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A Rare Variant of H-Type Recto-Vestiubular Fistula: A Case Report

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Abstract | **Case Report**

H-type recto-vestibular fistula was described in 1960 for the first time. This condition is characterized by a congenital connection between the rectum and urogenital tract, along with an external anal opening that may be normal or ectopic. We report a very rare case of a variant of this congenital malformation in a 3 years old female patient.

Keywords: H-fistula; H-type canal; Ano-rectal malformation; Children.

INTRODUCTION

The H-type anomaly in anorectal malformation has garnered interest due to its uncommon occurrence [1, 2]. It has rightly been classified as a rare and regional variant in the Krickenbeck classification [3]. Diagnosis is typically simpler in females than males [4].

Several surgical techniques have been documented in medical literature to address this issue [5].

We report the case of a 3-year-old patient presenting with a rare variant of the H-type fistula.

CASE REPORT

This concerns B. M., a 3-year-old child admitted to the Pediatric Surgery Department A of the Children's Hospital in Rabat, Morocco, for the management of a suspected anorectal malformation, indicated by discharge of feces and serous fluid through the vulva.

The patient was in good general condition, with perineal examination revealing the presence of an orifice at the vulvar level discharging whitish mucus, with well-placed anus and external genitalia typical of female anatomy.

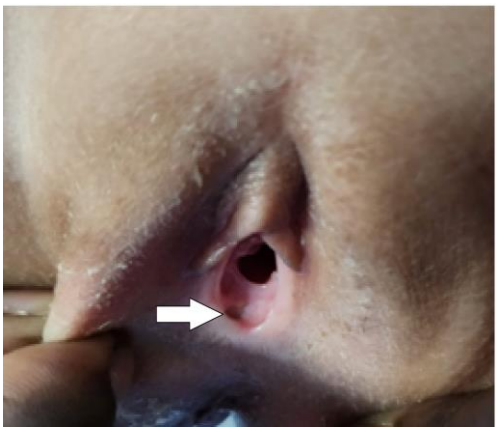

Figure 1: Image showing our patient vulvar fistula

The patient underwent examination under general anesthesia with a probe, allowing for the diagnosis of a recto-vestibular H-type fistula, for which surgery was planned.

Surgical Procedure: After placing the patient in the gynecological position with a urinary catheter in place (Fig 2). The fistula was catheterizedand we proceeded to a transverse incision halfway between the vulvar cleft and the anus.

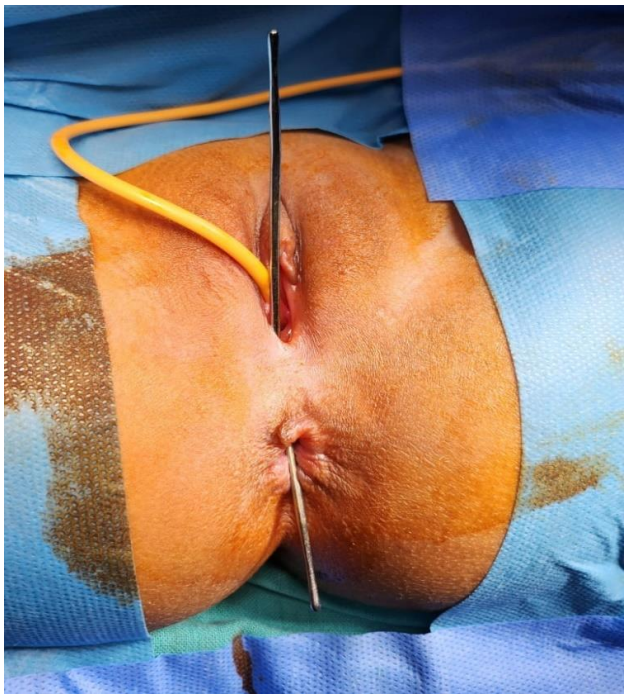

Figure 2: Image showing the catheterized H-type fistula

This was followed by dilation of the fibers of the anterior perineal central core, revealing a structure with a diameter of 1.6mm and a thick wall suggestive of an H-type canal fistula (Fig 3).

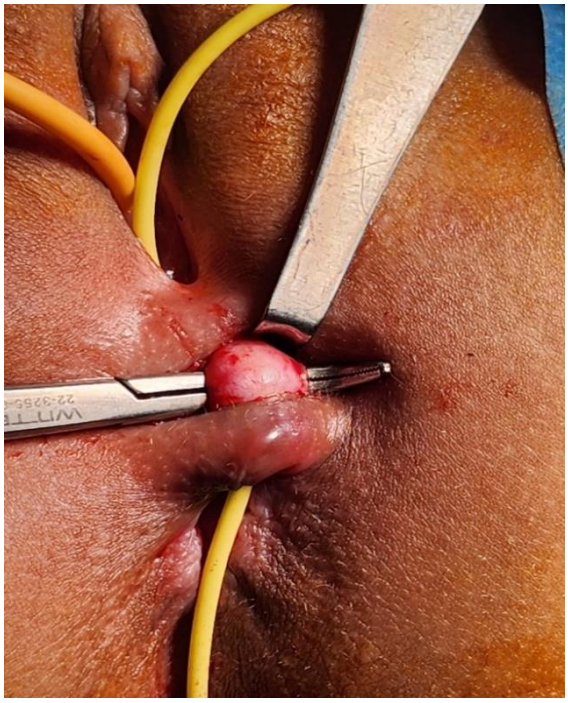

Figure 3: Image showing the H-canal type fistula

The canal was dissected, ligated and sectioned proximal to the vagina, pulled-through inside-out, ligated at the end of anal side and resected (Fig 4). The muscular fibers were approximated, and the skin was closed.

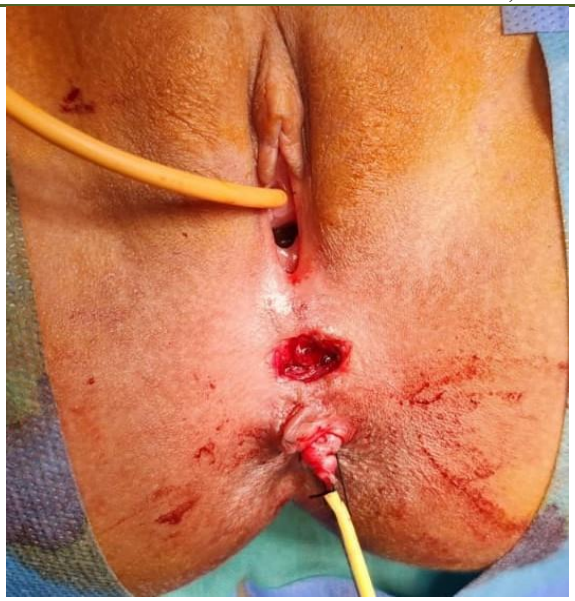

Figure 4: Image showing the pulled-through inside-out fistula at the anal side

The child stopped eating for 3 days with antibiotic coverage for 48 hours by amoxiclline/clavulanic acide.

The histopathological examination showed an anal-type wall. The patient's recovery was uneventful with good wound healing.

DISCUSSION

Bryndorf [6] initially reported this condition in 1960. H-type anorectal malformation is characterized by the presence of a congenital rectourogenital connection and an external anal opening, which may be in a normal or ectopic position. This anomaly has also been referred to as N-type anorectal malformations and, later, as a double termination of the digestif tract [4]. Historically, the term "congenital H-type fistula" primarily applied to females, as the anomaly is more prevalent in females than males. In females, the connection is typically with the genital tract, whereas in males, it is with the urinary tract.

In females, the anomaly can manifest in three types: rectovaginal, rectovestibular, and anovestibular, with either an ectopic or normal anus. In females, the anus is commonly found in its normal location and of normal size. Consequently, this condition has also been referred to as anovestibular or rectovestibular fistula with a normal anus [4]. When ectopic, the anus may be stenosed or of normal size. The term "perineal canal" has been used to describe the low H-type anomaly, which is similar to anovestibular fistula [4].

In literature, it was observed that several lesions closely resembling the typical H-type anomaly were previously categorized as H-type anorectal malformations. These variations could be considered as variants of the H-type, given their differing treatment approaches. To clarify these distinctions, terms such as "sinus" and "canal" have been introduced. Therefore, the following variants may be described [4].

1. H-type fistula with perineal fistula (more common inmales).
2. Perineal Groove.
3. H-type sinus, a blind-ending fistulous tract.
4. H-type canal. A large fistula more than or equal to1.5 cm that requires a formal closure should beconsidered as a variant.
5. Acquired H-type fistula following infection (morecommon in females).
6. Two perineal/penoscrotal/scrotal fistulae.
7. Three openings, pinhole anus with rectoscrotal andrectopenile urethral fistula.
8. H-type fistula with an imperforate anus.

In our patient it was an H-type canal variant.

The thickened appearance of the muscular layer, and the significant diameter in the case of a "canal-type fistula," could suggest the differential diagnosis of anal duplicity, as presented in our case, however, the term anal canal duplication is restricted to duplications along the posterior side of the anal canal, with a perineal orifice situated just behind the anus, at 6 oclock, with aphysical aspect of duplicate anus [14], this allows us to rule out the differential diagnosis of anal duplicity.

Since 1960, various surgical techniques have been attempted to reduce recurrence and complications. However, there is no consensus on the most effective approach. The early reports suggest that performing a protective colostomy can decrease the risk of wound dehiscence [7, 8]. However, more recent research suggests that repairing the fistula without colostomy does not increase the rates of wound dehiscence or infection. This may be due to improvements in preoperative and postoperative management, as well as

modifications to surgical techniques [9-12]. In our case, we did not perform colostomy, and there were no complications such as infection or wound dehiscence.

In 2013, Park described three cases treated using the endorectal mucosal advancement flap technique [13]. Only one case experienced constipation and anal fissures, which were successfully treated with laxatives and sitz baths.

Compared to the anterior rectal wall pull-through method, endorectal mucosal advancement flap technique causes less damage and tension, even when dealing with higher fistula positions. However, the number of reported cases utilizing this technique is limited [5].

CONCLUSION

There are numerous clinical forms of anorectal malformations and the H- canal type recto-vestibular fistula is a very rare variant of those malformations. Therapeutic options vary according to surgeons' experience.

Conflict of Interests: The authors have no conflict of interests to declare.

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