RECTOVESTIBULAR FISTULA WITH NORMAL ANUS: A treatment alternative

Anwar Abdul-Hadi MD, Humberto Lugo-Vicente MD

ABSTRACT

Congenital rectovestibular fistulas with normal anus are a rare form of anorectal malformations, especially in the Western hemisphere. Due to its rarity, consensus on preoperative management, surgical technique and postoperative care is still in debate. We describe a specific case with its management plan and outcomes while providing an up to date literature review on current management trends.

Index words: rectovestibular, fistula, normal, anus, treatment, alternative

INTRODUCTION

Anorectal malformations are well known congenital entities that comprise a spectrum of anomalies. Rectovestibular fistulas with a normal anus, also known as H-type fistulas or double termination of the alimentary tract, are an uncommon subtype comprising about 2.4% to 3.2% of all anorectal malformations in the Western Countries. Due to its rarity, consensus about preoperative management, surgical options and postoperative care have not been established. Several case series have reported different approaches with variable results. We hereby present a case to evaluate a specific management option and outcome.

Case History

A one-month-old-girl born at term was brought to the Emergency Room by her mother who noticed stool output from the vaginal vestibule since two weeks. The mother denied fever, changes in behavior, constipation, vaginal swelling, redness, tenderness, suppuration or any other symptoms. On physical exam the patient had a normally positioned anus with adequate sphincter tone. Stool output was evident through the vaginal orifice. Pelvic sonogram was performed which showed a normal pelvic anatomy and skeletal survey films had no evidence of anomalies. A Barium enema failed to show a rectovestibular fistula. The patient was taken to the operating room for a recto-genital exam under anesthesia were a rectovestibular fistulous tract one cm proximal to the dentate line was easily canalized using a 24 GA angio catheter (see Figure 1). The patient had a normal non-stenotic anus, without any associated fissures, erythema, or evidence of infection or trauma. The diagnosis was consistent with a Congenital H-Type rectovestibular Fistula. The child was discharged home.

Follow up in the Pediatric Surgery outpatient clinics showed no clinical deterioration without evidence of infection. Scheduling for surgery at three months of age was done with a day before surgery admission for mechanical bowel preparation with polyethylene glycol until clear stool output, clear liquids diet and oral metronidazole preparation. At the operating room under general anesthesia the child was placed in a prone jackknife position. The perianal and vaginal area were prepared and draped in sterile fashion. Silk 5-0 perianal sutures were used for anal exposure and a small guide wire was passed through the rectovestibular fistula for anatomical demarcation. Silk 5-0 perianal sutures were placed at the rectal fistula opening to serve as retraction during dissection (see Figure 2).

Needlepoint cautery was used for close circumferential dissection of the fistulous tract in an anus-to-vestibular direction, dividing the anterior external sphincter muscle in the midline. Dissection of the fistula continued up to the vestibule (see Figure 3).

After resection of the fistula the sphincteric muscles were approximated with interrupted vicryl 6-0. A U-shaped anterior rectal flap was created cephalad to the defect with a one cm
reported. Three weeks after dis-charge follow-up a small muco-
sal tag was observed along su-
ture line. Patient was scheduled
for an exam under anesthesia a
week later, were the mucosal
tag was excised and approxi-
mated primarily with vycryl 6-0.
No evidence of re-fistulization
or anal stenosis was appreci-
ated (see Figure 5). Pathol-
ogy was pertinent for an acro-
chordon. During the next none
months the child has developed
properly, gaining weight with no
evidence of fistula recurrence.

DISCUSSION

Due to its low incidence (0.7% - 3.2%) in
Western Countries among all the ano-
rectal malformations, H-type rectovestibular fistula
treatment lacks consensus. Our case
debuted with passage of stool per vagina, the
most common presenting sign, without evi-
dence of infection or abscess formation by his-
tory and physical examination. Diagnosis was
difficult to achieved using conventional radiol-
ogy (barium enema and pelvic sonogram) and
it necessitated an exam under anesthesia for
thorough examination and clear demarcation
of the fistulous tract. Multiple diagnostic work-
ups have been suggested, including endosco-
pies, contrast enemas and direct inspection
under anesthesia. We agree on exam under
anesthesia being the best method for diagno-
sis with the added benefit of anatomical de-
marcation with a probe, useful for operative
planning. Additionally, patients should be eval-
uated for any associated anomalies, especially
cardiac and pelvic (including presacral masses
and anal stenosis), as they may be pres-
ent in up to 60% of patients.

A procedure at a later age was planned as mul-
tiple reports suggest that it is not necessary to
manage this pathology emergently as long as
close follow up and adequate attention is pro-
vided by caretakers. Multiple preoperative
regimens have been suggested for this type
of defect. We agree on bowel preparation as
it should theoretically reduce the risk of post-
operative infection and wound dehiscence. We
opted for polyethylene glycol solution until evi-
dence of clear stools, 24 hours of oral metroni-
dazole and clear liquids diet. Other suggested
bowel preparations including three days con-

wide apex and 3 cm proximal length bilaterally.
The flap was advanced over the previous fis-
tula site and secured with interrupted vicryl 6-0
sutures to the anterior anal canal (see Figure
4). Pathology evaluation agreed with fistula histology, no associated inflammatory changes
were reported.

The patient was transferred to ward with Acet-
aminophen for pain control, intravenous Piper-
acillin/Tazobactam, nil per os status and TPN
to maintain hydration and adequate nutrition.
The first bowel movement was achieved on
postoperative day #1. On day #3, with an in-
tact suture line and no evidence of infection,
a clear electrolyte diet (PedialyteTM, Abbott
Laboratories) started. On day #4 the diet was
progressed to a half-strength milk solution. The
patient continued to stool with each feeding.
On postoperative day #5 diet was progressed
to regular formula, which was well tolerated.
The patient was discharged home on day #6
without complications and acetaminophen
elixir for pain control.

Follow up was done one week after discharge.
An intact suture line, no anal stenosis and no
evidence of infection were noted. No other
events of stool output through vestibule were

Figure 1. Rectal exam under anesthesia with a plastic probe passed along the recto-vestibular fistula. Denotate line is appreciated distal to the fistula opening.

Figure 2. Guide wire passed along the fistulous tract and silk 5-0 sutures were used to retract fistula.
Figure 3. Fistula was completely resected. There is a communication between the rectum and vestibule.

Figure 4. End result after the anterior rectal flap was advanced over the previous fistula site and secured with interrupted vicryl 6-0 sutures to the anterior anal canal.

Figure 5. Complete resection of mucosal tag prior to primary closure.
sisting of simple enemas twice daily, liquid diet, and oral metronidazole; saline enemas and clear liquids diet for 24 hours; saline enemas, liquid diet and oral metronidazole for 24 hours; polyethylene glycol and cleansing enemas for 24 hours; and liquid diet for one day with saline enemas the night before the operation. There is no evidence that suggests any regime to be superior.

A transanal approach was done for resection of fistula with an anterior endorectal mucosal flap to cover the previous fistula site, similarly to the approach described by Park with an added complete resection of the fistula instead of curettage. Complete resection of fistula with an anterior endorectal flap mobilization, as opposed to fistula curettage to cover previous fistula site has been suggested as an important measure to reduce the risk of recurrence, and maintaining the posterior and lateral aspects of the sphincters and perineal body intact, thus preserving continence. We believe that the addition of fistula resection does not imply a more technically challenging surgery, while providing similar results with preservation of the internal anal sphincters and the added benefit of complete fistula removal; which theoretically should reduce the recurrence rate. Other suggested surgical approaches include anterior sagittal anorectoplasty, posterior sagittal anorectoplasty, fistulectomy, vestibuloanal pull-through, perineal repair, fistulotomy and curettage with or without a diverting colostomy, primary closure of the fistula with diverting colostomy and endorectal advancement flap. We also agree that primary repair does not necessarily need a protective colostomy, even in a patient with a history of vulvar abscess or infection as long as the infection is well managed and cleared, and good bowel preparation has been achieved.

The procedure was well tolerated and no evidence of recurrence was seen during hospitalization and follow up at two and 5 weeks post operatively. At 5 weeks from the procedure a minor complication of a mucosal tag, confirmed by pathology, along the right lateral aspect of the suture line was noted which was easily excised and repaired primarily in the operating room. Continued follow up at none months showed no signs of recurrence or infection, adequately healed wounds and good sphincter tone. Postoperative intravenous antibiotics have been previously suggested and we opted for postoperative broad spectrum antibiotics until discharge.

Histological evaluation does suggest a congenital etiology when an epithelialized tract is evidenced without associated inflammatory changes, as in our case. The anatomical origin of the fistula one cm above the dentate line coupled with no history or evidence of vestibular abscess, as seen in our patient, also supports a congenital etiology, especially when not related to any inflammatory or infectious presentation. In patients with associated abscesses or cellulitis, differing surgical treatment to clear the infection may reduce the chances of recurrence or complications. Fistula recurrence and wound disruption are common, recurrence is reported between 5% to 30%, and may be associated with preoperative infections. Complications can be varied; perineal body dehiscence, recurrence, anal stenosis, bowel incontinence. In our case a small mucosal tag along the lateral aspect of the rectal flap developed one month from operation, which was excised and closed primarily without recurrence.

Close postoperative follow up should be performed to evaluate for recurrence and the possibility of anal stenosis, not developed in our patient. Yazici et al. suggest the inclusion of rectal dilation program on follow up. Follow up is also necessary to assess for fecal incontinence at a later age, although due to the preservation of the anal sphincters and preservation of a near normal anatomy outcomes are expected to be good.

It is necessary to reach consensus for the treatment of this specific type of anomalies to reduce comorbidities, especially associated with fistula recurrence and reoperations.

**CONCLUSIONS**

**H-type recto-vestibular fistulas are rare malformations among the ano rectal anomalies spectrum. Multiple approaches have been suggested with different recurrence rates and associated comorbidities. We favor direct inspection under anesthesia for diagnosis and a transanal approach with resection of fistula and anterior rectal mobilization over previous fistula site. We believe it reduces recurrence rates and has minimal comorbidities.**
REFERENCES


RESUMEN

Las fistulas congénitas rectovestibulares que tienen ano en posición normal son un tipo de malformación anorectal extremadamente rara, especialmente en Occidente. Dado su rareza, existe debate en el consenso de su manejo preoperatorio, técnica quirúrgica y cuidado postoperatorio. Describimos un caso específico de fistula congénita rectovestibular con ano normal con un plan de manejo y resultados excelentes a luz de la literatura actual de esta condición.