CASE REPORT

Rare urological manifestation of Henoch-Schönlein purpura: testicular torsion

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SUMMARY

The Henoch-Schönlein purpura (HSP) is a systemic vasculitis that mostly occurs in children. Besides the most common clinical manifestations such as palpable purpura, arthralgia, abdominal pain and renal disease, it can have urological manifestations. We report a rare case of a 2-year-old boy with the HSP who developed a testicular torsion under corticosteroid treatment.

BACKGROUND

Henoch-Schönlein purpura (HSP), also known as IgA vasculitis, is a systemic disease that occurs mostly in infants. HSP may develop from the age of 6 months to adulthood. Nevertheless, about half of all cases occur in children under 5 years of age, and 75% of patients are under 10 years of age. The median age at onset is 4 years.¹ It is of unknown aetiology but probably related to an autoimmune process. HSP is characterised by non-thrombocytopenic palpable purpura, mostly accompanied by abdominal pain, arthritis and renal involvement. Urogenital involvement has been described in rare cases. This is the report of a 2-year-old boy that had been diagnosed with HSP who developed 10 days after admission to the hospital a testicular torsion under corticosteroid treatment.

CASE PRESENTATION

A 2-year-old boy presented to the emergency room with fever and a swollen left knee. The boy had fever up to 40°C since 10 days, accompanied by a sore throat and coughing. A few days earlier, his general practitioner initiated antibiotic treatment because of a clinical otitis media. While he was in the emergency room, he started developing purpura on his extremities and face. The swelling of his knee had started a few hours earlier without a prior trauma. Ultrasonography of his left knee showed a limited amount of effusion in the bursa suprapatellaris. Blood tests showed a slightly elevated ESR (36 mm/hour) and CRP (9 mg/L). The patient was admitted to the hospital for observation with the working and tentative diagnosis of HSP. During his stay, the patient developed symptoms such as oedematous joints, abdominal pain, bloody stool, hypertension, proteinuria and purpura, all described to HSP. Corticosteroid treatment was started because of renal involvement and hypertension. Skin biopsy showed a leukocytoclastic vasculitis and IgA depositions, suggestive for HSP.

Ten days after admission, the boy started to develop a painful and swollen right testicle. An ultrasound was performed and showed a decreased

blood flow on both sides with normal homogenous parenchyma. Physical examination as well as radiological examination could not rule out testicular torsion. A paediatric urologist in a nearby academic hospital was consulted before surgical exploration was performed. After a few hours, the swelling increased and extended to both sides, and consequently surgical exploration was performed with testicular torsion being our working diagnosis.

INVESTIGATIONS

Blood tests showed besides a slightly elevated ESR (36 mm/hour) and CRP (9 mg/L) no abnormalities. Extended blood tests showed an elevation of AST (1610 IE/mL) and anti-DNAse B (301 IE/mL).

Ultrasonography of his knee showed a limited amount of effusion in the bursa suprapatellaris.

Abdominal ultrasonography was performed to rule out an invagination as the cause of bloody stools, and no abnormalities were found. Since the boy was developing a swelling on his head, a CT cerebrum was performed which showed a subperiosteal fluid collection left occipitotemporal and a subcutaneous fluid collection right parietal. Scrotal ultrasonography showed a reduced blood flow on both sides with fluid around testes and epididymis; a testicular torsion could not be ruled out.

DIFFERENTIAL DIAGNOSIS

Owing to ongoing HSP, scrotal involvement of HSP was in our differential diagnosis. Since scrotal involvement had been developed under corticosteroid treatment,² the differential diagnosis also included orchitis, epididymitis and testicular torsion. Owing to the later developed oedemas, we also considered acute haemorrhagic oedema of infancy (AHEI).

AHEI is a variant of leukocytoclastic vasculitis and is characterised clinically by purpura, fever and oedema of the skin in very young children (<2 years). Involvement of internal organs is rare. Preferred locations of the skin abnormalities are ankles, wrists, scrotum and face (cheeks, eyelids and eyes). AHEI is very similar to HSP. The differences between them are preferred age (<2 years in AHEI and 3–7 years in HS), place of the skin abnormalities, histopathology and immunofluorescence.³

TREATMENT

Surgical exploration was performed since testicular torsion could not be ruled, and swelling and pain increased. The right testicle was oedematous and purple coloured. After distorsion, the testicle

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regained its normal colour, and both testicles were fixated using Winkelman's method.

OUTCOME AND FOLLOW-UP

The diagnosis testicular torsion was confirmed during the operation. Postoperatively, the patient had scrotal pain, which reduced in a few days. Still he developed a stridor for which he was treated with epinephrine atomisation and dexamethasone. As mentioned earlier, the HSP showed an atypical course, and the patient developed more swellings in his face, feet and arms. These swellings continued for more than 1 month after he was diagnosed with HSP.

DISCUSSION

Scrotal involvement of HSP is a well described but rare phenomenon that has been reported to occur in 2–38% of boys with HSP.⁴ In most cases, it is a self-limiting disease that responds well to antibiotic and/or corticosteroid treatment.⁵ According to the literature, HSP-related acute scrotum, sometimes mimicking testicular torsion,⁶ can also be caused by orchitis, epididymitis and spermatic cord complications.⁷ This a young case of testicular torsion as it mostly occurs, apart from newborn infants, in boys during puberty.⁸ As a scrotal manifestation of HSP testicular torsion is highly unlikely, it, however, is a serious diagnosis that should definitely be ruled out⁴ since there is only 6–12 hours period to prevent testicular necrosis.

Our patient did not receive antibiotics and was already undergoing corticosteroid treatment when he developed scrotal pain, and under this therapy, there was progression of disease. Surgical exploration was performed because ultrasonography could not rule out testicular torsion, and the acute scrotum did have the appearance of being neither self-limiting nor responsive to corticosteroid treatment.

To the best of our knowledge, testicular torsion has only been reported once in 1974,⁹ and we would like to use this case to bring testicular torsion back under attention as a serious urological manifestation of HSP.

Learning points

- In boys with Henoch-Schönlein purpura (HSP) always perform a full physical examination, including the urogenital area.
- HSP can cause serious and sometimes irreversible damage to the testicles.
- Testicular torsion can be an acute manifestation of HSP that requires adequate work-up and treatment.

Contributors PO has written the introduction and background. MD has written the case presentation. LF has read and edited the case report.

Competing interests None declared.

Patient consent Obtained.

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