter-individual variability existing in autistic children with identical characteristics, as shown in the graph of two children with mild autism with the sameage (Figure 4).

This study investigates the expression and assessment of pain in venipuncture in children with ASD using a rating scale inspired by the GED-DI and CFCS.

The hypotheses of this study were: the expression of pain in children with ASD depends on the intensity of autism and intellectual disability, and we would expect that children with ASD produce more facial reactions when during painful stimulation. Concerning the pain expression, a descriptive analysis of the responses in this grid shows that children with ASD express their pain through various behaviors (vocal, facial and behavioral).

Facial reactions appear as the dominant mode of expression of pain in our sample. These descriptions are consistent with the research carried out by Lilley, Craig, and Grunau (1997), E, Pernon, C, Rattaz, 2003).

The relationship between pain expression and the intensity of autism in childrenwith ASD has been little investigated so far: Courtemanche and Black (2016) have carefully examined this link in addition to Garcia and al (2018). In their research, they suggested that the more the severity of ASD is, the responsiveness to pain decreases. This observation is consistent with the results of our research.

Other researchers, however, have not found evidence that supports the existence of a link between the intensity of ASD and pain threshold (Duerden et al. 2015; Yasuda et al 2016).

The limitations of the study**:**

The size of the sample.

Another limitation is that we were unable to directly test the role of intellectual functioning on pain response in this study.

Future studies will hopefully deepen and confirm these results.

6. Conclusion

To conclude, the present study revealed that the expression of pain in children with ASD is most of the time hyporeactive in response to the child's significant deficiency in terms of communication and interaction and that the more the severity of ASD, the more responsiveness to the pain decreases. It is important to make educators and those taking care of these children aware of the existence of this pain as they should pay attention to it.

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