Scientific Research Publishing

Journal of Biosciences and Medicines, 2023, 11, 26-31
https://www.scirp.org/journal/jbm
ISSN Online: 2327-509X
ISSN Print: 2327-5081

Infectious Pneumonitis Revealing an Anal Imperforation with Recto-Vaginal Fistula: A Case of Late Discovery in a 70-Year-Old Woman

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How to cite this paper: Assane, S., Massamba, B., Rokhaya, D., Sarr, A.A.C., Dalahata, B., Vahana, D., Maimouna, T.E. and Mamadou, C. (2023) Infectious Pneumonitis Revealing an Anal Imperforation with Recto-Vaginal Fistula: A Case of Late Discovery in a 70-Year-Old Woman. *Journal of Biosciences and Medicines*, **11**, 26-31.
https://doi.org/10.4236/jbm.2023.111004

Received: November 5, 2022
Accepted: January 10, 2023
Published: January 13, 2023

Abstract

Introduction: Anorectal malformations concern all anomalies of the terminal part of the digestive tract often diagnosed in the perinatal period. We report a clinical case of anorectal malformation discovered incidentally and very late at 70 years, following complications from the infectious syndrome. **Observation:** 70-year-old female patient, housewife, uneducated was referred to the hospital for acute febrile, cough, and mental confusion associated with chronic pelvic pain. The genito-anal examination showed an anal imperforation associated with a recto-vaginal fistula complicated by cervico-vaginitis. The evolution during hospitalization was favorable, but the patient died at home, 3 weeks after discharge. **Conclusion:** The discovery of anorectal malformation in adults is rare and exceptional in an elderly subject. This case demonstrates once again the challenges encountered in developing countries on the socio-economic and health levels.

Keywords

Anorectal Malformation, Elderly Subject, Dakar, Senegal

1. Introduction

Anorectal Malformations (ARMs), of which anal imperforation is a part, concern all anomalies in the placement of the terminal part of the digestive tract. Fistulas are frequently associated and can be: recto-vesical, recto-prostatic, recto-urethral, or perineal in boys, and recto-vaginal, recto-vestibular, or perineal in girls [1]. Although they are rare, they are the most frequent malformations affecting the perineal sphere [2]. Their prevalence varies from 1/1500 to 1/10,000 live births

[3]. The distribution is heterogeneous with a higher prevalence in developed countries [4] [5]. They are often diagnosed in the perinatal period during a routine clinical examination. Pelvic Magnetic Resonance Imaging (MRI) is the gold standard for ante- and post-natal diagnosis of imperforations with fistula [6]. However, cases of delayed diagnosis and management have been reported, especially in developing countries [7]. To our knowledge, no case of such late revelation (70 years) is reported in the literature.

In this work, we report a case of anal imperforation, associated with a recto-vaginal fistula in a 70-year-old patient. It is original by its very late age of discovery and its atypical mode of revelation by an infectious pneumonitis.

2. Observation

This was a 70-year-old female patient, housewife, illiterate, mother of 3 boys all alive and well after 3 gestures and 3 pares with vaginal delivery, one of which was in the hospital. Elsewhere, she reported chronic episodic pelvic pain since childhood which would have motivated numerous medical consultations and traditionally without any real improvement. She was also known to have high blood pressure for 20 years and was poorly monitored. She was referred to us from a local health center for exploration and follow-up of acute mental confusion associated with a refusal to eat, subacute cough, and fever of allegation evolving progressively for about a month, in a context of family conflict and abuse.

The clinical examination on admission revealed: a systemic inflammatory response syndrome, a bilateral pulmonary condensation syndrome predominantly on the left, irritation of the lower urinary tract; stage 2 inter gluteal eschar, erythema of the groin folds, clinical anemia, purulent leucorrhea, moderate extracellular dehydration and an alteration of the general condition.

She had also unfavorable geriatric conditions with frailty (FRIED score 5/5), progressively worsening loss of functional independence (Katz Activities of Daily Living 2.5/6), severe malnutrition with a Mini Nutritional Assessment score below 17, and probable depression (Mini Geriatric Depression Scale 3/4).

The biological investigations are summarized in **Table 1**

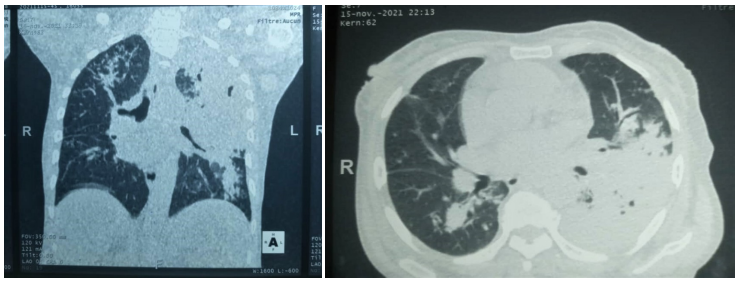
Table 1. Summary table of biological abnormalities.

Abnormalities of biological investigations
o Hyper leukocytosis with 17,000 elements/mm^3
o Lymphopenia at 400 U/mm^3
o Microcytic anemia at 8.6 g/dl
o Major elevation of C-Reactive Protein > 200 mg/l
o Serum hypo albumin at 17.7 g/L
o A severe alteration of the renal function with Glomerular Filtration Rate (GFR) according to MDRD at 32.5 ml/min

Chest Computed Tomography (CT) scan showed bilateral bronchial disease (**Figure 1(a)**, **Figure 1(b)**). The diagnosis of community-acquired pneumonia was made as follows.

In the context of the search for an infectious gateway, a genital infection site was found and the thorough examination revealed an anal imperforation associated with a recto-vaginal fistula complicated by cervico-vaginitis (**Figure 2**).

For imaging investigations, pelvic ultrasound showed intra-cavity fluid retention. Magnetic Resonance Imaging (MRI) confirmed a basal posterior ano-vaginal fistula associated with an anal imperforation (**Figure 3(a)**, **Figure 3(b)**).



(a) (b)

Figure 1. Images of bilateral bronchopneumopathy predominantly on the left on CT.

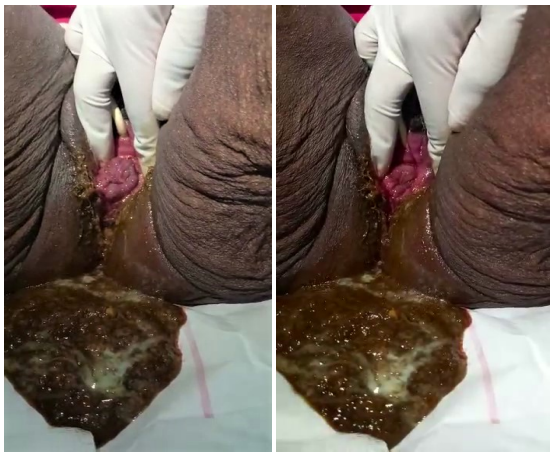
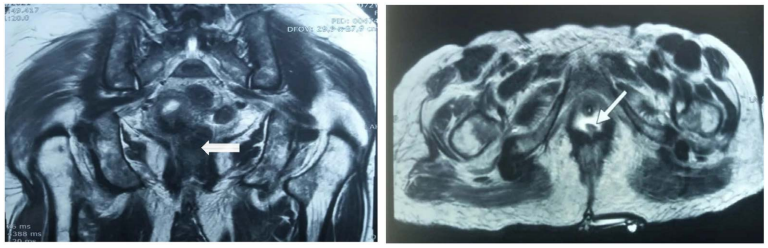


Figure 2. Emission of stool through the vaginal orifice with cervical prolapse.



(a) (b)

Figure 3. (a) The white arrow showing the anal imperforation (MRI); (b) White arrow showing the seat of the ano-perineal fistula (MRI).

The therapeutic approach was carried out following a medical-surgical and psychological approach. The medical approach was a bi-antibiotic therapy with metronidazole and amoxillin for genital and pulmonary purposes. An antidepressant treatment with a selective serotonin reuptake inhibitor (Sertraline) and a preventive anticoagulation were also initiated. Supportive and family psychotherapy was provided. The surgical technique considered was a colostomy then proctoplasty and cure of the fistula after control of the comorbidities.

During hospitalization, the clinical and biological evolution was favorable, allowing her to be discharged after 24 days of hospitalization while waiting for surgery. She died at home, 3 weeks after her discharge. The suspected etiology was sepsis or pulmonary embolism.

The written consent of the patient's guardian and the approval of the research ethics committee of our institute were obtained.

3. Discussion

Anorectal malformations are a heterogeneous group of abnormalities of the rectal canal development. A true birth incidence of ARM is difficult to obtain because there are no formal birth registries in most parts of Africa and most reports in the literature are hospital based. The incidence remains low in general and is somewhat higher in some developing countries [3]. In the Democratic Republic of Congo, they represented 10.6% of all malformations [6].

Their etiology is still very unclear and certainly multifactorial. A genetic component is sometimes found [8]. Maternal consumption of toxic substances, as well as environmental factors such as maternal exposure to cleaning agents, especially solvents, are factors that have been implicated in the development of this type of malformation [9]. We could not be informed by the patient about the notion of fever during the first trimester of her mother's pregnancy or about maternal exposure to certain incriminating toxic products. There were no similar malformations reported in other members of her family.

In Africa, most studies show a clear male predominance of 55% to 71% [10]. In Tanzania, the study by Mfinanga [11] identified a female predominance.

An association with other malformations is frequent (9% - 44%) [10]. In our patient there was no other obvious malformation found. The distal form found in our patient would be the most frequent type [6].

ARMs occur early in the first weeks of embryonic life. The diagnosis is usually made at birth or within the first 4 weeks of birth (19% - 89%) [10]. The presentation of anorectal malformations in adulthood is rare, they appear in a sporadic pattern [8]. In a large African study [11], the latest form was found in a 25-year-old female patient [12]. To our knowledge, no case of such late onset (70 years) has been reported in the literature. Delayed presentation in our case can be explained some times by discharge through the vagina fistula. In addition, in developing countries such as ours, socioeconomic factors, low intellectual level and misinformation are among the main reasons for delayed diagnosis. Coming from an

uneducated family, our illiterate patient had benefited from numerous medical and traditional consultations due to her recurrent illnesses. Her mother, on the other hand, told her to keep quiet about her ailments because "a good woman must learn to endure". This shows once again the challenge of medical education in our countries. This lack of education and information may explain the use of traditional treatments. Poverty, which makes the number of health personnel and the technical platform less effective, is also a factor incriminated in the delay of diagnosis and treatment [13]. Delay in diagnosis has been correlated with poorer outcome and a higher mortality [11]. The circumstances of a late diagnosis of an anorectal malformation can be digestive complications, such as intestinal obstruction or infectious complications, as in the case of our patient who presented an infectious cervico vaginitis associated with septicemia. Other associated complications such as malnutrition and depression could be multifactorial: due to pain during stool emission with a stool avoidance behavior, social isolation, maltreatment, etc.

The circumstances of discovery of this malformation are quite particular; the diagnosis of infectious pneumonitis with a common germ does not presage the discovery of an anal imperforation.

Management had a double objective: anatomical and functional. It is necessary to ensure the maintenance of urino-fecal continence. Several surgical interventions may be necessary. The distal forms seem to have a more favorable functional prognosis than the other forms [7]. The treatment for this form in adults can be done by anorectal lowering with a V-Y anoplasty allowing an anatomically correct location of the anus and an acceptable continence for the patient [14]. In our case, a 2-stage surgical treatment (colostomy + proctoplasty with cure of the fistula) was considered, after psychological preparation.

The prognostic factors were essentially described in children and adults and not in the elderly. The functional prognosis depends much more on the type of anorectal malformation and the quality of the surgical repair performed [10]. Similarly, aging of the pelvic musculature (muscular atrophy or hypotrophy, fibrosis, etc.) could also have a negative impact on the functional prognosis with often disappointing results. However, the immediate vital prognosis depends mainly on the severity of the associated malformations. In our frail elderly patient, the deleterious geriatric conditions such as malnutrition, depression, loss of autonomy, and immobilization darkened the vital prognosis with major infectious and thromboembolic risks. The latter was even incriminated in the death of our patient. This clinical case raises the question of the weight of tradition in the dissemination of certain anomalies or malformations and constitutes support for the implementation of interventions to break taboos.

4. Conclusion

The discovery of anorectal malformation in adults is rare and exceptional in an elderly person. This delay in diagnosis, which often has serious consequences, is

essentially due to socio-economic and cultural factors in our context. This shows once again the challenges encountered in developing countries in terms of health care.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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