#### **BRIEF REPORT**

## ACTA PÆDIATRICA

# Girls with cognitive impairment had a higher risk of ovariectomy for delayed recognition of adnexal torsion

Adnexal torsion is referred to when the ovaries, and sometimes the fallopian tubes, twist, and this interrupts the arterial supply and causes ischaemia. It is a relatively rare problem in paediatric emergency departments, and the annual incidence has been estimated to be 4.9 per 100,000 in the paediatric population.<sup>1</sup> There are established risk factor for adnexal torsion in adult women, such as inducing ovulation, ovarian hyperstimulation syndrome, a history of adnexal torsion, polycystic ovary syndrome, previous tubal ligation, teratomas and pregnancy.<sup>2</sup> However, little is known about the specific risk factors in the paediatric population. Moreover, the diagnosis is often difficult due to non-specific symptoms. It mainly relies on imaging, such as pelvic ultrasounds<sup>3</sup> or computed tomography or magnetic resonance imaging scans, but the role of blood tests in diagnoses is controversial.<sup>4</sup> Little has been published about the specific risk factors for delayed diagnosis and oophorectomy.

The aim of this retrospective study was to identify specific risk factors for delayed diagnosis and oophorectomy. It was conducted in two hospitals in northern Italy, the IRCCS Burlo Garofolo Pediatric Institute in Trieste and the Santa Maria Degli Angeli Hospital in Pordenone. The study was approved by the Institutional Review Board of the IRCCS Burlo Garofolo Pediatric Institute in September 2021.

We focused on patients aged between 11 and 18 years who presented to the paediatric emergency departments of the two hospitals and were diagnosed with adnexal torsion from January 2013 to December 2021.

The following data were collected: demographic characteristics, any cognitive impairment, type of symptoms, time from arrival in the paediatric emergency department to diagnosis and type of imaging and any referral to subspecialists. We also looked at any laboratory results that did not fit normal the parameters for beta-human chorionic gonadotropin, complete blood count and C-reactive protein. We compared quantitative variables with the Mann–Whitney U test or Student's *t* test on the basis of Shapiro–Wilk's test, and qualitative variables with the chi-squared test or Fisher test, when appropriate. The level of statistical significance was set at p = .05 and SPSS, version 21 (IBM Corp) was used for the analyses.

We detected 40 cases of adnexal torsion and studied the 36 girls with complete data. For 8/36 (22%) of the patients, oophorectomy was performed. The symptoms at onset included pain, nausea, vomiting and peritonism (Table 1). None of those symptoms, alone or in association with each other, were related to ovarian loss. We found that cognitive impairment was a risk factors for the oophorectomy (p-value < .001) in four patients with limited verbal expression, chromosomal abnormalities or cerebral palsy and a Gross Motor Function Scale of 4-5. As expected, the time between symptom onset and diagnosis was related to the ovarian loss risk (p = .033). The average time was 26 h in the ovarian loss group compared to 12 h in the group who were restored to their original condition. The average time that the in-hospital diagnostic work-up took did not differ substantially between the groups: 6.0 h in the loss group versus 6.5 h in the restored group (p = .76). This means that the delay was not mainly due to poor paediatric emergency department management, but to other factors, such as delayed hospital referrals. Analgesic administration did not differ in the two groups and did not mask the clinical picture. In the ovarian loss group, seven of the eight subjects who underwent blood tests had results that deviated from normal levels. Additional diagnostic and therapeutic data are reported in Table S1, but none of these showed significant differences between the two groups.

The study's main limitations were the retrospective design, small sample size and the lack of data on surgery timing and techniques. These may have been different between the two hospitals and could have influenced the outcomes.

This was the first study to demonstrate the high risk of ovarian loss due to ovarian torsion in girls with cognitive impairment. The findings can easily be explained by the inability of these patients to self-report pain, which could have led clinicians to underestimate their symptoms.<sup>5</sup> It is important that physicians are aware of the risk of ovarian torsion and are highly suspicious of this possibility when peri-pubertal girls present with the possible key symptoms of pain, vomiting and peritonism. Close collaboration with the girls' caregivers is important, and they provide the best assessment of pain. Dedicated pain scales also play an essential role in facilitating a timely diagnosis.

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	Outcome			
Variables	Total <i>n</i> = 36	Ovarian preservation n = 28	Ovarian loss n = 8	p Value
Age (years), mean (SD)	13.9 (2.3)	13.8 (2.4)	14.4 (2.4)	.502
Cognitive impairment, n (%)				
None	32 (88.9)	28 (100)	4 (50.0)	<.001*
Yes	4 (11.1)	0	4 (50.0)	
Symptoms, n (%)				
Only pain	24 (66.7)	19 (67.9)	5 (62.5)	.216
Nausea/vomiting	7 (19.4)	6 (21.4)	1 (12.5)	
Peritonism	2 (5.5)	2 (7.1)	0	
Pain and other symptoms	2 (5.6)	1 (3.6)	1 (12.5)	
Other	1 (2.8)	0	1 (12.5)	
Time from onset of symptoms to diagnosis of ovarian torsion (hours), average (IQR)	16.5 (7-38)	12 (5.5–37)	26 (21-47)	.033*
Mean number of clinical assessments (IQR)	2 (1-4)	1.5 (1-3.5)	4 (3-4)	.051
Pain reduced n (%)				
None	13 (36.1)	10 (37.5)	3 (37.5)	.618
Yes	23 (63.9)	18 (64.3)	5 (62.5)	
Diagnostic work-up length (hours), average (IQR)	6 (4-12)	6 (4-12)	6.5 (3.5–15)	.760

Abbreviations: IQR, interquartile range; SD, standard deviation.

 $p^*$  < .05 = significant higher risk of having ovarian loss instead of ovarian preservation.

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#### CONFLICT OF INTEREST

The authors have no conflicts of interest to declare.

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**TABLE 1** Demographic and paediatric emergency department variables

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#### SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.