



Case report

Voiceless disability: A worth case of bilateral infrainguinal testicular torsion in a patient with cerebropalsy[☆]

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ABSTRACT

Introduction: A fast surgical treatment is the gold standard when a testicular torsion is diagnosed.

However, an early diagnosis of torsion may not be feasible in case of torsion associated with undescended testis in the patients affected by cerebropalsy.

Case presentation: A Bolivian 16 year old male with acquired cerebropalsy and spastic neuromuscular disease was admitted to our Institute for a right inguinal swelling observed by the father in the morning. Indeed, the father had reported that the swelling had may be started two days before without pain or any other symptoms apparently. Two episodes of vomiting were only reported. At the general examination the patient, apparently, seemed to laugh repeatedly and a spastic movements increase were observed. The child had an infrainguinal bilateral cryptorchidism. An urgent left infrainguinal orchiectomy had been performed in the past and contralateral cryptorchidism was not corrected. At the right inguinal exploration, a complete twist of the spermatic chord was observed and a right orchiectomy was then performed.

Discussion: Testicular torsion in the inguinal canal is a rare reported condition that usually can involve patients with spastic neuromuscular disease. Processing, communication and verbalization of a chronic or acute pain seems to be different in a child with or without intellectual disability. It could be a lot more difficult to correct pain interpretation, with an important repercussion on pain accurate assessment and management.

Conclusion: In the patients with intellectual disability, a control of the testicles, it should always be done, mostly in case of atypical behaviour.

1. Introduction

The work has been reported in line with the SCARE criteria [1].

Written informed consent was obtained from the parents of the patient. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

The parents of the patient have given consent for possible publication of this case report.

Testicular torsion is a potential reversible acute vascular event involving the testis vascular cord that becomes twisted on its axis resulting in an avascular testis [2].

A fast surgical treatment is the gold standard when a testicular torsion is diagnosed.

However, an early diagnosis of torsion may not be feasible in case of

torsion associated with undescended testis in the patients affected by cerebropalsy with spastic neuromuscular disease that usually can be predisposing event [3,4].

So, we propose a case of delayed diagnosis of an infrainguinal testicular torsion in a patient with cerebropalsy in order to discuss the problems highlighted from this history.

2. Case report

A Bolivian 16 year old male with acquired cerebropalsy was admitted to our Institute for a right inguinal swelling observed by the father in the morning.

Indeed, the father had reported that the swelling had may be started two days before without pain or any other symptoms apparently and,

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on seeing worsening, had brought the boy to the emergency room in the evening at the end of the work shift.

Two episodes of vomiting were only reported.

The story of this boy started in Bolivia sixteen years earlier, when the mother died in childbirth and the boy developed an ischemic encephalopathy resulting in a spastic neuromuscular acquired disease.

The child had an infrainguinal bilateral cryptorchidism.

Because of work and economical problems, the father had entrusted the son to the maternal grandmother for many years and the boy had been mainly fed through a nasogastric tube.

At the age of thirteen, an urgent left infrainguinal orchyectomy was performed for a late diagnosed testicular torsion.

Controlateral cryptorchidism was not corrected.

At the general examination the patient, apparently, seemed to laugh repeatedly and the spastic movements increase compared to normal were observed.

On physical examination, a significant right inguinal swelling was observed.

The overlying skin was hyperaemic.

Furthermore, his hands, arms and chin appeared to be characterized by some scratches.

At the palpation of the right inguinal side, only a slight groan was heard.

Ultrasonography (US) showed an oval formation with thin-walled, anechoic, markedly uneven, non-vascularized to color Doppler, size of $5 \times 4 \times 2$ cm, possible expression in the first case of testicular twisted and necrotic. Marked edema of the surrounding soft tissues were observed (Fig. 1).

At the right inguinal exploration, a suprafascial necrotic mass was observed with a complete twist of the spermatic chord, as a consequence of a previous torsion of an ectopic undescended testicle (Figs. 2 and 3).

A right orchyectomy was then performed.

No post operative complications were observed and the child was discharged at day 2.

The boy was referred to an endocrinological consult in order to set up an hormonal therapy.

3. Discussion

This case has led us to reflect on various points.

Testicular torsion in the inguinal canal is a rare reported condition that can usually involve patients with cerebropalsy and spastic neuromuscular disease [3,5].

In fact, it seems that the torsion could be associated to the spasms of the cremasteric muscle that could be increased in spastic neuromuscular diseases. Furthermore, the same cryptorchidism could be linked to abnormal cremasteric muscle contraction that prevents the normal testicular to entry into the scrotum [6].

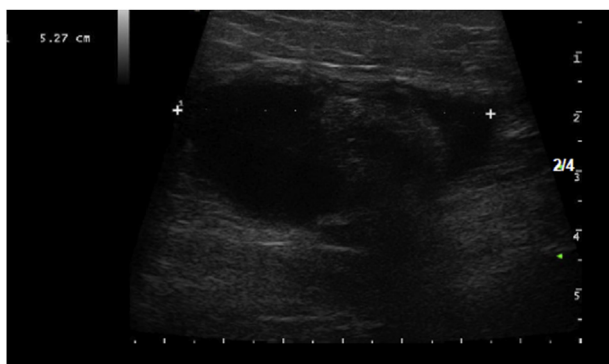


Fig. 1. Ultrasonography (US) showed an oval formation with thin-walled, anechoic, markedly uneven, non-vascularized.



Fig. 2. The necrotic testis after the inguinal canal opening.



Fig. 3. The complete twist of the spermatic chord, as a consequence of a previous torsion of ectopic undescended testicle.

Processing, communication and verbalization of a chronic or acute pain seems to be different in a child with or without intellectual disability (ID). Recently, some authors have highlighted how pain communication may depend on the child's ability to verbalize the pain. The children with ID though, at the same time able to verbalize the pain, would actually behave similarly to the children with normal intellectual development.

So, facial expression and pain verbalization may be the signs to consider in those children with capacity to verbalize the pain, and body movements and posture would not seem relevant. Furthermore, in these children, ID reduces the ability to control intentionally, to conceal or modify their behaviour. Therefore pain verbalization and facial expression may be even more evident than they are in children without ID [7].

Conversely, the children with ID and without capacity to verbalize their pain, would process and communicate the pain through documented expressions such as paradoxical laughs, increasing of the stereotyped movements or spastic movements, abnormal posture, and self-reliant behaviour [7–11].

Stereotyped body movements in these children, do not always seem to be associated with a communicative goal, perhaps having a self-centred goal rather than involving the others in their painful experience [9,12].

Consequently, in these patients, the interpretation of pain could be extremely difficult, with an important repercussion on pain accurate assessment and management.

In conclusion, in the patients with ID, a control of the testicles should always be carried out, mostly in case of atypical behaviour such as those described. It might be useful to customize pain assessment tools basing on the children's abilities, such as the new pain's scale creation, being aware of the specific differences in the expression of pain in children with ID.

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This study did not need of any registration in a publicly accessible database, not being a research study.

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Consent

Written informed consent was obtained from the parents of the

patient. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Author contribution

Edoardo Guida and Enrica Verzotti wrote the first draft of the manuscript.

Daniela Codrich and Federica Pederiva contributed to data collection.

Massimo Di Grazia contributed to the psychological part of the manuscript.

Jurgen Schleaf supervised this study.

Guarantor

Edoardo Guida, MD.

Appendix A. Supplementary data

Supplementary data related to this article can be found at <https://doi.org/10.1016/j.amsu.2018.08.011>.

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