JCEM Case Reports, 2025, **3**, luae239
https://doi.org/10.1210/jcemcr/luae239
Advance access publication 26 December 2024
Case Report

An Infant With *DHX37* Variant: A Novel Etiology of 46,XY DSD and Literature Review

R. Sena Turk Yilmaz,[1] Adam B. Hittelman,[2] Alla Vash-Margita,[3] Catherine Dinauer,[1] Stuart A. Weinzimer,[1] and Jasmine Gujral[1]

[1]Department of Pediatrics, Division of Pediatric Endocrinology, Yale School of Medicine, New Haven, CT 06510, USA
[2]Department of Urology, Division of Pediatric Urology, Yale School of Medicine, New Haven, CT 06510, USA
[3]Department of Obstetrics, Gynecology and Reproductive Sciences, Division of Pediatric and Adolescent Gynecology, Yale School of Medicine, New Haven, CT 06510, USA

Correspondence: R. Sena Turk Yilmaz, MD, Department of Pediatrics, Division of Endocrinology and Diabetes, Yale School of Medicine, PO Box 208064, New Haven, CT 06510, USA. Email: rukiye.turkyilmaz@yale.edu.

Abstract

46,XY sex reversal 11 (SRXY11) is a rare and recently identified form of 46,XY difference in sexual development (DSD), caused by variants in the DEAH-Box Helicase 37 gene (*DHX37*). *DHX37* is crucial for ribosome biogenesis, but its specific role in gonadal development remains unclear. The genital phenotype varies widely, ranging from typical female to typical male. We present a 46,XY infant with prenatal ultrasound findings of atypical genitalia. Amniotic fluid gene analysis revealed a known heterozygous pathogenic variant in *DHX37*, p.R308Q (c.923G>A), confirmed postnatally. The patient was born with markedly undervirilized genitalia with posteriorly fused labioscrotal folds, a single introitus, no clitoromegaly, and nonpalpable gonads. Laboratory evaluation at multiple points showed undetectable anti-Müllerian hormone (AMH) and inhibin B levels, elevated gonadotropin levels, and negligible testosterone levels. Clinical course was complicated by urine retention in the vagina and uterus and hydronephrosis requiring catheterization. Endoscopy revealed a urogenital sinus with separate urethral and vaginal openings and 2 cervices leading into 2 separate uteri suggestive of a bicornuate bicollis uterus. Laparoscopy revealed 2 intra-abdominal gonads adjacent to the fallopian tubes. Evidence for inheritance, penetrance, genotype-phenotype correlation, and risk of malignancy in SRXY11 is limited to case reports.

Key Words: *DHX37*, 46, XY gonadal dysgenesis, differences of sex development, ambiguous genitalia, testicular regression syndrome

Abbreviations: DOL, day of life; DSD, difference in sexual development; PGD, partial gonadal dysgenesis; TRS, testicular regression syndrome.

Introduction

Differences in sexual development (DSDs) refer to a collection of congenital conditions in which the development of chromosomal, gonadal, or anatomical sex deviates from the typical pattern in both 46,XX and 46,XY individuals [1]. The reported incidence of atypical genitalia in newborns is approximately 1 in 4500 births, but the exact prevalence remains uncertain [2]. DSDs arising from single gene variants typically lead to specific anatomical and functional disorders of the internal and/or external genitalia without causing other anomalies [3].

Disorders of testicular development include gonadal dysgenesis, which can be complete or partial, and the phenotype depends on the degree of testicular tissue differentiation. Complete gonadal dysgenesis (CGD) presents with typical female external genitalia, intact Müllerian structures, and bilateral streak gonads due to the absence of testicular development [3, 4]. Partial gonadal dysgenesis (PGD) results from incomplete development of the testes and is associated with a wide range of phenotypes, from mild (eg, isolated infertility) to severe (eg, undervirilization of the external genitalia and the presence of both Müllerian and Wolffian derivatives) [3, 4]. Another etiology of 46,XY DSD is testicular regression syndrome (TRS) in which the testes develop and operate normally during early embryogenesis but later regress. The phenotypic spectrum is highly varied based on the gestational week in which regression occurs [5]. The clinical features of TRS often bear similarities to those seen in PGD.

Although several genes crucial to sexual determination and differentiation have been described, a large percentage of genetic etiologies of 46,XY DSDs remain elusive. While *SRY*, *NR5A1*, and *MAP3K1* variants are the most common causes of 46,XY DSDs, their collective contribution accounts for less than 40% of all nonsyndromic forms [6, 7].

In this report, we describe a patient with 46,XY DSD, diagnosed prenatally with a pathogenic variant in DEAH-Box Helicase 37 gene (*DHX37*), a recently discovered etiology of DSDs.

Case Presentation

A 32-year-old gravid female of Brazilian descent, G4P0, with no history of consanguinity with her male partner and no significant medical history, was referred to our DSD clinic at 30 weeks of gestation due to ultrasound findings of atypical fetal genitalia. At 12 weeks of gestation, cell-free fetal DNA

Received: 27 September 2024. **Editorial Decision:** 5 December 2024. **Corrected and Typeset:** 26 December 2024

Table 1. Genes included in the extended gene analysis of the amniotic fluid

AKR1C2	*AKR1C4*	*AMH*	*ANOS1*	*AR*	*CBX2*	*CHD7*	*CYP11B1*	*CYP17A1*
CYP21A2	*DHH*	*DHX37*	*DUSP6*	*ESR1*	*FEZF1*	*FGF17*	*FGF8*	*FGF9*
FGFR1	*FLRT3*	*FSHB*	*GNRH1*	*GNRHR*	*HS6ST1*	*HSD3B2*	*IL17RD*	*KISS1*
KISS1R	*LHB*	*MAP3K1*	*NR0B1*	*NR5A1*	*NSMF*	*PROK2*	*P ROKR2*	*SEMA3A*
SEMA3E	*SOX9*	*SPRY4*	*SRXY10*	*SRY*	*STAR*	*TAC3*	*TACR3*	*WDR11*
WNT4	*WT1*	*ZFPM2*						

analysis revealed a 46,XY karyotype. Ultrasounds at 18 weeks and 23 weeks of gestation showed atypical genitalia with fused labial structures and a midline prominence. An amniocentesis with confirmatory chromosome analysis revealed the karyotype to be 46,XY with a normal microarray and the presence of *SRY*. An extended gene analysis for 46,XY DSDs (Table 1) on the amniotic fluid detected a heterozygous pathogenic missense variant, p.R308Q (c.923D>A), in *DHX37*. Parental testing revealed that this variant was maternally inherited.

The patient was born at 37 weeks via vaginal delivery with weight, length, and head circumference appropriate for gestational age. Physical examination showed prominent undervirilization of genitalia with nonpalpable gonads, hypoplastic outer and inner labioscrotal folds with posterior fusion, an edematous clitorophallic structure without clitoromegaly, and a single opening in a narrow introitus (Fig. 1) along with an orthotopic anal opening. The patient exhibited spontaneous voiding through the single introitus. The remainder of the physical examination was within normal limits.

Diagnostic Assessment

A pelvic ultrasound performed on the first day of life (DOL) revealed a fluid-filled uterus posterior to a decompressed bladder and bilateral hydroureteronephrosis without identifiable gonadal tissue. During an attempted voiding cystourethrogram on DOL 2, a catheter inserted through the introitus catheterized the vagina (Fig. 2) and drained urine. Subsequent pelvic ultrasounds revealed increased pelvicalyceal dilation. Laboratory evaluation on DOL 2 indicated undetectable levels of anti-Müllerian hormone (AMH), inhibin B, and estradiol with a negligible testosterone level (Table 2). The patient had a repeat single gene analysis of *DHX37* confirming the prenatally diagnosed variant p.R308Q. At 2 months of age, biochemical re-evaluation revealed a negligible testosterone level, undetectable AMH and inhibin B levels, and an elevated follicle-stimulating hormone level with a low but detectable luteinizing hormone (Table 2).

At 7 months of age, endoscopy revealed a urogenital sinus, 2.5-cm common channel, with distinct urethra and vagina, as well as 2 cervices. Diagnostic laparoscopy confirmed the presence of 2 separate hemi-uteri with a small indentation at the external fundus, consistent with a bicornuate bicollis uterus with 2 fallopian tubes with fimbriated ends attached to 2 gonads that resembled dysplastic testicles (Fig. 3).

Treatment

Urinary reflex into the bladder with associated hydroureteronephrosis, likely from ureteral compression, prompted the initiation of daily intermittent catheterization as well as prophylactic antibiotic therapy on DOL 2. At the 2-month follow-up visit, the patient was spontaneously voiding and receiving daily intermittent catheterization. A repeat pelvic ultrasound confirmed the resolution of hydroureteronephrosis, as well as the resolution of the vaginal distention. The patient also underwent labiaplasty to widen the introitus at 7 months of age, performed simultaneously with the vaginoscopy and diagnostic laparoscopy (Fig. 4).

Outcome and Follow-Up

After multiple counseling sessions provided by the multidisciplinary DSD team and shared decision-making, the family elected to raise the infant as a female. Prophylactic antibiotic therapy was discontinued at 9 months of age. At the current age of 13 months, the patient continues to void spontaneously without urinary tract infections and is growing and developing well, with Z scores for weight at 1.82 SD, height at 0.78 SD, and body mass index at 1.92 SD, meeting age-appropriate milestones. Repeat biochemical evaluation showed persistent elevations of gonadotropins.

Discussion

DHX37 is an autosomal gene on chromosome 12 (12q24.31) that produces an ATP-dependent RNA helicase implicated in multiple cellular processes such as translation initiation, nuclear and mitochondrial splicing, and assembly of ribosomes and spliceosomes [8, 9]. Prior to 2019, homozygous or compound heterozygous variants resulting in a loss of function of *DHX37* had been associated with neurodevelopmental disorders with brain anomalies, with or without vertebral or cardiac anomalies. Deletions or rearrangements of the 12q24 chromosomal region, which includes *DHX37*, have also been associated with atypical genital development [10]. However, it was only in 2019 that missense variants in *DHX37* were identified for the first time in individuals with nonsyndromic 46,XY DSDs presenting as PGD or TRS [9] and the disease was titled *46,XY sex reversal 11* (SRXY11) (OMIM:617362).

In the last 5 years, *DHX37* variants have been recognized as more prevalent than expected in studies utilizing whole exome analysis for 46,XY DSDs, with a proportion comparable to or higher than commonly tested genes such as *SRY and NR5A1*. Studies have reported a proportion ranging from 14% to 40% in previously undiagnosed 46,XY DSD patients [9, 11, 12]. To our knowledge, 60 patients of *DHX37* variants have been reported so far [9, 11, 13].

Although *DHX37* is a gene that is ubiquitously expressed in multiple tissues in the body, its heterozygous variants only cause DSDs and no other syndromes. It is hypothesized that

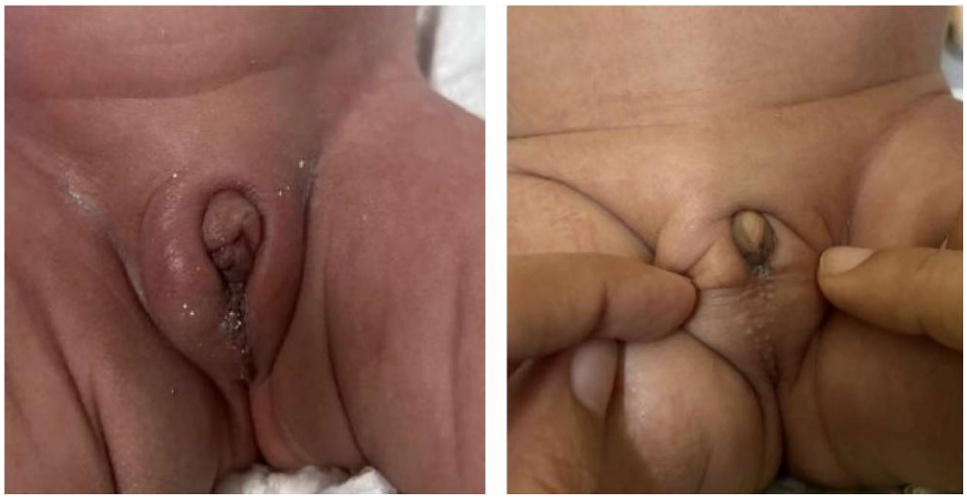

Figure 1. Genitalia exam on DOL 2 (left) and 2 months of age (right). DOL 2 shows genitalia with significant genital edema and erythema that subsequently resolved. Genitalia notable for posterior fusion, hypoplastic outer and inner labioscrotal folds, and a narrow, single introitus which is difficult to visualize.

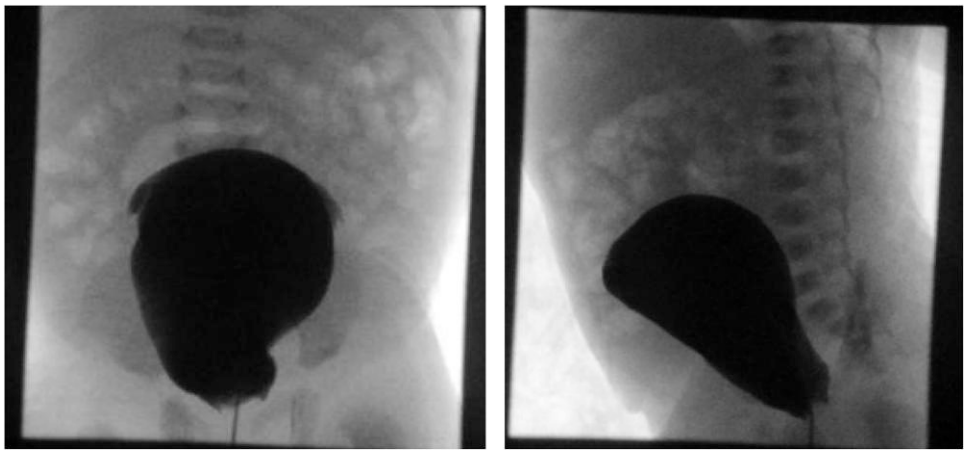

Figure 2. Voiding vaginogram on the first day of life, with preferential catheterization of vagina.

its variants lead to an abnormal increase in *WNT* signaling, leading to β-catenin protein overexpression which disrupts the normal processes of testis determination and development [10].

In SRXY11, the genital phenotype is distinguished by significant variability, encompassing a spectrum from typical female to typical male. The most observed pathogenic variant to date is p.R308Q, which was seen in our patient. This missense variant arises from a nucleotide substitution at position 923 of exon 6, where guanine is replaced by adenine, leading to an amino acid change from arginine to glutamine at position 308 (c.923G>A/p.(Arg308Gln). The phenotypes of the 23 patients carrying the p.R308Q range from gonadal dysgenesis to testicular regression, with some children being reared as females and others as males. Among the 15 patients in which transmission of the p.R308Q variant could be established, 8 were de novo, 5 were maternally inherited, and 2 were paternally inherited [9, 11-16] (Table 3).

There is one other reported patient with the same variant, who had a somewhat similar presentation with a vaginal septum and didelphys uterus [8], while our patient presented with a vaginal band and bicornuate bicollis uterus (Fig. 3). This collective data from 23 patients with the same variant demonstrating wide phenotypic variability underlines the lack of genotype-phenotype correlation in SRXY11. A distinctive feature in our patient, however, is the presence of urinary reflux causing urine retention in the vagina and hydroureteronephrosis. We believe this retrograde reflux to be from the combination of a narrow introitus along with the urogenital sinus. Compression of the distal ureters by the distended vagina likely caused the hydroureteronephrosis.

In our patient, the variant was inherited from an asymptomatic mother. While most patients in the literature with *DHX37* variants were either maternally inherited or occurred de novo, exceptions such as the p.R308Q and p.T477 M variants were passed down from a fertile father to the probands [9, 10]. This pattern aligns with a male sex-limited autosomal dominant mode of inheritance with variable penetrance. To our knowledge, there have been no reported patients of 46, XX individuals with DSDs linked to this gene. Studies conducted on goat fetuses have revealed that *DHX37* is expressed at higher levels in male gonads than in female gonads, suggesting its critical role in testicular development [17].

The gonads in our patient are located adjacent to the fimbriae of fallopian tubes, which is the typical location of ovaries; however, their gross morphology resembled testes. There

Table 2. Biochemical evaluation (SI units in parentheses)

	2 days	2 months	13 months
Luteinizing hormone (1 mIU/mL = 1 IU/L)	**<1.0 mIU/mL**	**0.17 mIU/mL**	**10.8 mIU/mL**
	Ref range 1-5 days: male: <1.35 mIU/mL female: <1.8 mIU/mL	Ref range 2-3.5 months: male: 0.62-4.08 mIU/mL female: <0.98 mIU/mL	Ref range 13 months: male: <1.3 mIU/mL female: ≤ 0.5 mIU/mL
Follicle-stimulating hormone (1 mIU/mL = IU/L)	**1.7 mIU/mL**	**16.74 mIU/mL**	**65.6 mIU/mL**
	Ref range 1-5 days: male: <2.16 mIU/mL female: <4.74 mIU/mL	Ref range 2-3.5 months: male: 0.41-3.02 mIU/mL female: 1.23-17.4 mIU/mL	Ref range 13 months: male: <1.9 mIU/mL female: 0.5-6 mIU/mL
Total testosterone	**4 ng/dL (0.13 nmol/L)**	**8.2 ng/dL (0.28 nmol/L)**	**4.8 ng/dL (0.16 nmol/L)**
	Ref range 1-10 days: male: <187 ng/dL (<6.5 nmol/L) female: 1-13 ng/dL (0.03-0.45 nmol/L)	Ref range 2-3.5 months: male: 38.9-325.9 ng/dL (1.3-11.3 nmol/L) female: <6.05 ng/dL (<0.2 nmol/L)	Ref range 13 months: <7-20 ng/dL (<0.2-0.7 nmol/L)
Anti-Müllerian hormone	**<0.08 ng/mL (<0.5 pmol/L)**	**<0.08 ng/mL (<0.5 pmol/L)**	**<0.1 ng/mL (<0.7 pmol/L)**
	Ref range 1-30 days: male: 23.8-124 ng/mL (170-885 pmol/L) female: <1.3 ng/mL (<9.2 pmol/L)	Ref range 1-4 months: male: 46.8-173 ng/mL (334-1235 pmol/L) female: <6.4 ng/mL (<45.7 pmol/L)	Ref range 13 months: male: 43.5-199.5 ng/mL (310-1425 pmol/L) female: 0.17-6.1 ng/mL (1.2-43.5 pmol/L)
Inhibin B (1pg/mL = 1 ng/L)	**<10 pg/mL**	**<10 pg/mL**	**<10 pg/mL**
	Ref range 1-2 days: male: 185-300 pg/mL female: < 68.6 pg/mL	Ref range 2-3.5 months: male: 229-631 pg/mL female: <184 pg/mL	Ref range 13 months: male: 24-300 pg/mL female: <183 pg/mL
Estradiol	**<4 pg/mL (14.6 pmol/L)**	**<2 pg/mL (<7.3 pmol/L)**	**<20 pg/mL (<73.4 pmol/L)**
	Ref range Newborn: male: < 13 pg/mL (<47.7 pmol/L) female: < 20 pg/mL (<73.4 pmol/L)	Ref range 2-3.5 months: male: <47 pg/mL (<172.5 pmol/L) female: <26.7 pg/mL (<98 pmol/L)	Ref range 13 months: male: <13 pg/mL (<47.7 pmol/L) female: <20 pg/mL (<73.4 pmol/L)

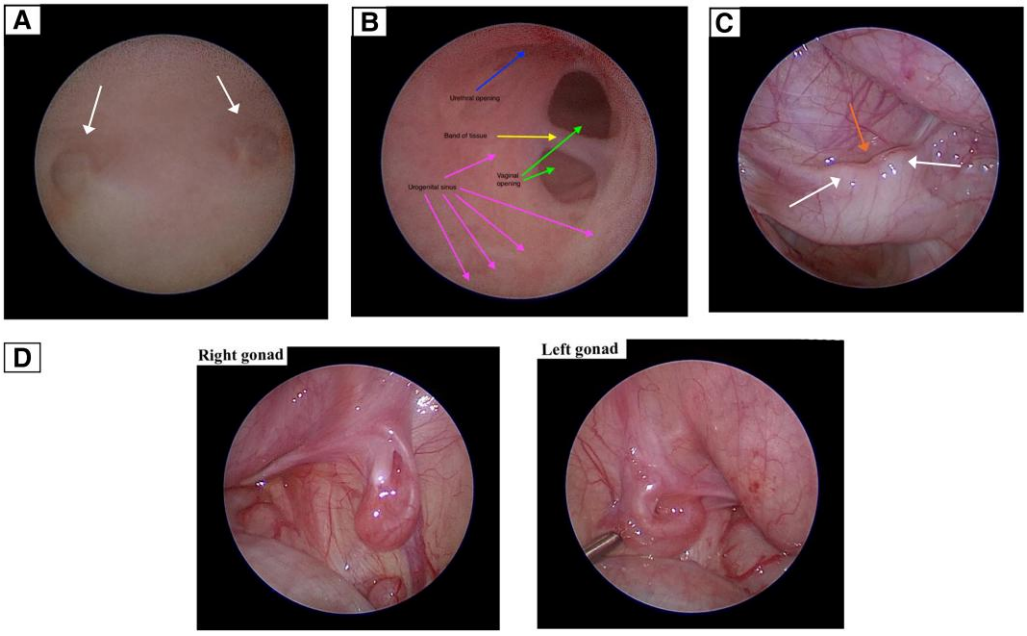

Figure 3. (A) Two cervices identified on the vaginogram (white arrows); (B) urethral opening (blue arrow), band of tissue (yellow arrow), vaginal opening (green arrows), and urogenital sinus (pink arrows) visualized through the vaginogram; (C) laparoscopic imaging of 2 separate hemi-uteri (white arrows) with a shallow fundal indentation at the fundus (orange arrow) suggestive of a bicornuate bicollis uterus; (D) laparoscopic imaging of the right and left gonads.

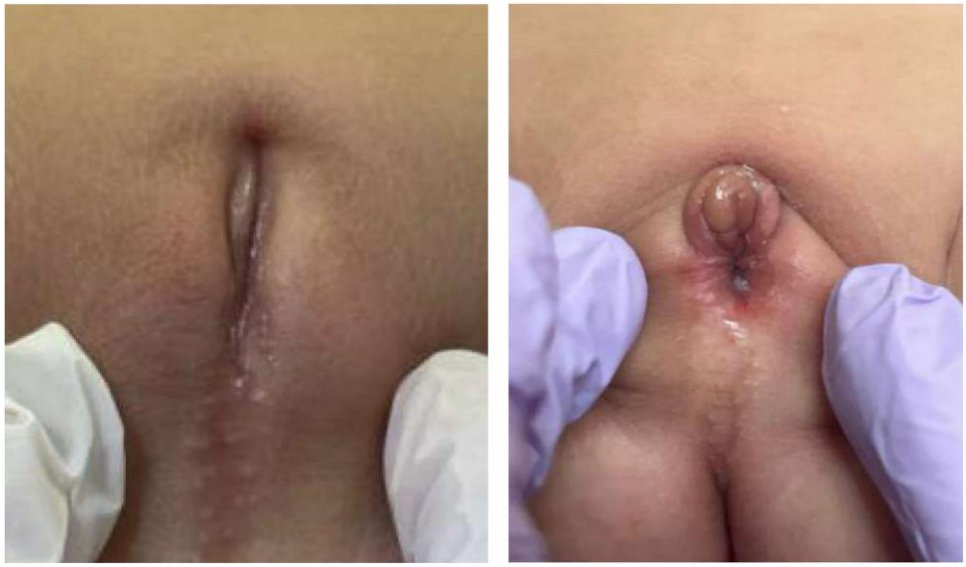

Figure 4. Hypoplastic labial folds with a narrow introitus: pre (left) and post (right) labiaplasty.

Table 3. Phenotype, sex of rearing, and inheritance patterns in studies that reported the p.R308Q variant.

No	Phenotype	Sex of rearing	Inheritance	Study
1	TRS	Male	Paternal	Da Silva et al, 2019 [9]
2	TRS	Male	Paternal	
3	TRS	Male	*De novo*	
4	TRS	Male to female	NA	
5	TRS	Male	Maternal	
6	TRS	Male	Maternal	
7	PGD	Female	*De novo*	
8	PGD	Male	NA	De Oliveira et al, 2023 [12]
9	GD	Female	*De novo*	McElreavey et al, 2020 [14]
10	GD	Female	NA	
11	GD	Female	NA	
12	GD	Female	NA	
13	TRS	Male	*De novo*	
14	PGD	NA	*De novo*	Buonocore et al, 2019 [15]
15	PGD	NA	*De novo*	
16	PGD	NA	*De novo*	
17	TRS	Male	*De novo*	Zidoune et al, 2021 [16]
18	TRS	Male	Maternal	
19	TRS	Male	NA	
20	GD/TRS	Male	Maternal	Shimura et al, 2024 [11]
21	GD/TRS	Male	Maternal	
22	GD/TRS	Male	NA	
23	GD	Female	Paternal	Margiotti et al, 2024 [13]

All patients had heterozygous variants.

Abbreviations: GD, gonadal dysgenesis; NA, not applicable; PGD, partial gonadal dysgenesis; TRS, testicular regression syndrome.

have been no reports of ovarian or ovotesticular gonadal histology in patients with *DHX37* variants. Typically, the gonads have been dysgenetic, or no gonadal tissue has been found [8].

The presence of a Y chromosome and intra-abdominal gonads increases the risk of malignancy in these individuals; however, data are lacking regarding the risk of malignancy with *DHX37* variants. To date, only one 46,XY patient has been reported to have germ cell neoplasia [8]. Overall, due to the limited number of reported patients, absence of gonadal tissue, and limited duration of follow-up, it is difficult to draw definitive conclusions regarding the incidence of malignancy. We will utilize described methods of gonadal surveillance such as imaging, gonadal biopsy, as well as potential gonadectomy in conjunction with family discussions since the gonads are not functional and likely dysgenetic.

In conclusion, we present a patient with 46,XY DSD, diagnosed prenatally from a pathogenic variant in *DHX37* causing SRXY11. Our patient presented with significant undervirilization and Müllerian structures including 2 hemiuteri and gonads adjacent to the fallopian tubes. SRXY11 is a newly described sex-limited, nonsyndromic form of 46,XY DSD that is more prevalent than previously reported. It demonstrates a wide phenotypic variability with incomplete penetrance and no genotype-phenotype correlation. Testing for this gene in 46,XY DSDs, along with the ongoing collection of clinical data, is critical to better understand this disease.

Learning Points

- *DHX37* is a newly recognized etiology of a significant percentage of 46,XY DSDs. The most common pathogenic variant so far is p.R308Q.
- The phenotype varies widely, presenting with gonadal dysgenesis or testicular regression, with no genotype-phenotype correlation.
- The most common inheritance is through an asymptomatic mother; however, paternal inheritance has been rarely observed.

Contributors

All authors contributed individually to the authorship. A.H., A.V., C.D., S.W., and J.G. were involved in the diagnosis and management of this patient. R.S.T., A.H., A.V., C.D., S.W., and J.G. participated in writing, reviewing, and editing the manuscript. R.S.T. and J.G. prepared the first draft and managed the manuscript submission. All authors reviewed and approved the final draft.

Funding

No public or commercial funding.

Disclosures

None declared.

Informed Patient Consent for Publication

Signed informed consent obtained directly from the parent's relatives or guardians.

Data Availability Statement

Original data generated and analyzed during this study are included in this published article.

References

1. Lee PA, Houk CP, Ahmed SF, Hughes IA; International Consensus Conference on Intersex organized by the Lawson Wilkins Pediatric Endocrine Society and the European Society for Paediatric Endocrinology. Consensus statement on management of intersex disorders. International consensus conference on intersex. *Pediatrics*. 2006;118(2):e488-e500.
2. Flück CE, Güran T. Ambiguous genitalia in the newborn. [Updated 2023 Nov 13]. In: Feingold KR, Anawalt B, Blackman MR, *et al.*, editors. *Endotext [Internet]*. (MA): MDText.com, Inc.; 2000-. Available from: https://www.ncbi.nlm.nih.gov/books/NBK279168/
3. Berkovitz GD, Seeherunvong T. Abnormalities of gonadal differentiation. *Baillieres Clin Endocrinol Metab*. 1998;12(1):133-142.
4. Mendonca BB, Domenice S, Arnhold IJ, Costa EM. 46,XY disorders of sex development (DSD). *Clin Endocrinol (Oxf)*. 2009;70(2):173-187.
5. Pirgon Ö, Dündar BN. Vanishing testes: a literature review. *J Clin Res Pediatr Endocrinol*. 2012;4(3):116-120.
6. Elzaiat M, McElreavey K, Bashamboo A. Genetics of 46,XY gonadal dysgenesis. *Best Pract Res Clin Endocrinol Metab*. 2022;36(1):101633.
7. Granados A, Alaniz VI, Mohnach L, *et al.* MAP3K1-related gonadal dysgenesis: six new cases and review of the literature. *Am J Med Genet C Semin Med Genet*. 2017;175(2):253-259.
8. de Oliveira FR, Guaragna MS, Maciel-Guerra AT, *et al.* DHX37 and the implications in disorders of sex development: an update review. *Horm Res Paediatr*. 2024;97(5):433-444.
9. da Silva TE, Gomes NL, Lerário AM, *et al.* Genetic evidence of the association of DEAH-box helicase 37 defects with 46,XY gonadal dysgenesis Spectrum. *J Clin Endocrinol Metab*. 2019;104(12):5923-5934.
10. Wan Y, Yu R, Luo J, *et al.* A novel DEAH-box helicase 37 mutation associated with differences of sex development. *Front Endocrinol (Lausanne)*. 2023;14:1059159.
11. Shimura K, Ichihashi Y, Nakano S, *et al.* DHX37 variant is one of common genetic causes in Japanese patients with testicular regression syndrome/partial gonadal dysgenesis without müllerian derivatives. *Horm Res Paediatr*. 2024. doi:10.1159/000537761
12. de Oliveira FR, Mazzola TN, de Mello MP, *et al. DHX37* and *NR5A1* variants identified in patients with 46,XY partial gonadal dysgenesis. *Life (Basel)*. 2023;13(5):1093.
13. Margiotti K, Libotte F, Fabiani M, Mesoraca A, Giorlandino C. Digenic origin of difference of sex development in a patient harbouring DHX37 and MAMLD1 variants. *Case Rep Pediatr*. 2024;2024(1):4896940.
14. McElreavey K, Jorgensen A, Eozenou C, *et al.* Pathogenic variants in the DEAH-box RNA helicase DHX37 are a frequent cause of 46,XY gonadal dysgenesis and 46,XY testicular regression syndrome. *Genet Med*. 2020;22(1):150-159.
15. Buonocore F, Clifford-Mobley O, King TFJ, *et al.* Next-Generation sequencing reveals novel genetic variants (SRY, DMRT1, NR5A1, DHH, DHX37) in adults with 46,XY DSD. *J Endocr Soc*. 2019;3(12):2341-2360.
16. Zidoune H, Martinerie L, Tan DS, *et al.* Expanding DSD phenotypes associated with variants in the DEAH-box RNA helicase DHX37. *Sex Dev*. 2021;15(4):244-252.
17. Elzaiat M, Jouneau L, Thépot D, *et al.* High-throughput sequencing analyses of XX genital ridges lacking FOXL2 reveal DMRT1 up-regulation before SOX9 expression during the sex-reversal process in goats. *Biol Reprod*. 2014;91(6):153.