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CASO PRESENTATION

Fournier's gangrene in a male patient with insulin-dependent diabetes mellitus: case report

Gangrena de Fournier en paciente masculino con diabetes mellitus insulinodependiente: reporte de caso clínico

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ABSTRACT

Introduction: congenital dacryocele is a rare entity due to nasolacrimal duct obstruction.
Case report: female neonate, 17 days old, born after euthyroid delivery, at term, with no significant prenatal history. She presented with swelling of the right eye since birth, abundant conjunctival secretions and conjunctival hyperemia. Physical examination revealed the presence of an 8 mm diameter tumor in the area of the right lacrimal sac, not painful to palpation, bluish color. Complementary images such as ultrasound were indicated. The diagnosis of congenital dacryocele was determined. Non surgical treatment was decided. Warm compresses were indicated in the area of the affected lacrimal sac for five minutes, three times a day. It was decided to apply antibiotic therapy with tobramycin + dexamethasone in eye drops, one drop every four hours for 10 days. At the end of the treatment, the patient showed improvement, without the need of further interventions. Follow-up by ophthalmology was indicated.
Conclusions: congenital dacryocele is a congenital entity of the lacrimal ducts of low incidence. Its diagnosis is clinical; however, imaging tests are necessary to rule out other entities. Conservative medical treatment may lead to resolution of the entity, with massage and antimicrobial therapy being useful; however, surgical intervention may be required.

Keywords: Lacrimal Duct Obstruction; Infant, Newborn; Congenital; Surgical Procedures, Operative.

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RESUMEN

Introducción: el dacriocele congénito es una entidad poco frecuente por obstrucción del conducto nasolagrimal.
Presentación del caso: neonato, del sexo femenino, de 17 días de nacida, producto de un parto eutócico, atérmino, sin antecedentes prenatales de importancia. Acude a los servicios por presentar inflamación en ojo derecho desde el nacimiento, secreciones conjuntivales abundantes e hiperemia conjuntival. A examen físico se constata presencia de un tumor de 8 mm de diámetro en zona del saco lagrimal derecho, no doloroso a la palpación, de coloración azulada. Se indica como complementario imágenes como ecografía. Se determina el diagnóstico de dacriocele congénito. Se decidió tratamiento no quirúrgico. Se indicó compresas tibias en la zona del saco lagrimal afectado por cinco minutos, tres veces al día. Se decide colocar antibioticoterapia con tobramicina + dexametasona en colirio, indicada una gota cada cuatro horas por 10 días. Al cabo del tratamiento se mostró mejoría, sin necesidad de otras intervenciones. Se indicó seguimiento por consulta de oftalmología.
Conclusiones: el dacriocele congénito constituye una entidad congénita de las vías lagrimales de baja incidencia. Su diagnóstico es clínico, sin embargo, para descartar otras entidades resultan necesarias pruebas de imagen. El tratamiento médico conservador puede llevar a la resolución de la entidad, resultando útil el masaje sumado a terapia antimicrobiana; sin embargo, puede requerirse una intervención quirúrgica.

Palabras clave: Obstrucción del Conducto Lagrimal; Recién Nacido; Congénito; Procedimientos Quirúrgicos Operativos.

INTRODUCTION

Fournier's syndrome, also called Fournier's gangrene, was described in 1764 by Baurienne as a fatal idiopathic necrotizing process, resulting in gangrene of the male genitalia. Later, in 1883, it was detailed for the first time by Jean Alfred Fournier, reporting the appearance of five cases of fulminant gangrene of idiopathic character in the scrotal region and penis, in young male patients, in such a way, he described the first particularities of Fournier's gangrene as: sudden onset and accelerated progression. Over the years, Fournier's gangrene has adopted several names such as "synergistic necrotizing cellulitis", "streptococcal gangrene" and "periurethral phlegmon", which describe an infectious, destructive and fatal disease of the soft tissues.[1,2,3,4,5]

Fournier's gangrene is a rare type I necrotizing fasciitis involving the soft tissues of the perineal, abdominal, external genitalia and perianal region. The main microbial category for Fournier's gangrene is polymicrobial, i.e., it is in the type I category; whereas for monomicrobial necrotizing fasciitis it would be categorized as type II. Therefore, the pathophysiology is carried out by the entry of commensal bacteria into the traumatized area, commonly the perineum. Commensal bacteria such as: Escherichia coli, Staphylococcus spp. and Streptococcus, trigger an inflammatory response, resulting in an obliterating endarteritis of the surrounding vasculature, thus, unleashing tissue ischemia and thrombosis of the vessels, subsequently generating necrosis of the skin, subcutaneous and adjacent tissue. For this reason, the oxygen tension in the tissues is reduced, which leads to a greater proliferation of anaerobic bacteria.[1,3,4]

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Trauma with involvement in the perineal, genital and perianal region intentionally or accidentally, can trigger a fulminant polymicrobial disease, giving quality to necrotizing fasciitis type I, for this reason, it is an established cause in 95 % of cases. The responsible polymicrobial agents include: fungi, gram-negative bacteria (Escherichia coli, Proteus mirabilis, Klebsiella sp, Pseudomonas, Bacteroides, Acinetobacter sp) and gram-positive bacteria (Staphylococcus, Streptococcus, Enterococcus, Clostridium); causing anorectal infection (30-50 %), urogenital (20-40 %) in genital skin (20 %).[4,5,6]

The remaining 5 % of cases of Fournier's gangrene are urological, anorectal, cutaneous and traumatic causes. In adult men they are: urethral strictures, calculi, prostatic massage, perianal, perirectal and ischiorectal abscesses, anal fissures, rectal cancer, inguinal hernia repair, prostatic biopsy, vasectomy, diathermy for genital warts, foreign body anal perforation, penile prostheses, genital piercings, penile injection, steroid enemas and urethral instrumentation. In women: septic abortions, vulvar abscess, Bartholin's abscess, HPV lesions, hysterectomy and episiotomy. In children: circumcision, congenital strangulated inguinal hernia, post-varicella rash and urethral instrumentation.[4]

It has been established that Fournier's gangrene is an unusual condition, accounting for only 0.02% of hospital admissions, it should be noted that the incidence is increasing with the aging of the population. Therefore, epidemiological data show an overall incidence rate of 1,6 cases per 100,000 men/year, showing a peak after the age of 50 years with 3,3 cases per 100,000 men/year. Thus, Fournier's gangrene sustains a higher prevalence in adult men than in women and children; in a ratio of 10:1.[1,3,4,7]

Fournier's gangrene is considered a urological emergency, for which it is opportune to establish an early diagnosis, due to its high mortality rate (20-30 %); in spite of an immediate surgical intervention.[1,3,4,7]

In Ecuador, attention to Fournier's gangrene has generally been sporadic; however, in recent years it has been observed that the incidence and severity has increased significantly, so it is estimated at approximately seven cases per 100,000 male patients.[7] The case of a patient treated for this reason in the "Hospital General del Puyo" during the month of March 2022 is presented.

PRESENTATION OF THE CASE

On March 13, 2022, a 47-year-old male patient, originally from Venezuela, resident of Puyo, mestizo, divorced, was received at the "Hospital General del Puyo", with a personal pathological history of Diabetes Mellitus type 2, diagnosed 10 years ago, under treatment with insulin NPH 15 intramuscular units (IM), which he abandoned three months ago; arterial hypertension diagnosed 4 years ago, under treatment with losartan 100mg every day, which he abandoned three months ago. As surgical history, she reported surgery for diabetic retinopathy.

On interrogation, she reported that a month ago she presented with highly progressive edema of the lower limbs.

Physical examination revealed a temperature (T) of 36,4°C, blood pressure (BP) of 124/70 mmHg, mean arterial pressure (MAP) of 88 mmHg, oxygen saturation (SpO2) of 98 % and inspired oxygen fraction (FiO2) of 21 %. In the central nervous system, the patient was conscious, oriented in PET, with no signs of neurological focalization. In the hemolymphopoietic system, bilateral inguinal adenopathies were found, in the inguinal region there were areas of hyperpigmentation. In the subcutaneous cellular tissue in the lower limbs there was bilateral

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edema of the lower limbs (+++) and lumbar region (+++) with pitting due to suspected arteriosclerosis, and in the urogenital system a pustular abscess was identified in the left testicle.

Laboratory studies showed leukocytosis with a left shift. In imaging studies, marked edema of the scrotal wall was identified with no areas of collection: bilateral epididymitis, left orchitis and bilateral scrotal cellulitis were found in the ultrasound. The color Doppler study showed no signs of ischemia.

After the examination and with the corresponding tests, stage V renal failure was diagnosed, so dialysis was recommended. It was decided to admit him for further study, evaluation by specialties and treatment.

In the weeks following his admission the patient presented hypotension, testicular pain and testicular edema (++/+++), erythema, accompanied by fever (38,5°C) for which broad-spectrum antibiotic therapy (Meropenem) was indicated to maintain a focus on soft tissue sepsis after taking cultures. She also presented with semi-liquid stools on five occasions with no evidence of alteration.

Table 1. Report of evolutionary imaging studies

	Scrotal level	**Color Doppler study**	**Testicular ultrasound**		
22/03/2022	Very thickened soft tissues without defined collections. Bilateral Varicocele Grade I	**Testicles:** increased vascularity, heterogeneous parenchyma.	Diffuse soft tissue edema. Signs suggestive of scrotal cellulitis. Free fluid at pelvic level	Orquiepididimitis Bilateral Funiculitis Bilateral Ateroesclerosis de Miembros Inferiores	Orchiepididymitis. Bilateral Funiculitis Atherosclerosis of Lower Limbs Bilateral
	Bilateral Inguinal Adenopathies Spermatic cord: thickened and heterogeneous appearance bilaterally.	Left/right epididymis: increased vascularity, regular borders, thickened, heterogeneous echostructure.			
	Peri testicular fluid preserved.	Pampiniform plexus at Valsalva maneuver thickened.			
22/03/2022	Very thickened soft tissues with reticular aspect, laminar collections, echogenicity changes at scrotal and suprapubic level.		❖ Epididymitis left. ❖ Bilateral Hydrocele	❖ Bilateral Simple Cyst of Epididymis	❖ Bilateral Varicocele Grade I

The patient does not respond favorably to treatment (Table 1 and 2), where the testicular lesion evolves into a Fournier's Gangrene type necrotizing fasciitis, so surgical resolution is decided.

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Table 2. Evolutionary Blood Biometry

Test	Reference value	March 2022						Report
		Day 16	Day 17	Day 20	Day 21	Day 27	Day 31	
Leukocytes	[4,800-10,800] k/ul	7,14	6,16	27.95	35,00	23,28	26,45	Leukocytosis
Red cells	[3,90-5,20] m/ul	1,9	1,85	1,73	1,77	2,11	1,83	Erythropenia
HGB	[12,1-16,2] g/dl	7	6	5,8	6	9	5,6	Low hemoglobin
HTO	[38,0-48,0]%	21	18,0	16	18,0	27	17	Low hematocrit
VCM	[80,0-100,0] fl	93,4	92,8	93,1	92,5	85,1	92,9	Normal
HCM	[27,0-31,0] pg	28,6	28,7	27,7	27,4	28,9	30,6	Normal
MCHC	[32,0-36,0] g/dl	37,6	33,3	32,6	36,4	34,0	32,9	Normal
Platelets	[150-450] k/ul	372	306	269	263	312	265	Normal
NEU %	[43,0-65,0]%	77,8	79,4	94,3	94,8	87,6	92,3	Neutrophilia
LYM %	[20,5-45,5]%	10,9	8,3	1,3	1,7	4,7	1,2	Lymphopenia
EOS %	[1,0-5,0]%	2,8	2,9	0,1	0,6	0,1	0,1	Eosinopenia
Quantitative PCR	0,00-5,0 mg/l	125,37	211,02	--	279,80	120,22	79,55	Positive PCR

In the surgical resolution reports, it should be noted that the patient was admitted on several occasions with a diagnosis of Fournier's necrotizing fasciitis with projections towards surgical cleaning of the affected area, debridement of skin, subcutaneous tissue and muscle.

In the exploration and surgical findings there is evidence of good quality granulomatous tissue, signs of necrosis in the perineal border and devitalized tissue. The most relevant operative procedures included debridement of devitalized tissue, profuse lavage with 0,9 % saline solution with 3000 ml, drying and control of hemostasis, placement of prostosan and approximation of the wound with anti-tension stitches. These procedures were required repeatedly in all interventions. The patient's treatment ended with the resolution and good evolution of the patient, being discharged.

DISCUSSION

In the case presented, the patient showed typical triggering factors of this process with the subsequent evolution and aggravation of the clinical picture, associated with previous pathologies such as: DM2, nephropathy and subsequent sepsis. In the patient, the onset of the disease and its subsequent aggravation, has a characteristic clinical spectrum: severe toxic picture, scrotal necrosis and delimitation of an erythematous and crepitant area due to gas in the affected area.

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Fournier's gangrene has an indolent onset, but often as the disease progresses the clinical picture begins to appear as a simple abscess or cellulitis with progression to necrotizing soft tissue infection, its common clinical picture includes scrotal pain, swelling and erythema; accompanied by systemic features such as fever, rigidity and tachycardia. Symptoms will exacerbate as time passes, intensifying the picture of pruritus, pain and discomfort.(3,4)

Therefore, exploratory physical examination reveals a purulent discharge, hyperemia, crepitus, skin discoloration, putrid odor, and patches of necrotic tissue with surrounding edema that may progress to florid gangrene. Basically, the diagnosis is reached through the clinic, a complete anamnesis and physical examination, which include a traumatic history in the perineal region or external genitalia. Complementary imaging studies are soft tissue ultrasound, computed tomography (CT) and nuclear magnetic resonance (MRI), which report air in the planes of the soft tissues and help determine the extent of the disease. (3,4)

Although a triggering cause for this disorder is now identified in more than 90 % of cases, most cases are secondary to one of the following mechanisms: underlying urethral genitourinary disorder, inflammatory or infectious anorectal pathology, trauma or injury to the perineal and anogenital skin. It has been suggested that those patients with Fournier's gangrene in whom no etiology is demonstrated (as occurred in the case described), have an underlying undiagnosed genitourinary disorder. There are underlying diseases in up to 90-95 % of cases, with diabetes mellitus (in up to 40-60 % of cases), chronic alcoholism or immunosuppressed patients being considered the most important predisposing factors, in whom the prognosis is less favorable.(8,9)

In general the etiopathogenesis of the disease is not well defined, since after infection a subcutaneous cellulitis limited by the muscular fasciae originates, with subsequent tissue and skin necrosis, as in the case of the patient detailed above, who although he arrived at the hospital unit with several adjoining comorbidities, the trigger of Fournier's gangrene can be associated with an initial picture of pustule of unknown origin, erythema in the testicular region, which with the passage of weeks was deteriorating his condition. Once gangrene has set in, it can progress by 2-3 cm/hour. The involvement of the testicle is unusual, being protected by several tunics and having independent vascularization; this fact occurred in the patient, forcing immediate surgical measures to prevent its progression, making this case interesting for study due to the rapid and lethal infectious dissemination.(10)

Another aspect to take into account is the flora of the urethral region and rectum, being a typically polymicrobial infection. The facultative or strict anaerobic and aerobic bacteria in each patient, there is much bacterial synergism in the development of this infection. This means that the input of the isolated germs varies, with bacteria such as: Clostridium; if it is urinary gram-negatives such as Streptococcus and Staphylococcus are isolated; if it is cutaneous, Staphylococcus is cultured. The most frequently isolated aerobe is E. coli and the anaerobe Bacteroides fragilis. Overall, the most frequent germ is E. coli, so the complementary laboratory and radiological tests were very supportive, always highlighting leukocytosis with left shift, hyperglycemia (in the case of the patient who was diabetic), compensated metabolic acidosis, increased urea and creatinine (due to renal pathology). Also, imaging tests were useful to confirm the diagnosis and evaluate the response to treatment; those used were simple radiography and ultrasound (8,11,12).

The fundamental therapeutic principles to control Fournier's gangrene are urgent hemodynamic stabilization, early surgical debridement and IV broad-spectrum antibiotic therapy, together with a multidisciplinary approach, which will contribute to lower the mortality rate and increase the possibilities of surgical reconstruction of the affected areas. That is why timely diagnosis helps us to perform a correct approach, however, Fournier syndrome not treated in time leads to

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serious clinical conditions such as deformities, urinary tract infection, wound infection, renal failure, thromboembolic disease of the lower extremities and septic metastases.[1,4,13,14,15]

Treatment was multidisciplinary and urgent, combining medical and surgical treatment and maintenance life support. In addition to the broad-spectrum antibiotic treatment that was performed intravenously. As well as the use of fourth generation cephalosporins (cefepime), given its effectiveness against resistant germs. Surgical debridement on multiple occasions improved the access, visualization and cleaning of the affected area due to the progress of the gangrenous process. In addition, radical extirpation of necrotic tissue was performed, with wide debridement until healthy tissue was found, and then the exposed tissue was covered with compresses. Reinterventions were performed according to the evolution and appearance of the wound.

CONCLUSIONS

Despite technical advances and broad-spectrum antibiotics, mortality is currently high in this type of ailment, which reflects the reality of a serious disease and a true urological emergency. Therefore, timely diagnosis at the first medical contact and urgent referral to the hospital for extensive debridement are essential to improve the prognosis.

Conflicts of Interest
The authors declare no conflicts of interest in relation to the present investigation.

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Declarationo Authorship
All authors participated in the conceptualization, research, writing - initial draft, writing - revision and editing.

BIBLIOGRAPHIC REFERENCES

1. He X, Xiang X, Zou Y, Liu B, Liu L, Bi Y, et al. Distinctions between Fournier's gangrene and lower extremity necrotising fasciitis: microbiology and factors affecting mortality. Int J Infect Dis [Internet]. 2022 [citado 27/09/2022]; 122: 222-9. Disponible en: https://pubmed.ncbi.nlm.nih.gov/35598736/

2. Almaiman SS, Alfraidi OB, Alhathal NK. Fulminant corporal infection induced by Fournier gangrene: A case report with unusual presentation. Urol Case Rep [Internet]. 2022 [citado 27/09/2022]; 40: 101942. Disponible en: https://pubmed.ncbi.nlm.nih.gov/34824979/

3. Basukala S, Khand Y, Pahari S, Shah KB, Shah A. A rare case of retroperitoneal extension in Fournier's gangrene: A case report and review of literature. Ann Med Surg (Lond) [Internet]. 2022 [citado 17/09/2022]; 77: 103595. Disponible en: https://pubmed.ncbi.nlm.nih.gov/35638004/

4. Calderón W, Camacho JP, Obaíd M, Moraga J, Bravo D, Calderón D. Tratamiento quirúrgico de la gangrena de Fournier. Rev. Cir [Internet]. 2021 [citado 17/09/2022]; 73(2): 150-157. Disponible en: https://www.scielo.cl/scielo.php?pid=S2452-45492021000200150&script=sci_arttext

ISSN: 1561-3194 **RNPS:** 1877
Rev Ciencias Médicas. 2023; 27(S1): e6008

5. Sobrinho AGB, Geraldelli TV, Bitencourt EL, Brandão RGD, Vaz GP, Galvão JA, et al. Síndrome de fournier em idoso: um relato de caso. Revista de Patologia do Tocantins [Internet]. 2021 [citado 15/09/2022]; 8(3): 71-4. Disponible en: https://sistemas.uft.edu.br/periodicos/index.php/patologia/article/view/12286

6. Cruz Jordán. V, Moncayo Anaslema F, Beltran Alejandro M. Gangrena de Fournier complicada, en hospital de tercer nivel. REVFCM-UG [Internet]. 2022 [citado 21/12/2022]; 3(2): 26-31. Disponible en: https://revistas.ug.edu.ec/index.php/fcm/article/view/1820

7. Egas Ortega W, Granja Rousseau I, Luzuriaga Graf J, Egas Romero W, Moncayo C. Características de los casos de gangrena de Fournier atendidos en el Hospital Luis Vernaza de Guayaquil-Ecuador. Rev Med Vozandes [Internet]. 2017 [citado 17/09/2022]; 28: 27-32. Disponible en: http://fi-admin.bvsalud.org/document/view/843xu

8. Viel-Sánchez P, Despaigne-Salazar R, Mourlot-Ruiz A, Rodríguez-García M, Martínez-Arzola G. Gangrena de Fournier. Revista Cubana de Medicina Militar [Internet]. 2020 [citado 19/09/2022]; 49(1): 206-213. Disponible en: https://revmedmilitar.sld.cu/index.php/mil/article/view/333

9. Díaz-Martínez AR, De los Cobos-Gutiérrez E, Hernández-Ávila PH, Arias-de la Cruz Y, Hernández-González N. Caracterización clínica de pacientes con gangrena de Fournier del Hospital General Docente "Dr. Agostinho Neto", 2008-2018. Rev Inf Cient [Internet]. 2021 [citado 19/09/2022]; 100(4): e3528. Disponible en: https://revinfcientifica.sld.cu/index.php/ric/article/view/3528

10. Vargas Rubio T, Mora Agüero S de los Ángeles, Zeledón Aguilera AS. Gangrena de Fournier: generalidades. Rev. méd. sinerg [Internet]. 2019 [citado 19/09/2022]; 4(6): 100-17. Disponible en: https://www.revistamedicasinergia.com/index.php/rms/article/view/217

11. Lombardo Vaillant TA. Clinical-epidemiological study on Fournier's gangrene in a Luanda hospital. From January 2016 to December 2021. Medisur [Internet]. 2022 [citado 19/09/2022]; 20(3): 515-526. Disponible en: http://scielo.sld.cu/scielo.php?script=sci_arttext&pid=S1727-897X2022000300515&lng=es

12. Yeniyol CO, Suelozgen T, Arslan M, Ayder AR. Fournier's gangrene: experience with 25 patients and use of Fournier's gangrene severity index score. Urology [Internet]. 2004 [citado 19/09/2022]; 64(2): 218-22. Disponible en: https://pubmed.ncbi.nlm.nih.gov/15302463/

13. Lacruz-Pérez B, García-Montero A, Guinot-Bachero J. Abordaje postquirúrgico de un caso de gangrena de Fournier desde atención primaria. Enfermería Dermatológica [Internet]. 2019 [citado 19/09/2022]; 13(37): 52-58. Disponible en: https://dialnet.unirioja.es/servlet/articulo?codigo=7088023

14. Ochoa DXT, Montesinos CEF, Ortiz GIM. Perfil bacteriológico e regimes antibióticos utilizados no tratamento da Gangrena de Fournier. Braz. J. Hea. Rev. [Internet]. 2023 [citado 19/02/2023]; 6(1): 3382-91. Disponible en: https://ojs.brazilianjournals.com.br/ojs/index.php/BJHR/article/view/57287

15. Zakariya-Yousef BI, Trujillo Díaz N, de la Herranz Guerrero P. Grangrena de Fournier secundaria a un absceso inguinoperineal por Acidaminococcus intestini y Streptococcus gallolyticus spp. pasteurianus. Rev Esp Quimioter [Internet]. 2021 [citado 19/02/2023]; 34(6): 679-681. Disponible en: https://www.ncbi.nlm.nih.gov/pmc/articles/PMC8638768/