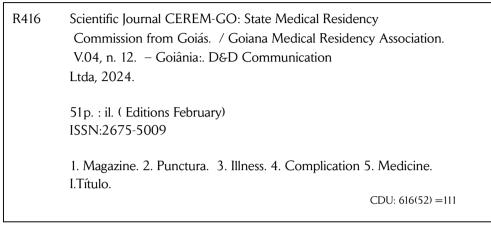
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# WALDEMAR NAVES DO AMARAL TÁRIK KASSEM SAIDAH

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## **ORIGINAL ARTICLE**

# EVALUATION OF PREDICTORS IN-HOSPITAL MORBIMORTALITY IN PATIENTS UNDERGOING URGENCY AND EMERGENCY SURGERY

ANTÔNIO FERNANDO CARNEIRO<sup>1</sup>, TOLOMEU CASALI<sup>1</sup>, QUÉTILAN LOPES<sup>1</sup>

#### ABSTRACT

According to data from the WHO, 254 million surgeries are performed every year around the world. Of these, around 7 million patients suffer some post-operative complication and one million die. Urgent and emergency surgeries are identified as independent predictors of mortality and perioperative care is essential for the patient's good clinical development. This study aimed to characterize the population undergoing this type of procedure at HC-UFG and identify clinical and surgical factors related to higher mortality in the intraoperative period up to the seventh postoperative day. This is an analytical, prospective study. Data were obtained by reviewing patient records and printed anesthesia records. In the assessment of postoperative complications, the Postoperative Morbidity Survey scale, developed and validated specifically for this purpose, was used. 71 patients were followed, aged 41.7 +/- 24.5 years. 42.2% had some pre-operative risk situation. The presence of complications was observed in 40.8% and 8.4% died. Anemia (odds ratio [OR] 16.0, p < 0.05), major surgery (OR 13.3, p < 0.05), acute or acute chronic renal failure (OR 16.8, p < 0.05) and September (OR 0.5, p < 0.05) were significant for the occurrence of complications. These results may reflect the high complexity of the institution's patients. Knowing the profile of patients served helps in defining management strategies and creating specific lines of care for high-risk groups.

#### KEYWORDS: PERIOPERATIVE MORTALITY; NON-ELECTIVE SURGERIES; PERIOPERATIVE CARE.

#### INTRODUCTION

According to the World Health Organization (WHO), approximately 254 million surgeries are performed worldwide each year. Of these, about seven million patients experience some postoperative complication, and approximately one million die during or after the surgical procedure <sup>1</sup>. When considering only procedures performed on an urgent or emergency basis, the numbers are even more alarming, with some studies showing values that are 10 times higher than those found for elective procedures, in terms of mortality and surgical complications <sup>2</sup>.

In many healthcare services, a series of challenges are encountered to provide efficient and quality care for critically ill patients. Among them, we can highlight the delayed hospital flow between the surgical indication and the procedure, including the internal bed regulation system, preoperative clinical optimization, routine performance of safe surgery checklists, among others <sup>3</sup>.

The creation of institutional protocols emerges as an appropriate way to improve care and the quality of assistance provided. For this purpose, it is necessary to study the demand for patients undergoing emergency procedures, as well as the logistics and specific resources of each service <sup>4</sup>. Thus, the use of instruments developed to analyze perioperative risk is indispensable for a previous institutional analysis and may not reflect the reality of a location under study.

Considering that emergency surgeries are identified as independent predictors of mortality and that perioperative care is fundamental for the good clinical outcome of the patient, this study aimed to characterize the population undergoing this type of procedure at the Hospital das Clínicas of the Federal University of Goiás (HC-UFG).

Characterization of the study

This is an analytical, prospective and quantitative cohort study.

Characterization of research locations

The research was carried out at the Hospital das Clínicas of the Federal University of Goiás (HC UFG).

Characterization of the population

The sampling plan was based on the number of urgent and emergency surgeries that took place in the HC UFG surgical center.

Data Collection Instrument

Trans- and postoperative data were obtained by re-

1 Hospital das Clínicas da Universidade Federal de Goiás

#### ADDRESS

QUÉTILAN SILVA LOPES Endereço: Av. 1ª (Avenida Qd 68 Lt Ar 1), Goiânia, GO, 74605-020. Email: quetilan@hotmail.com. viewing the patients' medical records and printed anesthesia records, for up to 7 days postoperatively. Among the data related to surgery, the following were evaluated: surgical size; the time between surgical indication and its completion; the duration of the procedure and the need for reintervention.

In the assessment of postoperative complications, the validated Postoperative Morbidity Survey (POMS) scale was used, consisting of nine domains, which record morbidity according to the presence of established criteria.

#### Data analysis method

Fisher's test and Chi-square were used to compare categorical variables and Student's t-test was used for continuous variables. In order to select the predictor variables with the least possibility of overfitting, the logistic regression technique was used. A p value < 0.05 was considered. BioStat 5.0 was used.

Ethical aspects of the research

The study was carried out after consideration by the Research Ethics Committee (CEP) of HC UFG (Opinion report Number: 5,540,241). Participants or guardians signed the Free and Informed Consent Form before undergoing the study.

#### RESULTS

Demographic data, clinical characteristics and surgical procedures.

During the study period, 71 patients were followed, the mean age was 41.71 (+/- 24.58), 63% female, 16.9% ASA I, 45% ASA II, 32.3% ASA III and 5% ASA > or = 4.

Among the surgeries, 47.8% were stratified as small, 29.5% as intermediate and 22.5% as major. The average time to perform the procedure was 1.8 +/- 0.98 hours. Regarding the time between the surgical indication and the procedure, 16.9% < 12 hours, 33.8% between 12 and 24 hours, 23.9% between 24 and 48 hours and 25.3% > 48 hours. The surgical specialties that received the most attention were orthopedics (25.3%), general surgery and urology (16.9%) and neurosurgery (14%). Postoperative complications were observed in 40.8% of the sample and 8.4% died.

Morbidity and mortality and postoperative complications. Postoperative mortality was 8.4%, and 42.2% had one or more preoperative risk situations present. Among the preoperative clinical risk situations, anemia, sepsis, AKI or acute CKD, acid-base disorder, hemodynamic instability, water-electrolyte disorder, and neoplasia stood out, in order of prevalence. Anemia, the most common, was present in 65.5% of patients who presented peri- and postoperative complications.

Among the patients with peri- and postoperative complications, the presence of sepsis (42.2%) and acute kidney injury (AKI) or acute exacerbation of chronic kidney disease (CKD) (27.5%) was also noteworthy. Regarding postoperative follow-up, complications were recorded using the POMS scale in 40.8% of patients, with the following being most common: pain (16.9%), respiratory (15.4%), hematologic (15.4%), and infectious (14%).

#### **Risk predictors**

The significant variables identified by univariate analysis or those with the highest plausibility of being associated with the outcome were included in the logistic regression technique. This strategy was adopted to reduce the possibility of overfitting due to the small number of events compared to the possible predictors. The ASA classification was excluded from the model, despite being universally accepted and having a defined prognostic value, as it is composed of the clinical factors defined in the study as preoperative clinical risk situations. Age was grouped into age groups because it showed a non-linear behavior and still did not enter the final model. Anemia (OR = 16.0; 95% CI 4.72 - 54.57), AKI or acute exacerbation of CKD (OR = 16.8; 95% CI 1.96 - 143.65), and sepsis (OR = 20.5; 95% CI - 4.13 - 101.4) were kept as significant patient-related risk factors for perioperative complications within 7 days. Among the surgical factors, only the large surgical procedure category (OR = 13.3; 95% CI 3.3 - 53.4) was related to the outcome after logistic regression.

#### DISCUSSION

Your study confirmed a high presence of postoperative complications in patients undergoing non-elective surgeries (40.8%). The mortality rate found (8.4%) was very close to the data found in the national literature on critically ill patients admitted to Intensive Care Units (ICU) in the postoperative period of non-cardiac surgeries <sup>5</sup>. This result reflects the high complexity of the population treated (37.3% of patients with ASA score  $\geq$  III) and mirrors the difficulty of access and early diagnosis of surgical diseases in the population served by the Brazilian Unified Health System (SUS).

A study conducted at the Hospital das Clínicas in Porto Alegre, also considered a reference for tertiary level healthcare, evaluated the 30-day postoperative mortality of 187 patients undergoing non-elective surgeries over a two-month period. The study found a mortality rate of 14.4%. Advanced age, patient severity, and surgical complexity were significant factors associated with this primary outcome <sup>6</sup>.

In order to seek strategies to improve outcomes for surgical patients, this study aimed to examine the clinical and surgical factors involved in the higher incidence of complications and death in emergency surgeries. Preoperative risk situations such as anemia, AKI or CKD, and sepsis made patients more susceptible to postoperative complications and death. Anemia is a common finding in critically ill patients and those undergoing high-risk surgical procedures (around 60% of those admitted to the ICU)<sup>7</sup>. A recent systematic review demonstrated that anemia is an important risk factor for the occurrence of anastomotic leakage and postoperative infection, in addition to being associated with hemodynamic instability and tissue hypoperfusion in critically ill patients <sup>8</sup>.

The medical literature does not define a minimum acceptable preoperative hemoglobin value. However, it is clear that chronic anemia is much more tolerable than acute anemia. The decision to perform a preoperative transfusion should take into account various factors, such as the type of surgery, anticipated blood loss, coexisting disease, and duration of anemia <sup>9</sup>.

Iron deficiency anemia is the most common type of anemia. In this situation, oral or intravenous iron supplementation in the preoperative period is the preferred treatment, which may even reduce transfusion needs <sup>9</sup>.

The high prevalence of sepsis among patients who developed surgical complications (48.2%) corroborates the profile of patients treated in the emergency room of this hospital, which does not include trauma. Brazilian studies show that sepsis is present in up to 73% of deaths and is responsible for the high prevalence of dysfunction of multiple organs and systems <sup>5</sup>. To minimize this outcome, it is important to administer antibiotics early and fluid resuscitation in the first hours to prevent tissue hypoperfusion <sup>10</sup>.

Acute AKI or CKD was also another factor identified in our study. Its perioperative incidence has variable etiology, however, for all cases, renal failure is associated with mortality rates of 60% to 90%. Postoperative renal dysfunction is related to a higher incidence of gastrointestinal bleeding, respiratory infection and sepsis. The best evidence for treatment involves maintaining normovolemia <sup>11</sup>.

The large size variable was an independent predictive factor for the occurrence of perioperative complications and death. The data presented corroborate current literature <sup>12</sup>. It is known that large emergency surgeries, such as abdominal surgeries, are accompanied by various factors that increase the risk of postoperative complications, especially in more vulnerable populations, such as fasting, use of multiple drugs, immobility, use of nasogastric tubes, and indwelling urinary catheters <sup>3</sup>. An audit conducted in 35 hospitals in the United Kingdom showed a high mortality rate (14.9%) in patients undergoing laparotomies. This evidence led to the creation of the NELA project - National Emergency Laparotomy Audit, which consists of a series of pre-, intra-, and post-operative measures to improve outcomes in this population. Among these measures are the surgeon-led care plan and prompt diagnostic definition, formal access to risk assessment for death and complications, early administration of antibiotics, and early surgery <sup>14</sup>.

The time interval between surgery indication and the

procedure, although an important factor in the context of emergency surgeries, was not significant for the occurrence of postoperative complications and mortality. This may be related to the fact that structured preoperative management minimizes the impact of identified risk situations and appears to be more relevant than the rapid performance of surgery.

The average time to perform the procedure was 1.8 +/-0.98 hours. Studies show that elective surgeries lasting more than 2.1 hours are an independent risk factor for complications and increased hospital stay. In view of this, the importance of supervision of resident doctors by preceptors during the entire period of surgical intervention is highlighted <sup>15</sup>.

Our cohort had a limited number of patients, although the number of individuals who developed postoperative complications was quite significant. This study can contribute significantly to standardizing care and helping to define management priorities.

It is also worth highlighting that the study was carried out in a tertiary hospital in a developing country. Access to the service is difficult and, together with the severity of the disease stage at the time of admission, may have influenced the results. The literature shows that there is an association between perioperative mortality in patients undergoing general anesthesia and the Human Development Index (HDI), with mortality being higher in developing countries compared to developed countries <sup>16</sup>. These studies demonstrate the importance of the organization of health systems in post-surgical outcomes. Data observed in American hospitals, for example, showed that post-surgical survival is higher in those services that recognize the most seriously ill patients early, despite the number of complications being similar between institutions <sup>17</sup>.

Therefore, it can be concluded that improving outcomes depends mainly on two factors: recognition of patients at higher risk and early treatment of complications. Factors that may be related to the difficulty in recognizing and treating complications include the high volume of patients at increased risk, reduced nursing staff, communication failure and lack of risk escalation <sup>17</sup>.

There are some algorithms that more practically measure the risk of complications and death in non-cardiac surgery, which may vary according to the patient's morbidities and clinical condition. To calculate the risk, data about the person are considered, such as age, physical fitness, history, physical examination of the patient, laboratory tests and the type of surgery they will undergo.

In most hospitals, as is the case at HC UFG, obtaining an operating room for emergency surgery depends on dialogue and negotiation. In others, emergency surgeries are performed on a first-come, first-served basis <sup>18</sup>.

Recognizing failures during the preoperative patient care process is important for developing measures to optimize the outcome and reduce fragmentation of care.

- 1. WHO Guidelines for Safe Surgery 2009. Safe Surgery Saves Lives. Geneva: World Health Organization; 2009. ISBN-13: 978-92-4-159855-2.
- Adriene S, Betânia N, Luiza AF, Sávio CP, Jairo ADS, Elaine AF, Patrícia WG, Wolnei C, Luciana PCS. Preditores de mortalidade intra-hospitalar em pacientes submetidos a cirurgias não eletivas em um hospital universitário: uma coorte prospectiva. Rev Bras Anestesiol. 2018;68(5):492-498.
- Gouveia JL, Ruiz PBO, Silveira DM, Calil ASG, Vilela RPB, Ruiz PBO. Indicador de qualidade: taxa de mortalidade intra-operatória e pós-operatória. Rev. Sobecc, São Paulo. Out./dez. 2020; 25(4): 234-240.
- Ben AG, Alan MB, Elke K, Louise C, David Y, Helen M, Karen K, Gerard RD. Validade da Pesquisa de Morbidade Pós-Operatória após reparo de aneurisma de ao mesmoza abdominal — um estudo observacional prospectivo. Medicina Perioperatória. Artigo:10 (2015).
- Lobo SM, Rezende E, Knibel MF, et al. Epidemiologia e desfecho de pacientes cirúrgicos não cardíacos em unidades de terapia intensiva no Brasil. Rev Bras Ter Intensiva. 2008;20:376-84.
- Stahlschmidt A, Novelo B, Freitas LA, Passos SC, Dussán-Sarria JA, Félix EA, Gamermann PW, Caumo W, Stefani LPC. Preditores de mortalidade intra-hospitalar em pacientes submetidos a cirurgias não eletivas em um hospital universitário: uma corte prospectiva. Rev Bras Anestesiol. 2018;68(5):492-498.
- 7. Hajjar LA, Fukushima JT, Almeida JP, et al. Strategies to reduce blood transfusion: a Latin-American perspective. Curr Opin Anaesthesiol. 2015;28:81-8.
- Van Rooijen S, Huisman D, Stuijvenberg M, et al. Intraoperative modifiable risk factors of colorectal anastomotic leakage: Why surgeons and anesthesiologists should act together. Int J Surg. 2016;36 Pt A:183-200.
- Semião C, Nogueira C, Campos J, Coelho A, Augusto R, Coelho N, Pinto E, Ribeiro J, Peixoto J, Canedo A. Anemia Pré-Operatória como Factor Predictor de Morbilidade — Análise Retrospectiva de um Departamento de Cirurgia Vascular. Publicação Oficial SPACV. 2020; 16 (3).
- Rhodes A, Evans LE, Alhazzani W, et al. Surviving sepsis campaign: international gudelines for management of sepsis and septic shock: 2016. Intensive Care Med. 2017;43:304-77.
- Sear JW. Kidney dysfunction in the postoperative period. Br J Anaesth. 2005;95:20-32.
- Jakobson T, Karjagin J, Vipp L, et al. Postoperative complications and mortality after major gastrointestinal surgery. Medicina. 2014;50:111-7.
- Merani S, Payne J, Padwal RS, et al. Predictors of in-hospital mortality and complications in very elderly patients undergoing emergency surgery. World J Emerg Surg. 2014;9:43.
- Saunders DI, Murray D, Pichel AC, et al. UK emergency laparotomy network. variations in mortality after emergency laparotomy: the first report of the UK Emergency Laparotomy Network. Br J Anaesth. 2012;109:368-75.
- Grocott MP, Pearse RM. Prognostic studies of perioperative risk: robust methodology is needed. Br J Anaesth. 2010;105:243-5.
- Pearse RM, Moreno RP, Bauer P, et al. Mortality after surgery in Europe: A 7 day cohort study. Lancet. 2012;380:1059-65.
- Johnston MJ, Arora S, King D, et al. A systematic review to identify the factors that affect failure to rescue and escalation of care in surgery. Surgery. 2015;157:752-63.
- Costa ADS Jr. Assessment of operative times of multiple surgical specialties in a public university hospital. Einstein (São Paulo). 2017;15(2):200-5.

# "EPIDEMIOLOGICAL ANALYSIS OF REPORTS OF SELF-INFLICTED INJURIES IN THE CITY OF ANÁPOLIS (GO), FROM 2018 TO 2022"

PEDRO RODRIGUES; THAYNARA NAVES

#### ABSTRACT

Introduction: Self-inflicted injuries have been increasing in numbers over recent years, posing a challenge for the development of public health policies Objectives: Identify the epidemiological scenario of notifications of self-inflicted injuries and their variables, in the city of Anápolis-GO, between the years from 2018 to 2022

Methods: This is a cross-sectional, retrospective study, referring to data on notifications of self-inflicted injuries obtained through access to the Notifiable Diseases Information System (SINAN) platform, in DATASUS

Results: It was observed that the scenario of notifications of self-inflicted injuries in the city of Anápolis (GO) showed a statistical increase between 2018 and 2022. There was a predominance of notifications in females, in the mixed-race population, between the ages of 10-14 years and in educational levels of incomplete grades 5 to 8. When it comes to the means used, there was a predominance of less lethal means (poisoning and use of sharp objects). An association between alcohol consumption and self-inflicted injuries was verified.

Conclusions: Based on the epidemiological study, it was observed that the city of Anápolis presented increasing statistics of notifications. It is expected that the municipal pregnancy in the city of Anápolis (GO), observing the epidemiology of the variables involved, can maintain the basic conditions for mental health care, through Primary Care and with the support of Psychosocial Care Centers (CAPS), outpatient clinics of mental health and reference hospitals for the care of patients requiring psychiatric hospitalizations.

#### KEYWORDS:SELF-HARM, ANÁPOLIS, DATASUS, SINAN.

#### INTRODUCTION

Suicidal ideation is characterized as a thought of self-destruction, encompassing planning to end one's life. Self-injury, on the other hand, is defined as a type of violence in which the person inflicts harm on themselves, including acts of self-mutilation, scratching, biting, cutting, amputation, which can lead to mild to severe consequences.<sup>1</sup> Suicide attempt is considered the behavior taken with the purpose of harming oneself, intending to commit suicide. If the act is completed, the suicide attempt is defined as suicide. The difference between suicidal ideation, suicidal behavior, self-injury, and completed suicide appears to be subtle.<sup>2</sup>

The factors that lead an individual to commit self-injury are diverse<sup>3</sup>, complex, and intimately related to each other. Among the variables that generate self-harm are genetics, environmental factors, psychiatric illnesses, use of alcohol and illicit drugs, loneliness, social motivations, family and social factors, as well as environmental, religious, and political elements.

In Brazil, reports of self-inflicted injuries are made

through the individual notification form (interpersonal/ self-inflicted violence), the information from which is incorporated into the Ministry of Health's (MS) database and subsequently tabulated in DATASUS, specifically in the Notification of Injury Information System (SINAN). This is a fundamental document for the development of public policies at all levels of health care for the prevention of suicide. However, studies2 have shown that only 25% of self-inflicted injuries are treated in hospital settings.

#### METHODS

This is a cross-sectional, retrospective study regarding notifications of self-inflicted injuries in the city of Anápolis (GO), based on data obtained through access to the Notification of Injury Information System (SINAN) on the DATASUS platform. These data originate from individual notification forms for interpersonal/self-inflicted violence issued in the municipality between 2018 and 2022, which are available for public consultation. To calculate notification statistics, access was made to the SINAN website, under the "notifiable diseases and injuries - from 2007

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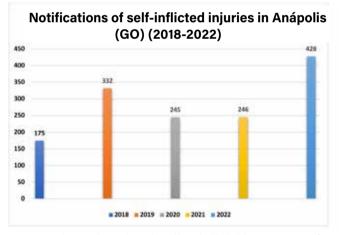
PEDRO HENRIQUE DOS SANTOS RODRIGUES Rua 23, quadra 44, lote 19 – Vale do Sol Anápolis (GO) – 75085700. E-mail: med.pedro6@gmail.com onwards" tab. Within DATASUS, the "year of notification" option was selected under "line", the "self-inflicted injuries" option was marked in the column, "frequency" was selected under content, the available periods were set to "2018-2022", and under "available selections", the municipality of "Anápolis" was selected, with age groups ranging from "10-14 to 60 years or more", with the option "self-inflicted injury". These filters were selected as basic for all searches.

Regarding the variable criteria in the searches on the SINAN platform, specific searches were conducted for each age group between 10-14 to 60 years, by male and female sex, skin color/race, education level, location of occurrence, marital status, with details on hanging, blunt object, sharp object, poisoning, and firearm, as well as the association with alcohol use.

The simple frequency data was tabulated in Excel and Tabwin, using a basic statistical methodology, with graphical representation in the form of tables and figures.

#### RESULTS

In terms of notification to the SINAN system, through the individual notification form on the DATASUS platform, it was demonstrated that over the years from 2018 to 2022, in the population aged 10-14 to 60 years or more, in both sexes, there was an increase of 59.12% in the reported cases (Figure 1).



Source: SINAN/VIVA/GVE/SUVISA/SES-GO – Data extracted on November 17, 2023.

## Figure 1. Absolute numbers of notifications of self-inflicted injuries in Anápolis (GO), between 2018 and 2022.

Regarding the notifications based on age groups, including ages equal to or above 10 years old, between the years 2018 and 2022, the following statistics were observed (Table 1), where the age group of 10 to 14 years old showed the highest increase in notifications (92.4%).

Age group						
10 a 14	15 a 19	20 a 29	30 a 39	40 a 49	50 a 59	60 ou mais
10	45	56	28	20	12	04
28	58	103	59	51	16	17
15	62	72	36	30	22	08
30	60	74	38	27	07	10
130	130	75	38	36	09	10
	10 28 15 30	10 45 28 58 15 62 30 60	10 a 14         15 a 19         20 a 29           10         45         56           28         58         103           15         62         72           30         60         74	10 a 14         15 a 19         20 a 29         30 a 39           10         45         56         28           28         58         103         59           15         62         72         36           30         60         74         38	10 a 14         15 a 19         20 a 29         30 a 39         40 a 49           10         45         56         28         20           28         58         103         59         51           15         62         72         36         30           30         60         74         38         27	10 a 14         15 a 19         20 a 29         30 a 39         40 a 49         50 a 59           10         45         56         28         20         12           28         58         103         59         51         16           15         62         72         36         30         22           30         60         74         38         27         07

Source: SINAN/VIVA/GVE/SUVISA/SES-GO – Data extracted on November 17, 2023.

#### Table 1. Prevalence of notifications by age groups between 2018 and 2022.

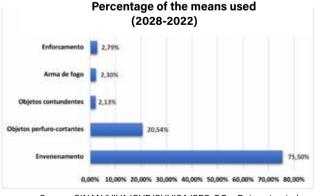
In the data regarding the level of education, the following statistics were noted (Table 2). The education levels that showed the highest increases in notifications during the observed period were between 5 and 8 incomplete elementary school grades, with an increase of 71.1%.

		Year				
Education	2018	2019	2020	2021	2022	
Illitirate	2	2	0	0	1	
1 to 4 incomplete elementary school						
grades	7	15	11	11	11	
Complete 4th grade	13	5	4	6	20	
5 to 8 incomplete elementary school						
grades	31	58	39	50	107	
Complete elementary school education	21	66	41	31	63	
Incomplete high school education	43	75	58	51	104	
Complete high school education	34	77	63	72	99	
Incomplete higher education	9	19	17	10	7	
Complete higher education	15	15	12	14	16	

Source: SINAN/VIVA/GVE/SUVISA/SES-GO – Data extracted on November 17, 2023.

# Table 2. Prevalence of notifications according to education levels between 2018 and 2022.

It is essential to understand the mechanisms through which self-injury occurred. An analysis of the objects and means by which self-injury was carried out was conducted (Figure 2). It was observed that the most commonly used objects/means were those considered less violent, such as poisoning (75.7%) and sharp objects (20.54%), while the more violent means, with greater force, accounted for only 2.3% (firearm) and 2.79% (hanging).



Source: SINAN/VIVA/GVE/SUVISA/SES-GO – Data extracted on November 17, 2023.

Figure 2. Prevalence of notifications according to the means used for selfinjury between 2018 and 2022. Regarding the locations where the injuries occurred, there was a predominance of the "Residence" location (Table 3), representing a percentage of 87.14% of the total notifications in 2022.

	Year					
Location of injury	2018	2019	2020	2021	2022	
Home	147	294	217	219	373	
Public road	11	13	13	12	19	
School	1	4	2	0	9	
Collective housing	1	2	2	3	2	
Commerce	3	1	2	0	6	
Industry	1	0	0	0	0	
Sports practice location	0	0	0	0	0	
Bars	2	2	1	4	2	
Others	4	3	4	4	10	
Unknown	5	13	4	4	5	

Source: SINAN/VIVA/GVE/SUVISA/SES-GO – Data extracted on November 17, 2023.

Table 3. Prevalence of notifications according to the locations of injury occurrence between 2018 and 2022.

Based on sex, the following distribution was observed between 2018 and 2022 (Table 4). The female sex predominated in the notifications, representing 69.4% of the sample, while the male population represented 30.6%.

Gender	Year						
	2018	2019	2020	2021	2022		
Male	50	105	76	82	122		
Female	125	227	169	164	306		

Source: SINAN/VIVA/GVE/SUVISA/SES-GO – Data extracted on November 17, 2023.

#### Table 4. Prevalence of notifications by gender between 2018 and 2022.

When correlating race/color, a predominance of the "brown" race was observed, with an increase of 61.8% from 2018 to 2022, representing 22.5% of the notifications in 2022 (Table 5).

Race	Year					
	2018	2019	2020	2021	2022	
White	37	85	50	56	78	
Black	8	5	5	5	10	
Brown	127	240	184	182	332	
Asian	2	1	3	1	4	
Indigenous	0	0	1	0	3	
Ignored	1	1	2	2	1	

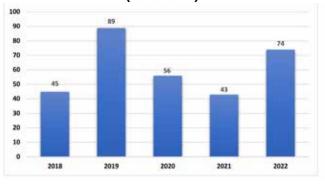
Source: SINAN/VIVA/GVE/SUVISA/SES-GO – Data extracted on November 17, 2023.

#### Table 5. Prevalence of notifications by race between 2018 and 2022.

An important factor to be correlated with the practice of self-injury was the verification of the association with alcohol consumption for the practice of self-inflicted injury.

It was observed that between the years 2018 and 2022, there was an increase of 39.2% in notifications of self-injuries associated with alcohol consumption (Figure 3).

Prevalence of association with alcohol use (2018-2022)



Source: SINAN/VIVA/GVE/SUVISA/SES-GO – Data extracted on November 17, 2023.

Figure 3. Prevalence of notifications by association with alcohol use between 2018 and 2022.

#### DISCUSSION

According to the latest demographic census from the Brazilian Institute of Geography and Statistics (IBGE) in 2022, the state of Goiás had an estimated population of 7,055,228 inhabitants. The city of Anápolis had a population of 398,869 people, with a population density of 426.29 inhabitants per square kilometer.

Anápolis (GO) is part of the health region called Pirineus, which includes a total of 10 municipalities. The city has 47 basic health units, divided between Basic Health Units (UBS) and Family Health Strategy (ESF). Regarding Psychosocial Care Centers (CAPS), the city has the CAPS Alcohol and Drugs Living (AD), CAPS Crescer (Child), and CAPS Vida Ativa, with support from the Mental Health Outpatient Clinic (Espaço Florescer). In terms of hospital care, Anápolis has services at the Emergency Care Unit (UPA 24H) of Vila Esperança, with support for hospitalization at the Eurípedes Barsanulfo Institute of Behavioral Medicine (INMCEB).

The preferential care is provided through the entry points of the Unified Health System (SUS), represented by Primary Care and CAPS. The first contact with the primary care level is essential to address a large part of the population's mental health demand, as mild, moderate, and severe (non-life-threatening) cases of mental disorders can be managed by trained teams on an outpatient basis. More severe cases, involving self-injury, suicidal ideation, planning, and suicide attempts, should be referred to other levels of care (secondary and tertiary). Primary care aims to provide prevention and promotion of mental health.<sup>4</sup>

Analyzing the results presented in the epidemiological review of self-inflicted injury notifications in the municipality of Anápolis (GO), the scenario of an increase in the number of cases follows the same pattern as the rest of the state of Goiás<sup>5</sup>, as well as the national and international scenarios. Regarding the age groups studied, the ages that showed the highest increase in notifications were the 10 to 14 age group, with an increase of 92.4% compared to the 2018 data. Similar data were found in a 2014 World Health Organization<sup>6</sup> study titled "Preventing suicide: A global imperative," where suicide was the second leading cause of death in the 15 to 29 age group.

Regarding gender, the female population in the city of Anápolis (GO) was the most affected by self-inflicted injuries, with a predominance of 69.4% compared to males. National statistics corroborate these findings, as a national study from 2016<sup>7</sup> observed that the prevalence of females in suicide attempts is an important health indicator for the development of public intervention policies.

Among the analysis of the race/color variable, the study observed that the most affected race by self-inflicted injuries was brown, with an increase of 61.8% compared to 2018. A study conducted by the Ministry of Health in 2014, published in 2015, found that race/ethnicity is a factor influencing the suicide rate in the population<sup>2</sup>. The factor of education level was also verified in the same study, with lower levels of education being correlated with a higher risk of suicidal behavior. In the present study, the main affected education levels were those between 5 and 8 incomplete elementary school grades, with an increase in incidence of 71.1%.

Given the notifications of self-inflicted injuries, it is imperative to observe how the act of self-harm occurred.<sup>8</sup> In a national study from 2018.<sup>9</sup> it was observed that the act of suicide can be carried out by less or more lethal/violent means. The most lethal means, such as firearm use and hanging, are preferred by men, as they tend to attempt suicide in a definitive manner, while women opt for less lethal/violent means, such as poisoning and the use of sharp objects. In the present study, this statistic correlated with the study9, where self-harm occurred by poisoning in 75.5% of cases, and in 20.54% by the use of sharp objects.

It is important to consider that suicide acts or attempts, as well as suicidal ideations and planning, are closely related to mental disorders, such as depression, anxiety disorders, bipolar disorder, and the use of alcohol and psychoactive drugs.<sup>10,11</sup> In the present study, the association between alcohol and notifications of self-inflicted injuries increased by 39.2% in 2022 compared to 2018. Alcohol is a depressant of the central nervous system, and its association is harmful in the context of mental disorders.<sup>12</sup>

#### CONCLUSION

The present epidemiological study of the scenario of compulsory notifications of self-inflicted injuries in the municipality of Anápolis (GO), in the time frame from 2018 to 2022, showed that the city of Anápolis (GO), with its increasing population growth and expanding development, has increasing statistics of self-harm notifications, following the state, national, and global scenarios. Individuals who commit self-inflicted injuries are in a progressive process of severe mental crisis, and early intervention is potentially able to interrupt the sequence of events that can lead to completed suicide.<sup>2</sup> The act of welcoming is essential for the user to be treated in a humane, welcoming, and effective manner, focusing on a horizontal approach, with a focus on health promotion. Nowadays, in the 21st century, self-inflicted injuries are seen as a taboo.<sup>13</sup> Such individuals are stigmatized not only by society but also by health professionals who provide initial care, as patients are seen as suffering from some mental disorder, not deserving of the proper seriousness at the time of care.<sup>14</sup>

Based on the epidemiological variables presented in the study, it is expected that the municipal management of the city of Anápolis (GO), especially through the Municipal Health Department (SEMUSA), can allocate financial and human resources to meet the basic needs for mental health care, focusing on prevention and promotion of mental health through Primary Care, with support from Psychosocial Care Centers (CAPS), increasing the number of available slots for mental health outpatient consultations (Espaço Florescer), strengthening reception mechanisms and professional training in hospitals for receiving patients who self-harm, and increasing the number of slots through the National Regulation System (SISREG) in reference hospitals for psychiatric hospitalizations in the city of Anápolis (GO).

- WHO. World Report on Violence and Health. New South Wales Public Health Bulletin. 2002;13(8):190. ok
- BRASIL. Ministério da Saúde. Secretaria de Vigilância em Saúde. Saúde Brasil 2014: uma análise da situação de saúde e das causas externas. Brasília: Ministério da Saúde, 2015.
- Bahia CA, Avanci JQ, Pinto LW, Minayo MC de S. Lesão autoprovocada em todos os ciclos da vida: perfil das vítimas em serviços de urgência e emergência de capitais do Brasil. Ciência & Saúde Coletiva. 2017 Sep;22(9):2841–50.
- Magalhães LS, Oliveira Andrade SM. Depressão e Comportamento Suicida: Atenção Primária em Saúde. Revista Psicologia e Saúde. 2019 Feb 8;11(1):107.
- Rodrigues M de F, Oliveira PP de, Silva HC da, Pinheiro JM da C. Comportamento suicida: o perfil epidemiológico das lesões autoprovocadas no Estado de Goiás. Revista Científica da Escola Estadual de Saúde Pública de Goiás "Cândido Santiago." 2020 Aug 12.
- World Health Organization. Preventing suicide: a global imperative. Whoint [Internet]. 2014;(1). Available from: https://apps.who.int/iris/handle/10665/131056.
- VASCONCELOS-RAPOSO J, SOARES AR, SILVA F, FERNANDES MG, TEIXEIRA CM. Níveis de ideação suicida em jovens adultos. Estud psicol (Campinas) [Internet]. 2016Apr;33(2):345–54. Available from: https://doi. org/10.1590/1982-02752016000200016.
- AÑUÁRIO BRASILEIRO DE SEGURANÇA PÚBLICA 2022. São Paulo: Fórum Brasileiro de Segurança Pública, ano 16, 2022. ISSN 1983-7364.
- Ribeiro JM, Moreira MR. Uma abordagem sobre o suicídio de adolescentes e jovens no Brasil. Ciência & Saúde Coletiva [Internet]. 2018 Sep 1 [cited 2022 May 23];23:2821–34. Available from: https://www.scielo.br/j/csc/a/ txZCWtk98yqSkvTTj6Vj74b/?format=html&lang=pt.
- Meleiro AMA da S. Prática psiquiátrica no hospital geral: interconsulta e emergência. Revista Brasileira de Psiquiatria. 2002 Jun;24(2):105–5.
- Cavalcante FG, Minayo MC de S. Estudo qualitativo sobre tentativas e ideações suicidas com 60 pessoas idosas brasileiras. Ciência & Saúde Coletiva. 2015 Jun;20(6):1655–66.

- Cordeiro EL, Silva LSR da, Mendes EWP, Silva LCL da, Duarte VL, Lima ÉCMP. Tentativa de suicídio e fatores associados ao padrão uso e abuso do álcool. SMAD Revista Eletrônica Saúde Mental Álcool e Drogas (Edição em Português). 2020 Feb 21;16(1):1–10.
- Eilers JJ, Kasten E. Finished with Life Anyway and Then Stigmatized for Attempting Suicide—An Overview. Healthcare. 2022 Nov 17;10(11):2303.
- Taylor TL, Hawton K, Fortune S, Kapur N. Attitudes towards clinical services among people who self-harm: systematic review. British Journal of Psychiatry [Internet]. 2009 Feb;194(2):104–10. Available from: https://www. cambridge.org/core/journals/the-british-journal-of-psychiatry/article/attitudes-towards-clinical-services-among-people-whoselfharm-systematic-review/99BAD49B1EA6FB9A0C9AE6450F7629ED.

# SPONTANEOUS INTRATHECAL HYPOTENSION: CASE REPORT

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#### ABSTRACT

Introduction: Intracranial hypotension is a condition characterized by a lower than normal volume of cerebrospinal fluid (CSF) due to CSF leakage across the dural membrane in one or multiple locations. Symptoms include orthostatic headache and symptoms associated with stiff neck, tinnitus, photophobia, nausea, and evidence of low CSF pressure or leakage through at least one imaging modality. Case report: male patient, 46 years old, physician, with report of spontaneous CSF leak during physical activity. It evolved with the following symptoms: pulsatile holocranial headache that worsens when lying down and getting up. He underwent oral treatment without success, requiring an invasive procedure such as a blood patch to improve symptoms. After a few days he underwent new imaging tests with no changes. Discussion: The most common site of CSF leak is the cervicothoracic junction or upper thoracic region. Epidural blood patches are frequently performed for the treatment of post-dural puncture headache, which may vary between the previous site of cerebrospinal fluid leak or the lumbar location.

#### KEYWORDS: CEREBROSPINAL FLUID; INTRACRANIAL HYPOTENSION; EPIDURAL BLOOD PLAQUE.

#### INTRODUCTION

Intracranial hypotension is a condition characterized by a lower-than-normal volume of cerebrospinal fluid (CSF) due to CSF leakage through the dural membrane, at one or multiple locations. The loss of CSF results in the displacement of brain structures, causing headaches and other neurological symptoms<sup>1</sup>. Although the headache may present benign signs, if left untreated, it can lead to serious adverse events such as stroke and neurological sequelae<sup>2,3</sup>.

Headache with low CSF pressure is almost always caused by CSF leakage, which can be iatrogenic, traumatic, or spontaneous in origin. The etiology of spontaneous CSF leakage is still unclear, and the site of the leak can be anywhere along the spinal column, but is usually at the level of the thoracic or low cervical region<sup>4</sup>. The most common presentation is orthostatic headache. Other features may include cranial nerve palsies, dizziness, tinnitus, photophobia, changes in hearing, or neck stiffness<sup>5</sup>.

Data from large community-based studies on the epidemiology of spontaneous intracranial hypotension are not available, but an estimate of the annual incidence is 4 to 5 cases per 100,000 people<sup>6</sup>, which is about half the incidence of aneurysmal subarachnoid hemorrhage. Including children and adolescents, spontaneous intracranial hypotension can affect patients of any age, particularly women aged 35 to 55 years<sup>14</sup>.

The present study aims to present a case of spontaneous cerebrospinal fluid (CSF) fistula. Based on this case report, we seek to explore methods of diagnosis and treatment for this manifestation.

#### CASE REPORT

Male patient, 46 years old, residing in the city of Goiânia/Goiás, physician, with no underlying pathologies, regularly practicing physical exercises (running and swimming), and no allergies. He reports that during a few days of vacation in December 2021, he started experiencing back pain in the cervical region after more strenuous physical exercise than usual at the beach, associated with pulsatile holocranial headache that worsened with lying down/standing up, with a pain score of 3/10. He used regular painkillers with partial relief of the pain. After 4 days, already in Goiânia, he returned to work and started experiencing diplopia, blurred vision, and decreased visual acuity. He underwent cervical spine magnetic resonance imaging (MRI) with a diagnosis of pachymeningitis, represented by Figure 01 below, leading to further investigation, during which he underwent lumbar puncture and viral panel, all of which were normal.

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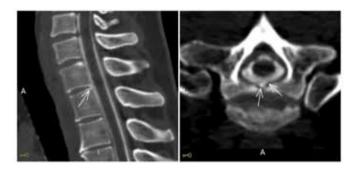


Figure 01: Computed tomography myelography of the cervical and thoracic spine showing epidural extravasation of contrast medium diffusely into the epidural space in the later acquisitions, from C5 to T5.

He underwent another MRI of the neuroaxis with contrast, which reported the presence of a collection in the C7-T1 region, leading to a diagnosis of cerebrospinal fluid hypotension. He rested at home for a week, with no improvement in mild pain symptoms. During this time, he was taking oral prednisolone for 5 days due to suspicion of viral meningitis. After the rest period, he returned to the hospital where an anesthesiologist performed a blood patch in the L3-L4 lumbar region with 20 ml, remaining at rest for another week without improvement. He returned to work with persistent symptoms, leading to another blood patch in the L2-L3 lumbar region with 20 ml. He remained at rest at home for another week, with no improvement.

He underwent a third blood patch with a higher location using 15 ml, without improvement. He decided to seek other means to continue the investigation, where he underwent a myelotomography with contrast, with a report of epidural extravasation of the contrast medium from T1-T2, spreading throughout the epidural space in the later acquisitions, from C5 to T5.

He returned to Goiânia, where a fourth blood patch was performed in the C7-T1 region guided by tomography with 10 ml, and after 15 days, the patient denied any symptoms. After five months, another MRI was performed, which was reported as normal.

#### DISCUSSION

The diagnostic criteria for spontaneous intracranial hypotension, as established by the International Classification of Headache Disorders, require the presence of (1) orthostatic headache, (2) associated symptoms of >=1 (neck stiffness, tinnitus, photophobia, nausea), and (3) evidence of low CSF pressure or leakage through at least one modality (brain MRI detecting indirect signs of low CSF pressure, lumbar puncture measuring CSF pressure less than 6 cm-H2O, CT myelography, or radionuclide cisternography RC)<sup>7</sup>.

The most common site of CSF leakage is the cervicothoracic junction or upper thoracic region. This usually occurs spontaneously or after minor trauma to the spinal cord. As a result of CSF leakage, the brain sinks, causing traction on the bridging veins, pain-sensitive meningeal structures, and cranial nerves, leading to symptoms<sup>8</sup>.

Epidural blood patches are commonly used for headaches attributed to CSF leakage. They are used to treat patients who experience headaches after a known dural injury. CSF leakage was first reported in 1955<sup>9</sup>. The etiology of this leakage is variable, with the most common cause being lumbar puncture. In a recent Cochrane Review, the blood patch was considered superior to conservative management for the treatment of post-dural puncture headache10. Most blood patches, however, are performed at the lumbar level. Cervical blood patching is feared for several reasons. Commonly cited complications include cranial nerve palsy<sup>11</sup>, altered mental status<sup>12,13</sup>, subdural hematoma<sup>14</sup>, seizures1<sup>5</sup>, and transient bradycardia<sup>16</sup>. Serious adverse events also include compression of nerve roots17 and chemical meningitis18. At the cervical level, spinal cord compression becomes the most feared complication. However, no systematic study has been done to elucidate the incidence of these complications.

There are several reports indicating that the lumbar blood patch can permanently relieve headache, regardless of whether the site of leakage is identified or not<sup>19,20,21</sup>. However, other reports demonstrate that the lumbar blood patch does not always result in permanent relief<sup>23,23,24</sup>. A study by Diaz<sup>25</sup> suggests that the site of leakage should be identified by radionuclide cisternography and treated with a blood patch targeted at the levels of the CSF leak. A report by Kantor and Silberstein<sup>26</sup> also suggests that cervical blood patch may be useful after failure of lumbar blood patch when the site of leakage is not identified.

Our case report exemplifies two unsuccessful lumbar epidural blood patch (EBP) attempts for spontaneous CSF leakage. Subsequently, when the site of the leakage was identified, a blood patch was performed at the C7-T1 level, resulting in symptom improvement.

A case report by Ferrante et al. indicated that the blood patch successfully treated spontaneous CSF leakage in the cervical region. This report also showed the spread of blood from the lumbar to the cervical region. Cousins et al.<sup>28</sup> suggested that placing the blood patch close to the site of CSF leakage is important. The proposed mechanism is that the injected blood seals the dural defect and stops the leakage. Another theory is that the blood injection causes an epidural tamponade effect on the leakage. It would seem sensible to target the treatment to the site of leakage to maximize the chances of success, but there is no clear evidence to support targeted blood patching, and randomized clinical trials are likely not feasible given the low incidence of the disease.

In anesthesia practice, a blood patch at the site of leakage is the treatment of choice if conservative measures fail. Other treatments have been suggested with varying success rates, including intrathecal fluid infusion, epidural saline infusion, epidural dextran infusion, epidural fibrin glue injection, CSF diversion, and surgical repair of the leak<sup>29</sup>. The procedure then becomes more technically challenging and higher risk.

#### REFERENCES

- tigation findings and treatment outcomes of spontaneous intracranial hypotension syndrome: a systematic review and metaanalysis. JAMA Neurol 2021;78:329-37. DOI:10.1001/jamaneurol.2020.4799
- Schievink W. Stroke and death due to spontaneous intracranial hypotension. Neurocrit Care. 2013;18:248–251. DOI: 10.1007/s12028-012-9800-3
- Zakaria R, Wilby M, Fletcher N. Spontaneous csf collection in the cervical spine may cause neurological deficit and intracranial. DOI: 10.2174/1874205X01307010001
- 4.Schievink WI, Michael LM II, Maya M, Klimo P Jr, Elijovich L. Spontaneous intracranial hypotension due to skull-base CSF leak. Ann Neurol 2021;90:514-6. DOI: 10.1002/ana.26175
- Panya N, Suyin GT. High thoracic/Cervical epidural blood patch for spontaneous cerebrospinal fluid leak: a new challenge for anesthesiologists. Anesth Analg 2011;113:1476 –9. DOI: 10.1213/ANE.0b013e31822e5665
- Schievink WI, Maya M, Moser FG, Simon PA, Nuño M. Incidence of spontaneous intracranial hypotension in a community. Beverly Hills, California, 2006- 2020. Cephalalgia 2021 September 23. https://doi. org/10.1177/0333102421104
- 7.Headache Classification Subcommittee of the International Headache Society. The International Classification of Headache Disorders: 2nd Ed. Cephalalgia. 2004;24(S1):9–160.
- 8.Sriganesh K, JS, VB. Targeted cervical epidural blood patch for the management of refractory headache in a patient with spontaneous intracranial hypotension. J Neurosurg Anesthesiol. 2015 Jan;27(1):68-9. DOI: 10.1097/ANA.000000000000059
- Nosik W. Intracranial hypotension secondary to lumbar nerve sleeve tear. J Am Med Assoc 1955;157: 1110–11.
- Boonmak P, Boonmak S. Epidural blood patching for preventing and treating post-dural puncture headache. Cochrane Database Syst Rev 2010; 1: CD001791.
- Perez M, Olmos M, Garrido FJ. Facial nerve paralysis after epidural blood patch. Reg Anesth 1993;18: 196–8.
- Beers RA, Cambareri JJ, Rodziewicz GS. Acute deterioration of mental status following epidural blood patch. Anesth Analg 1993;76:1147–9.
- Sperry RJ, Gartrell A, Johnson JO. Epidural blood patch can cause acute neurologic deterioration. Anesthesiology 1995;82:303–5.
- Tekkok IH, Carter DA, Brinker R. Spinal subdural haematoma as a complication of immediate epidural blood patch. Can J Anaesth 1996;43:306–9.
- Kardash K, Morrow F, Beique F. Seizures after epidural blood patch with undiagnosed subdural hematoma. Reg Anesth Pain Med 2002;27:433–6.
- Andrews PJD, Anaes FRC, Ackerman WE, et al. Transient bradycardia associated with extradural blood patch after inadvertent dural puncture in parturients. Br J Anaesth 1992;69:401–3.
- Waguri N, Tomita M, Hayatsu K, Okamoto K, Shimoji K. Epidural blood patch for treatment of spontaneous intracranial hypotension. Acta Anaesthesiol Scand 2002;46:747–50
- Oh J, Camann W. Severe, acute meningeal irritative reaction after epidural blood patch. Anesth Analg 1998;87:1139–40
- Ferrante E, Arpino I, Citterio A. Is it a rational choice to treat with lumbar epidural bloodpatch headache caused by spontaneous cervical CSF leak? Cephalalgia 2006;26:1245– 6
- Terjima K, Oi Y, Ogura A, Sakai N, Takei M, Tamaki T, Ogawa R. Blood patch therapy for spontaneous intracranial hypotension: safe performance after epidurography in an unconscious patient. Anesth Anal 2002;94:959 – 61
- Benzon HT, Nemickas R, Molloy RE, Ahmad S, Melen O, Cohen B. Lumbar and thoracic epidural blood injections to treat spontaneous intracranial hypotension. Anesthesiology 1996;85:920 –2
- Berroir S, Loisel B, Ducros A, Boukobza M, Tzourio C, Valade D, Bousser M-G. Early epidural blood patch in spontaneous intracranial hypotension. Neurology 2004;63:1950 –1
- Hannerz J, Dahlgren G, Irestedt L, Meyerson B, Ericson K. Treatment of idiopathic intracranial hypotension: cervicothoracic and lumbar blood patch and peroral steroid treatment. Headache 2006;46:508 –11
- 24. Nowak DA, Takano B, Topka H. Spontaneous cerebrospinal fluid hypovolaemia: a therapeutic dilemma? Eur J Neurol 2006;13:1128 –38
- 25. Diaz JH. Epidemiology and outcome of postural headache management in

spontaneous intracranial hypotension. Reg Anesth Pain Med 2001;26:582–7 26. Kantor D, Silberstein SD. Cervical epidural blood patch for low CSF pressure headaches. Neurology 2005;65:1138

- Ferrante E, Arpino I, Citterio A. Is it a rational choice to treat with lumbar epidural bloodpatch headache caused by spontaneous cervical CSF leak? Cephalalgia 2006;26:1245– 6
- Cousins MJ, Brazier D, Cook R. Intracranial hypotension caused by cervical cerebrospinal fluid leak: treatment with epidural blood patch. Anesth Anal 2004;98:1794 –7
- Mokri B. Headaches caused by decreased intracranial pressure: diagnosis and management. Curr Opin Neurol 2003;16:319 –26

# ECTOPIC PREGNANCY WITH OVOCYTE TRANSMIGRATION: A CLINICAL CASE REPORT

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#### ABSTRACT

The purpose of this article is to report a clinical case involving the occurrence of ectopic pregnancy (EP) and oocyte transmigration. A 33-year-old woman, native of Mozarlândia, Goiás, presented with polymenorrhea and dysmenorrhea after a positive pregnancy test. With no significant history of illnesses, the patient reported social alcohol consumption and previous use of oral contraceptives for 5 years. After the positive pregnancy test acquired at a pharmacy, she experienced vaginal bleeding and abdominal pain, leading her to an emergency consultation where the -hCG test confirmed the pregnancy and a Transvaginal Ultrasound (TVUS) showed normal findings. With persistent symptoms, a second -hCG test and subsequent TVUS revealed insufficient increase in hormonal levels and an adnexal mass with hemoperitoneum. A videolaparoscopy was performed, identifying an EP in the left fallopian tube and oocyte transmigration, treated with salpingostomy and ovarian cystectomy. The patient recovered well after surgery, without complications. This case highlights the importance of early diagnosis and timely surgical intervention in cases of EP, aiming to avoid severe complications and preserve the patient's fertility. The diagnosis and treatment of EP are challenging due to the ambiguity of symptoms. Technological advances have improved diagnosis, but half of the cases are not initially identified. Treatment can be medical or surgical, depending on the severity and reproductive desires of the patient.

#### KEYWORDS: ECTOPIC PREGNANCY; PREGNANCY TESTS; TUBAL PREGNANCY; ULTRASONOGRAPHY; ZYGOTE.

#### INTRODUCTION

The term ectopic pregnancy (EP) refers to the situation where fertilization and implantation of the eggs occur in a location other than the uterine cavity, making this condition one of the main emergencies in Gynecology and Obstetrics<sup>1</sup>. The rupture of the structures where EP occurs is the most common cause of maternal death in the first trimester of pregnancy and occurs because there is a structural incompatibility of the tissue with the gestational sac<sup>2</sup>. The precise rates related to the estimate of EP occurrence are not clear due to underreporting of cases that are often treated on an outpatient basis; however, it is estimated that in the United States of America, between 1 to 2% of pregnancies occur in this way<sup>3</sup>.

Among the possibilities, the uterine tube, better known as the fallopian tube, is the most recurrent site of EP, accounting for 90% of cases<sup>4</sup>. This event can be fatal since it can be a causal factor for severe internal bleeding, leading to the death of up to 6% of pregnant women<sup>2</sup>. The risk factors for this condition are diverse and can be related to the mother's lifestyle habits, such as smoking, and even genetic factors<sup>5</sup>.

The mechanisms by which the oocyte is captured by the fimbriae, structures present in the fallopian tubes, are still not clear in the world literature; however, there is evidence that, due to the anatomical structure and positioning of the uterus, tubes, and ovaries, oocyte capture occurs by the ipsilateral tube<sup>1</sup>. Despite this, it is known that there is an event, called oocyte transmigration, in which pregnancy occurs contralateral to the ovary responsible for ovulation, and the elucidation of the physiological mechanism for this explanation remains limited<sup>6</sup>. Thus, the objective of this article is to describe a case report in which there were two events, namely ectopic pregnancy and oocyte transmigration.

#### **CASE REPORT**

Patient, L. M. C., 33 years old, Caucasian, from Mozarlândia - Goiás (GO), sought gynecological medical care at a private clinic in the city of Goiânia - GO complaining of polymenorrhea and dysmenorrhea after being diagnosed with pregnancy through a pharmacy-sold test. Regarding her medical history, the patient denies a history of prior pregnancies, does not report chronic diseases, genetic conditions, blood pressure alterations, or smoking habits. She reports social alcohol consumption. She reports continuous use of Ofolato D 10,000 IU and Reconter 10mg (Escitalopram Oxalate), in addition to the oral contraceptive Stezza Merk 2.5mg/1.5mg (Nomegestrol Acetate/Estradiol) for the past 5 years which she stopped using 4 months before with the intention of getting pregnant.

From the detailed history, the patient reports that she

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started her menstrual cycle on December 6, 2023, which was expected, and that the bleeding continued until December 9, 2023, with a heavier flow than usual. The patient reports that until then, her cycle was normal with a medium flow lasting 3-4 days. On December 13, 2023, there was another episode of bloody discharge associated with mild pain, cramping, in the lower abdominal region, predominantly in the hypogastric region. Due to this, a pharmacy-sold pregnancy test was performed, which confirmed the positive result.

Therefore, on the following day, December 14, 2023, the patient sought emergency care in Goiânia - GO, where the Beta-human Chorionic Gonadotropin (β-hCG) test was performed, which was reactive, confirming the positive result with a value of 141.7 mIU/ml. Additionally, a Transvaginal Ultrasound (TVUS) was requested to investigate the origin of uterine bleeding, and the examination did not detect any apparent abnormalities. The exam showed the uterus in anteversion, centered, with normal shape and size, with well-defined and precise contours, myometrium with homogeneous acoustic texture and symmetrical walls, closed endocervical canal, and regular junctional zone. The endometrial echo was present, well-defined, regular, trilaminar, with a thickness of 6.9 mm. The ovaries were normal in size and echogenic pattern, with the left ovary measuring 2.56 x 1.75 x 1.89 cm and a volume of 4.40 cm<sup>3</sup>, and the right ovary measuring 2.67 x 1.98 x 2.0 cm and a volume of 5.58 cm<sup>3</sup>. (Figure 1). Consequently, the patient returned to work activities without restriction in her routine, maintaining exercises with effort, however, still experiencing constant, uncomfortable but non-limiting pain, which did not require analgesic medication.

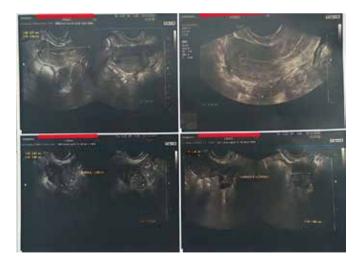


Figure 1 - TVUS performed on December 14, 2023, showing normal uterus and adnexal

On December 18, 2023, another  $\beta$ -hCG test was performed, showing a quantitative result lower than expected, with a value of 291.7 mIU/ml. Additionally, the picture of polymenorrhea remained constant. Therefore, considering these data, the patient was advised to seek medical care in Goiânia - GO, at a private clinic, in order to undergo TVUS for detailed gynecological analysis. Furthermore, she was recommended to take one tablet of Utrogestan 200 mg. The TVUS was performed on December 24, 2023, showing an adnexal mass of 4.0 cm on the right side and hemoperitoneum (Figure 2). Given these findings, laparoscopy was performed to investigate the findings of the exam.



Figure 2 - TVUS performed on December 24, 2023, showing a right adnexal mass and hemoperitoneum.

Therefore, the patient was immediately admitted to the hospital for treatment initiation. She was promptly referred to the operating room, where she underwent laparoscopy. During the procedure, a gestational sac was identified in the left fallopian tube, containing embryonic material inside. Salpingostomy and cystectomy of the right ovary were performed, followed by cleaning and aspiration of the hemorrhagic content in the abdominal cavity due to the ruptured ectopic pregnancy (Figure 3). Both fallopian tubes and ovaries were preserved. There were no surgical complications. The patient's progress was satisfactory, and she was discharged the day after the surgery.

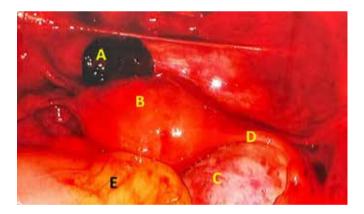


Figure 3 - Image obtained by laparoscopy showing A. Gestational sac after removal from the left tube; B. Uterus; C. Right ovary containing an adnexal cystic mass; D. Right fallopian tube; E. Rectum

#### DISCUSSION

This study presents a case report aiming to describe the patient's diagnostic process. Ectopic pregnancy (EP) is an important factor in maternal morbidity due to its challenging diagnosis<sup>5</sup>. Additionally, factors such as smoking, pelvic inflammatory diseases, and increased use of assisted reproduction techniques may be crucial in increasing the incidence of EP. However, these data are still hypotheses, as there is still no exact relationship regarding identifiable risk factors for EP. It is more clearly associated with factors that cause damage to the fallopian tubes<sup>5</sup>.

In the literature, several authors seek to identify more causal factors of EP, with a positive association found between age (>35 years), infertility, contraceptive failure, previous EP, and previous spontaneous abortion<sup>15,6</sup>. It is worth noting that in the present clinical case, there is no association with the aforementioned pathophysiological factors. Another similar case to the one described in this clinical case is reported in the literature, as described by Sikorski; Zrubek (2003). Therefore, the importance of early diagnosis and the need for more studies to understand the pathophysiological aspects of EP are emphasized. The work of Sikorski; Zrubek (2003) includes an important observation regarding the transperitoneal migration of spermatozoa, ova, or zygotes, found through the visualization of the corpus luteum in the ovary contralateral to the tubal implant<sup>8</sup>.

The clinical presentation of EP is ambiguous, with signs and symptoms commonly compatible with the onset of pregnancy, such as pain and vaginal bleeding, as reported in the current case, and there may even be an absence of signs and symptoms. Bleeding in EP is due to the decidual shedding of the endometrial layer and can vary in intensity, ranging from light spotting to heavy bleeding. As for pain, the variation is even greater depending on the patient, which can be localized on only one side or generalized <sup>15</sup>.

The diagnosis has substantially improved due to technological advances in ultrasound and hormonal testing. However, it still presents a challenge for gynecology, as approximately 50% of women do not receive the diagnosis at the first presentation, as described in the current case under evaluation<sup>5</sup>. Quantitative values with significance in  $\beta$ -hCG can already be identified from the 8th day of gestation, and monitoring its level is an important tool to assist in the diagnosis of EP<sup>1</sup>. The rate is measured every 48 hours to distinguish a normal from an abnormal pregnancy, and when there is a variation greater or lesser than expected for the stage of gestation, EP or early pregnancy loss is suspected<sup>1</sup>. Other methods such as serum progesterone levels, biomarkers, and transvaginal ultrasound can also be used<sup>1</sup>.

Regarding treatment, one should opt primarily for less invasive routes when possible, such as the administration of methotrexate, a medication that targets cells with high mitogenic activity. On the other hand, in more severe cases and even in advanced EP, surgical intervention is necessary, through salpingostomy or salpingectomy <sup>51</sup>. Regarding surgical technique, in general, an endoscopic approach is consensually preferable, as randomized studies confirm the advantage of a minimally invasive approach. Among the main advantages are: reduced operative time, lower risk of postoperative adhesions, shorter hospital stay, and a shorter and accelerated return to daily life activities, as well as work activities<sup>10</sup>.

Regarding the safety of the entry method, open or closed, studies show that there is no significant difference between the two<sup>10</sup>. The technique of salpingectomy consists of removing the fallopian tube; if it has not been planned before the start of the surgery, it is recommended to assess the viability of the contralateral fallopian tube<sup>10</sup>. During removal, priority is given to removing the EP in situ to avoid the risk of rupture in the abdominal cavity, with the risk of subsequent EP10. The salpingostomy technique consists of resecting the gestational sac while preserving the fallopian tube. The incision should be minimal, allowing the ectopic to be easily expelled or removed (usually between 1 and 2 cm). If this does not occur spontaneously, hydrodissection techniques can be used to facilitate removal; however, the surgeon must be careful not to rupture the ectopic<sup>10</sup>.

Salpingectomy is indicated for patients with EP larger than 5 cm in diameter, significant tubal damage, hemorrhage, or previous tubal ligation. However, for patients who wish to preserve fertility, salpingostomy is recommended to preserve the fallopian tubbe <sup>910</sup>. Regardless of the technique used, after the procedure, abdominal washing is recommended to aspirate any trophoblastic tissue accidentally spilled during surgery, given the risk of a new EP<sup>10</sup>. In general, surgical intervention presents more satisfactory results when compared to clinical treatment. On the other hand, both surgical and clinical management did not show a significant difference regarding fertility rate. Therefore, it is recommended that the physician consider other factors for decision-making, such as hemorrhage, patient stability, and fertility <sup>56,910</sup>.

#### CONCLUSION

Diagnosis and treatment of ectopic pregnancy (EP) represent significant challenges in clinical practice, given the ambiguity of symptoms and the need for early intervention to avoid serious complications. While technological advances have improved diagnosis, about half of cases are still not initially identified. Treatment, preferably less invasive, may include medical methods such as methotrexate or surgical intervention such as salpingostomy or salpingectomy, with the choice depending on the severity of the case and the patient's reproductive desires. Although there is no significant difference in fertility after surgical or clinical treatment, other factors such as patient stability should be considered in the therapeutic decision. Further studies are needed to fully understand the risk factors and optimal approaches for the diagnosis and treatment of EP.

- Hendriks E, Rosenberg R, Prine L. Ectopic pregnancy: diagnosis and management. Am Fam Physician. 2020 May 15;101(10):599-606.
- 2. Creanga AA, Syverson C, Seed K, Callaghan WM. Pregnancy-related mortal-

ity in the United States, 2011-2013. Obstet Gynecol. 2017 Aug;130(2):366-73.

- Creanga AA, Shapiro-Mendoza CK, Bish CL, Zane S, Berg CJ, Callaghan WM. Trends in ectopic pregnancy mortality in the United States: 1980-2007. Obstet Gynecol. 2011 Apr;117(4):837-43.
- Bouyer J, Coste J, Fernandez H, Pouly JL, Job-Spira N. Sites of ectopic pregnancy: a 10 year population-based study of 1800 cases. Hum Reprod. 2002 Dec;17(12): 3224-30.5. Sivalingam VN, Duncan WC, Kirk E, Shephard LA, Horne AW. Diagnosis and management of ectopic pregnancy. J Fam Plann Reprod Health Care. 2011 Oct;37(4):231-40.
- Benjamin I, Figueira JV, Miquilarena R, Rodriguez F, Lopez A, Lerner J. Ectopic pregnancy with a contralateral corpus luteum: case report. JBRA Assist Reprod. 2023 Jun 22;27(2):314-6.
- Sikorski M, Zrubek H. Sperm, oocyte or zygote transmigration as a cause of adnexal stump heterotopic pregnancy. J Obstet Gynaecol. 2003 Sep;23(5):577-8.
- Insunza A, de Pablo F, Croxatto HD, Letelier LM, Morante M, Croxatto HB. On the rate of tubal pregnancy contralateral to the corpus luteum. Acta Obstet Gynecol Scand. 1988;67(5)433-6.
- Mullany K, Minneci M, Monjazeb R, Coiado OC. Overview of ectopic pregnancy diagnosis, management, and innovation. Womens Health (Lond). 2023;19: 17455057231160349.
- Ozcan MCH, Wilson JR, Frishman GN. A systematic review and meta-analysis of surgical treatment of ectopic pregnancy with salpingectomy versus salpingostomy. J Minim Invasive Gynecol, 2021 Mar;28(3):656-67.

# OSTEOGENESIS IMPERFECTA IN A NORMAL PREGNANCY : A CLINICAL CASE REPORT

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#### ABSTRACT

The aim of this work is to report a clinical case of childbirth in a patient with Osteogenesis Imperfecta (OI) during an unplanned pregnancy. Pregnancy in these patients is associated with maternal and fetal complications, given that the presence of an autosomal dominant mutation in the mother determines a 50% risk of OI in the offspring. This is a case report of a pregnant patient with Osteogenesis Imperfecta, with a positive maternal and fetal outcome, aiming to discuss possible complications arising during pregnancy due to this comorbidity, focusing on recommendations for managing these women. Patient C.C.V., a 25-year-old female, diagnosed with OI, primiparous, with a history of multiple bone fractures, bone dysplasia, blue sclera, and endometrioma. She was admitted in March, 2023, for a cesarean section, hemodynamically stable with unaltered laboratory tests and negative serologies, with a family history of diabetes, heart disease, and glaucoma. The patient reported an unexpected pregnancy without prenatal care, and the fetus showed no suspicion of pathology. The delivery was via cesarean section, after the onset of labor, with spinal anesthesia in the operating room of the maternity hospital. The fatus was born healthy, without complications, and is not affected by OI. This case highlights the importance of multidisciplinary care and planning for pregnancies in women with OI, as well as the significance of comprehensive knowledge of the patient's history of previous fractures and clinical conditions, so that obstetricians and anesthesiologists can assist in choosing the best delivery method for both the mother's and the fetus's health.

#### KEYWORDS: CONGENITAL BONE DISEASES, PREGNANCY, OSTEOGENESIS IMPERFECTA, PRENATAL.

#### INTRODUCTION

Osteogenesis Imperfecta (OI), also known as brittle bone disease, is a group of connective tissue disorders with heterogeneous presentation, related to a deficiency in the synthesis of type I collagen. Its incidence is estimated at 1 case per 15,000-20,000 births, making it the most common genetically inherited connective tissue disorder<sup>1</sup>.

Of these cases, 85 to 90% are caused by autosomal dominant structural or quantitative mutations in collagen-related genes, with emphasis on the COL1A1 and COL1A2 genes, which encode the alpha chains of type 1 collagen. The location of this mutation within the protein determines the genotypic and phenotypic presentation of patients with Ol<sup>2</sup>.

In 1979, a classification of OI subtypes was developed based on clinical characteristics and severity. Type I is mild, non-deforming, with blue sclera; type II is characterized by a perinatal lethal form; type III is severe, with progressive deformity; type IV is moderate, with normal sclera<sup>3</sup>. Regarding genetic characteristics, 22 distinct types of the disease have been identified.

In general, clinically, this pathology is characterized by

bone fragility and deformities, associated with fractures from minimal trauma. It can also cause growth deficits depending on the clinical form presented by the patient. As it is a disorder of connective tissue, findings can extend to other systems besides the skeletal system, such as blue sclera, hearing loss, dentinogenesis imperfecta, and pulmonary dysfunction<sup>1</sup>.

With advances in the management of OI, there has been an increase in the life expectancy of patients, and more women affected by the disease reach reproductive age with a desire for pregnancy. Pregnancy in these patients is associated with maternal and fetal complications, as the presence of an autosomal dominant mutation in the mother determines a 50% risk of OI in the offspring<sup>4</sup>. Therefore, reproductive counseling in this group is essential, as well as proper management in prenatal care and childbirth to minimize negative outcomes among pregnant patients.

This is a case report of a patient with Osteogenesis Imperfecta (OI) who became pregnant, with a positive maternal and fetal outcome. The aim is to discuss possible complications of pregnancy due to this comorbidity, focusing on management recommendations for these women.

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Patient C.C.V., 25 years old, was admitted in March 2023 for cesarean section, hemodynamically stable, with normal laboratory tests and negative serologies. Primiparous, she became pregnant naturally, without planning, and was 36 weeks pregnant. She discovered the pregnancy during an ultrasound exam for preoperative evaluation before the removal of an endometrioma in the left ovary. The patient did not have prenatal care. She has osteogenesis imperfecta (type not specified), diagnosed at 6 months of age, with a history of more than 20 fractures, blue sclera, and a family history of genetic diseases such as heart disease, glaucoma, and diabetes. Despite the osteogenesis imperfecta, she has no other comorbidities, does not take medications, and has no history of hypertension, diabetes, or smoking. However, she reports social alcohol consumption. A karyotype was not performed. The fetus did not show any suspected pathology.

An elective cesarean section was performed with spinal anesthesia, a regional anesthesia with the injection of anesthetic into the subarachnoid space, blocking pain in the lower body. After delivery, a living child without OI, without apparent injuries, and with adequate vital signs was born.

#### DISCUSSION

Osteogenesis imperfecta, a rare and hereditary condition of the connective tissue, presents significant challenges for affected individuals and their families. Characterized by bone fragility and susceptibility to fractures due to defects in collagen synthesis, this condition requires a careful approach, especially during pregnancy.

For women planning to conceive, it is advisable to seek genetic counseling before conception in cases of osteogenesis imperfecta. This can help families better understand the implications of the condition and make decisions about family planning.

During pregnancy, meticulous preparation is essential, allowing for the monitoring of fetal development, early identification of complications, and guidance on appropriate prenatal practices. Complications during pregnancy, which can range from intense pain and bone deformities to more serious situations such as uterine rupture, highlight the importance of prenatal monitoring. This process requires a multidisciplinary approach, involving professionals such as obstetricians, geneticists, and psychologists, who should be fully aware of the patient's life history, including her fractures and complications resulting from the disease, to provide accurate and effective guidance during the preparation for pregnancy, childbirth, and the postpartum period.

So far, there is no definitive drug treatment for this disease. However, several methods have been associated with monitoring affected patients. One such approach includes the use of bisphosphonates, investigated as an alternative to improve bone density and reduce the chances of fractures in patients diagnosed with OI. However, the lack of conclusive scientific studies supporting the beneficial efficacy of these drugs during pregnancy highlights uncertainty about their safety during the gestational period.

Precautions during childbirth are expanded to ensure the health of all involved. The team must be prepared to offer support and specific interventions for each patient, aiming for a unique experience and adapting to the specific needs of each pregnant woman with OI, such as additional care with anesthesia, intubation, padding of the stretcher, and equipment used, among others.

The administration of anesthesia can generate uncertainty, especially when choosing medications, and challenges during tracheal intubation. It is recommended to apply anesthetics to the oropharynx area before laryngoscopy, while the patient is conscious, to assess possible difficulties in intubation. Furthermore, intubation via bronchofibroscopy and the use of a laryngeal mask airway are alternatives that should be considered<sup>10</sup>.

Delivery, often surgical, may be necessary due to the pregnant woman's pelvic deformities, cephalopelvic disproportion or the fetus' condition with osteogenesis imperfecta10. These considerations highlight the complexity of the act in patients with this condition and the importance of specific care to ensure safe and positive results.

Due to reports of different techniques, it is essential to analyze each case individually to determine the best approach to factors such as anesthesia and mode of delivery, a procedure implemented in the present clinical case. One study found that hemorrhage is a possible complication in 10% to 30% of patients with OI, due to tissue fragility and inadequate response to bleeding caused by collagen deficiency, which increases the risk of postpartum uterine atony<sup>7</sup>. During childbirth, it is crucial to observe special care, ensuring, for example, adequate positioning to avoid compression. Additionally, research has shown high rates of obstetric complications in women with osteogenesis imperfecta, including gestational diabetes, cesarean section, need for blood transfusion and occurrence of fractures both before and after childbirth<sup>8</sup>.

Diagnosis of fetal status can be conducted through the extraction of chorionic villi and imaging observations of the fetus<sup>9</sup>. During pregnancy, an ultrasound can detect OI and indicate the type, as the most serious and potentially fatal form of osteogenesis imperfecta. After the birth, the doctor checks the symptoms and performs a physical examination to make a diagnosis. If any doubts persist, the professional may choose to perform a skin biopsy or take a blood sample for genetic analysis, methods that help confirm the diagnosis and define the most appropriate treatment for the condition.

#### CONCLUSION

It is ideal that the pregnancy for those with Osteogenesis Imperfecta is planned and monitored by professionals who are experts in the patient's clinical condition and have extensive knowledge of the specific case to avoid future complications during pregnancy and childbirth. In the case reported, the patient had an unplanned pregnancy but was well managed and monitored, without serious complications.

After the pregnancy is discovered, the patient with OI must receive a multidisciplinary approach, involving professionals such as obstetricians, geneticists and psychologists, who are aware of the patient's life history, including her fractures and complications resulting from the disease. There are specific risks related to this condition during pregnancy, especially related to hemorrhages, due to tissue fragility and inadequate response to bleeding caused by collagen deficiency, which increases the risk of postpartum uterine atony, in addition to an increased risk of obstetric complications.

To date, there are no medications proven to be effective for treating this condition and there is a risk that the fetus may also be a carrier of this genetic condition. It is important and necessary to monitor and identify early OI in the baby during pregnancy.

It is generally recommended that the birth be cesarean section, given the cephalopelvic disproportion and/or pelvic deformations resulting from previous fractures. A careful analysis by the anesthetist and obstetrician is important to assess the best route of delivery for both the mother and the baby.

- Forlino A, Marini JC. Osteogenesis imperfecta. Lancet. 2016 Apr;387(10028): 1657-71.
- Panzaru MC, Florea A, Caba L, Gorduza EV. Classification of osteogenesis imperfecta: importance for prophylaxis and genetic counseling. World J Clin Cases. 2023 Apr;11(12):2604-20.
- Sillence DO, Senn A, Danks DM. Genetic heterogeneity in osteogenesis imperfecta. J Med Genet. 1979 Apr;16(2):101-16.
- Cozzolino M, Perelli F, Maggio L, Coccia ME, Quaranta M, Gizzo S, et al. Management of osteogenesis imperfecta type I in pregnancy; a review of literature applied to clinical practice. Arch Gynecol Obstet. 2016 Jan;293(6):1153-9.
- Rauch F, Glorieux FH. Osteogenesis imperfecta. Lancet. 2004 Apr;363(9418):1377-85.
- Sharma A, George L, Erskin K. Osteogenesis imperfecta in pregnancy: two case reports and review of literature. Obstet Gynecol Surv. 2001 Sep;56(9):563-6.
- das Neves JFNP, Sant'anna RS, de Almeida JR, Saldanha RM, Magalhães MG. Anestesia venosa total em paciente portador de Osteogênesis imperfecta: relato de caso. Rev Bras Anestesiol [Internet]. 2004 Sep;54(5):668-71. doi: 10.1590/S0034-70942004000500007
- Rao R, Cuthbertson D, Nagamani SCS, Sutton VR, Lee BR, Krischer J, et al. Pregnancy in women with osteogenesis imperfecta: pregnancy characteristics, maternal, and neonatal outcomes. Am J Obstet Gynecol MFM. 2021 Jul;3(4):100362.
- Cubert R, Cheng EY, Mack S, Pepin MG, Byers PH. Osteogenesis Imperfecta: mode of delivery and neonatal outcome. Obstet Gynecol. 2001 Jan;97(1):66-9.
- Vogel TM, Ratner EF, Thomas RCJ, Chitkara U. Pregnancy complicated by severe osteogenesis imperfecta: a report of two cases. Anesth Analg. 2002 May;94(5):1315-7.

# **BOERHAAVE SYNDROME: CASE REPORT**

MARCUS ALMEIDA; LEANDRO MOURA; TIMÓTEO VERÍSSIMO; RODRIGO CARVALHO; GUILHERME SOUZA; RENATA GUERRA; GUILHERME ARANTES; LEONARDO SANTOS; BRUNO SILVA; HENRIQUE LIMA

#### ABSTRACT

INTRODUCTION: Boerhaave Syndrome (BS) is defined as a spontaneous rupture of the esophagus, a rare but potentially fatal condition with a high morbidity and mortality rate, constituting the most lethal perforation of the gastrointestinal tract.

CASE REPORT: Male patient, 49 years old, alcoholic, presents with chest pain that has lasted for five days, triggered by emesis due to alcoholic beverages. He is normotensive, normocardic, afebrile and eupneic. Preserved chest expansion, left thoracic and cervical subcutaneous emphysema, and reduction in ipsilateral breath sounds. Computed tomography of the chest was requested and evidenced soft tissue emphysema in the cervical spaces, pleural effusion on the left and pneumomediastinum. Patient underwent double-mouth cervical esophagostomy, conventional gastrostomy, closed pleural orientation on the left and complementary therapy with endoscopic vacuum. There was good postoperative evolution with esophageal transit remaining during the same hospitalization.

DISCUSSION: The diagnosis of BS represents a real challenge, and its treatment, whether surgical or not, must be instituted immediately given the high morbidity and mortality. Although Non-Operative Treatment is reserved for selected patients, there is a tendency to adopt aggressive Operative Treatment for patients with late diagnosis, local or systemic complications. Complementary therapies such as esophageal stent or endoscopic aspirator present promising results with less invasiveness and reduced mortality rate.

CONCLUSION: SB should be remembered as a differential diagnosis of chest pain in the emergency department. Despite the scarce case series, early diagnosis and treatment constitute the most relevant prognostic factor.

#### KEYWORDS: BOERHAAVE SYNDROME; ESOPHAGEAL PERFORATION; MEDIASTINITIS.

#### INTRODUCTION

Boerhaave syndrome is defined as the spontaneous rupture of the esophagus, a rare but potentially fatal condition. Although it occurs at a low incidence rate of 3.1 per 1,000,000, its mortality rate is high, ranging from 35% to 40%, making it the most lethal perforation of the gastrointestinal tract.<sup>1</sup>

It was first described in 1724 by the Dutch surgeon Hermann Boerhaave, who reported in his monograph the symptoms experienced by Admiral Von Wassenauer: sudden and excruciating chest pain after vomiting following a copious meal, which led to his death. The pathology was confirmed by autopsy, which revealed an esophageal perforation.<sup>2</sup>

The pathophysiological mechanism involved in this condition is due to a sudden increase in intraluminal esophageal pressure with the absence of relaxation of the upper esophageal sphincter, leading to a complete rupture of all layers of the esophagus (Haba et al, 2020). The main reported symptoms are chest pain, vomiting, fever, subcutaneous emphysema, and dyspnea. They are classically described by the Mackler triad composed of vomiting, chest pain, and subcutaneous emphysema.<sup>1</sup>

The diagnostic workup requires a clinical history that includes risk factors for BS with suggestive manifestations.

Among the complementary tests, one can use chest X-ray (CXR), which is a method with high sensitivity, up to chest and abdominal CT with oral contrast, which is the most specific method, capable of identifying the site of esophageal perforation, as well as its extension and associated complications. Other tests such as contrast esophago-gram and upper gastrointestinal endoscopy (UGIE) may be used for selected patients.<sup>3</sup>

The management can be instituted by ST or NST, although endoscopic therapies have stood out more recently, especially those using vacuum therapy or stent placement, corresponding to the main or adjuvant therapy.

Early diagnosis is a real challenge and is one of the main prognostic factors, especially those diagnosed within the first 24 hours, as the mortality rate increases considerably in those with late evolution, reaching up to 65% mortality due to mediastinitis and rapid progression to septic conditions.<sup>4</sup>

#### **CASE REPORT**

Male patient, 49 years old, asthmatic and alcoholic, was referred for evaluation by the general surgery team due to ventilator-dependent chest pain and thoracic subcutaneous emphysema within five days of evolution. Symptoms

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#### Aparecida de Goiânia.

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MARCUS ALVES CAETANO DE ALMEIDA Endereço: Av. Diamante, s/n - St. Conde dos Arcos, Aparecida de Goiânia - GO, 74969-210. Email: marcuscaetanomed@gmail.com were triggered after vomiting due to alcohol intake. Absence of dyspnea, nausea, vomiting, or fever on admission. Patient undergoing antimicrobial therapy in the originating unit.

The physical examination revealed a patient in fair overall condition, with good skin color, well-hydrated, normotensive, normocardic, afebrile, and eupneic in ambient air. On examination of the respiratory system, preserved chest expansibility was noted, along with subcutaneous emphysema on palpation, predominantly in the thoracic and left cervical regions, and reduced vesicular murmur on auscultation ipsilaterally. Neurological, cardiovascular, and abdominal systems showed no alterations.

In the face of the diagnostic hypothesis of BS, a CT scan of the neck, chest, and abdomen with intravenous contrast was requested, which revealed marked soft tissue emphysema in the anterior and paravertebral cervical spaces (Figure 01), as well as left pleural effusion associated with pneumomediastinum (Figure 02). The abdominal CT scan was normal. Laboratory tests showed leukocytosis (15,370/mm<sup>3</sup>), with normal band neutrophil count, and elevated C-reactive protein (65.1 mg/L).

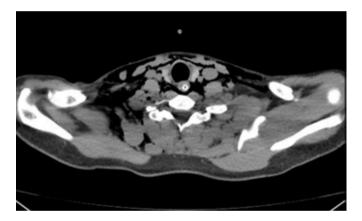


Figure 01 - Extensive soft tissue emphysema in cervical and paravertebral compartments. Source: Personal archive.



Figure 02 - Tomographic signs of pneumomediastinum, white arrow. Large left pleural effusion, yellow arrow. Source: Personal archive.

After clinical reassessment, closed water seal pleural drainage was indicated on the left, followed by urgent upper gastrointestinal endoscopy, which showed a 2 cm diameter esophageal perforation in the left lateral wall, located 40 cm from the upper dental arch and 1 cm from the esophago-gastric junction. It communicated with the left pleural space, which showed extensive contamination (Figure 03).

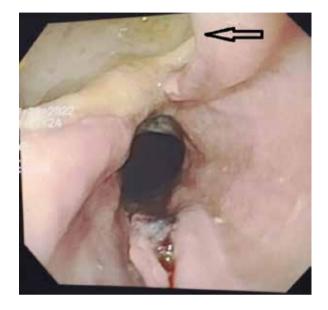


Figure 03 - Endoscopic evidence of transmural esophageal injury on the left lateral wall, indicated by the black arrow. Source: Personal archive.

It was indicated, in the same surgical procedure, the performance of cervical esophagostomy with double stoma for transit diversion associated with conventional gastrostomy tube for enteral nutrition (figure 04).



Figure 04 - Patient in immediate postoperative period. Source: Personal archive.

In the postoperative course, the thoracic drain showed enteral-like output, indicating the presence of a left esophago-pleural fistula. Due to its persistence, on the 12th postoperative day (POD), endoscopic vacuum therapy was indicated along with Total Parenteral Nutrition (TPN) and decompressive GT. After four days of this therapeutic approach, there was a significant reduction in thoracic drain output, followed by resolution of the fistula, leading to the early removal of the thoracic drain and endoscopic vacuum therapy.

Underwent a follow-up EGD on the 31st POD, which showed a healed esophageal lesion in good condition. Consequently, within 48 hours, esophageal transit reconstruction was performed with primary end-to-end anastomosis in a single plane associated with cervical laminar drainage. The patient was discharged 72 hours after the procedure following a negative methylene blue test.

#### DISCUSSION

BS is a rare cause of esophageal perforation in the general population but is associated with known risk factors, which involve increased esophageal intraluminal pressure. These include uncontrollable vomiting, often resulting from alcohol consumption, asthma attacks, straining during bowel movements, and heavy lifting.<sup>5</sup> In this case study, at least two of these factors are identifiable: episodes of vomiting and asthma, in addition to the significant association with alcohol consumption.

Despite the high mortality rate associated with delayed diagnosis of BS, this case demonstrates a prolonged clinical course, contrasting with the natural history of the disease. The patient presented to the emergency department with a stable clinical condition and no signs of sepsis based on the quick Sequential Organ Failure Assessment (qSOFA) score, despite at least 96 hours of esophageal perforation evolution. This outcome may be attributed to the intravenous antimicrobial therapy initiated at the referring facility, as well as clinical support and likely delayed but progressive contamination of the mediastinal and pleural compartments. In a classic progression, rapid onset of sepsis due to mediastinitis is expected within approximately 12 to 24 hours, with progressive organ dysfunction beyond 48 hours of evolution. However, the patient's presentation with thoracic pain, dyspnea, and subcutaneous emphysema made spontaneous esophageal rupture a feasible diagnostic hypothesis.

From an anatomical standpoint, it is noteworthy that during the endoscopic examination, the esophageal perforation occurred in the left lateral wall, located one centimeter from the esophagogastric junction, with a length of two centimeters. These data reinforce and corroborate with the literature, except for the significant proximity to the esophagogastric junction.<sup>2</sup>

Given the presented case, it is evident that the adoption

of NST is not feasible due to late clinical presentation, with at least five days of evolution, extensive contamination of the mediastinal and pleural compartments, as well as signs of ventilatory discomfort and chest pain. Based on objective criteria, as proposed by the Pittsburgh score, a sum of six points is calculated, including leukocytosis, pleural effusion, uncontained fistula, and delayed diagnosis, which also supports the need for aggressive surgical treatment.<sup>36</sup>

Therefore, a GT feeding tube was constructed as a method of long-term enteral nutrition, along with double-lumen cervical esophagostomy as a principle of contamination control, in addition to closed left pleural drainage. Although not performed, Nissen fundoplication with perforation blocking was also a feasible option due to its proximity to the esophagogastric junction.<sup>4</sup>

During the postoperative management, despite the surgical therapy instituted, persistence of the left esophagopleural fistula was evidenced, leading to the possibility of using endoscopic vacuum therapy, which represents a minimally invasive option with results comparable to primary closure, despite the low number of cases. In current literature, success rates range from 70 to 100%, with mortality rates of 7 to 18% and complication rates of 10 to 14%.<sup>7</sup> After 96 hours of application, aiming at reducing contamination and conditioning the lesion, there was closure of the fistula and healing of the perforation, objectively assessed by follow-up endoscopy.<sup>8</sup>

The reconstruction of the esophageal tract occurred during the same hospitalization and represented an important milestone for the patient's quality of life in the postoperative period, as oral diet became feasible.

#### CONCLUSION

SB, despite its rarity, should be considered as a differential diagnosis in cases of chest pain in the emergency department, especially in patients with known risk factors.

There is much debate about the ideal therapeutic modality, and among its possibilities, it is convenient to aim for risk stratification, with recommended scores such as the Pittsburgh score to define surgical therapy or not. Regardless of the therapeutic approach, it is necessary to follow principles for the treatment of this serious condition, which include treatment and control of local contamination, enteral nutrition, and wide drainage if necessary.

The gold standard surgical treatment is primary repair of the lesion for eligible patients. Other alternative modalities, such as exclusion and diversion of transit, are options in unfavorable clinical scenarios. Endoscopic treatment, either vacuum therapy or self-expanding stent placement, has gained prominence, with some studies showing outcomes similar to the gold standard treatment. Finally, despite the limited literature and case studies on BS, it is demonstrated that the most relevant prognostic factor is early diagnosis and treatment.

- Allaway MGR, Morris PD, B. Sinclair J, Richardson AJ, Johnston ES, Hollands MJ. Management of Boerhaave syndrome in Australasia: a retrospective case series and systematic review of the Australasian literature. ANZ Journal of Surgery. 2020 Dec 14;91(7-8):1376–84.
- Salvador-Ibarra IJ, Pizaña-Davila A. Síndrome de Boerhaave: reporte de un caso y revisión de la literatura. Cirugía y Cirujanos. 2021 Dec 21;89(92).
- Chirica M, Kelly MD, Siboni S, Aiolfi A, Riva CG, Asti E, et al. Esophageal emergencies: WSES guidelines. World Journal of Emergency Surgery. 2019 May 31;14(1).
- Dias P, Martins De Freitas V, Medina Da Fonseca C. REVISÃO DE LITERA-TURA: MANEJO NA PERFURAÇÃO DE ESÔFAGO. REVISTA EMESCAM DE ESTUDOS EM SAÚDE. 2019 Jan;1(1).
- Haba Y, Yano S, Akizuki H, Hashimoto T, Naito T, Hashiguchi N. Boerhaave syndrome due to excessive alcohol consumption: two case reports. International Journal of Emergency Medicine. 2020 Nov 30;13(1).
- Schweigert M, Santos Sousa H, Solymosi N, Yankulov A, Fernández MJ, Beattie R, et al. Spotlight on esophageal perforation: A multinational study using the Pittsburgh esophageal perforation severity scoring system. The Journal of Thoracic and Cardiovascular Surgery. 2016 Apr;151(4):1002–11.
- Pattynama LMD, Eshuis WJ, van Berge Henegouwen MI, Bergman JJGHM, Pouw RE. Vacuum-stent: A combination of endoscopic vacuum therapy and an intraluminal stent for treatment of esophageal transmural defects. Frontiers in Surgery. 2023 Feb 22;10.
- Loske G, Schorsch T. Endoskopische Vakuumtherapie beim Boerhaave-Syndrom. Der Chirurg. 2016 Jun 3;87(8):676–82.

# SURGICAL CORRECTION OF PULMONARY ARTERY ANEURYSM. CASE REPORT

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#### ABSTRACT

Pulmonary artery aneurysm (PAA) is a rare condition that can occur in both sexes, 89% in the main pulmonary artery and 11% in the branches of the pulmonary artery. It presents a risk of compression of adjacent structures, such as the left coronary artery (LMCA), among others. Rupture of the PAA can result in high mortality rates, ranging between 50 and 100%. We report a case on the clinical evolution after the surgical approach of a patient with symptomatic PAA, with potential compression of the LMCA.

#### KEYWORDS: ANEURYSM; THORACIC SURGERY; PULMONARY ARTERY.

#### INTRODUCTION

Pulmonary artery aneurysm (PAA) is a rare condition, first described in 1860. Based on autopsies, a prevalence of approximately 1 in 13,696 individuals was estimated <sup>1</sup>. It generally occurs in both sexes, with 89% in the main pulmonary artery and 11% in the branches of the pulmonary artery. Among the branches, the most common incidence occurs in the left branch <sup>2</sup>. They can arise secondary to various etiologies such as infection, malignancies, congenital heart disease, or vasculitis <sup>3</sup>.

PAA is characterized by focal dilatation of the pulmonary artery involving all three layers of the vessel wall <sup>4</sup>. The condition is diagnosed when the diameter of the vessel dilatation is greater than 1.5 times the upper limits of normal. In most studies, a gender-based cutoff was not followed, considering an aneurysm all cases with a pulmonary artery diameter greater than 40 mm <sup>3</sup>. PAA presents a risk of compression of adjacent structures, such as the left main coronary artery (LMCA), the main bronchus, and/or the recurrent laryngeal nerve. Among potentially fatal complications, rupture of the PAA can result in high mortality rates, ranging from 50 to 100% <sup>4</sup>.

There is no specific guideline for the best approach when PAA is diagnosed. Among the main surgical indications are an aneurysm diameter greater than 55mm; an increase in diameter greater than 5mm in six months; compression of adjacent structures; thrombus formation in the aneurysm sac; onset of clinical symptoms; evidence of valve pathologies; pulmonary hypertension; and signs of aneurysm rupture or dissection <sup>2</sup>.

The aim of this report is to demonstrate the clinical evolution following the surgical approach of a patient with symptomatic PAA, with potential adjacent compression of the left main coronary artery.

#### **CASE REPORT**

The patient is a 71-year-old male presenting for a cardiology evaluation for a dental implant procedure. During the consultation, he complains of typical chest pain during exertion, followed by syncope, even during routine bike rides, which started in the last few months. He reports experiencing near-syncope episodes while evacuating, but of different characteristics, with prodromes before the episodes. His medical history includes hypertension, dyslipidemia, prediabetes, and an ischemic stroke without sequelae in 1990. He brings an ultrasound (US) of the carotid arteries from 2018 showing plaques compromising 25% of the vessel lumen. He was previously diagnosed with pulmonary valve stenosis and underwent pulmonary valve commissurotomy in 1986. He is currently taking Diovan HCT®, Plenance Eze®, and Glifage . On cardiovascular auscultation, there is a +++/6+systolic and diastolic murmur in the pulmonary focus. The electrocardiogram shows sinus rhythm and right bundle branch block. An echocardiogram with stress, performed in 2020, revealed a left ventricular ejection fraction (LVEF) of 74% (Teichholz) with no segmental contractility abnormalities at rest or during stress.

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Due to cardiovascular risk, a new echocardiogram and carotid ultrasound were requested. The echocardiogram showed: LVEF of 55%. Posterior wall and septal wall thickness of 9 mm. Right Ventricle (RV) - Right Atrium (RA) gradient of 25 mmHg. Systolic pressure in the pulmonary artery of 30 mmHq. Significant ectasia in the pulmonary artery trunk and its left branch associated with moderate pulmonary valve regurgitation (average gradient of 9 mmHg and velocity of 2.0 m/s), as well as right chamber overload, with an internal diameter of the RV of 47 mm, S' wave of 13.2 cm/s. Due to the pulmonary artery ectasia, a CT angiography (CTA) of the pulmonary and coronary arteries was requested for further investigation, with the following findings: Pulmonary Artery Aneurysm (PAA) (internal diameter of 47 mm in the trunk region and diameter of the left branch of 24 mm), with its trunk showing intimate relationship with the TCE (11 mm distance) (Figure 1), without causing compression at rest. Moderate luminal reduction in the right coronary artery (RCA) and slight reduction in the second marginal artery (Mg2).

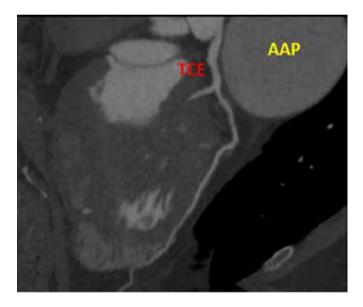


Figure 01. Angiotomography showing pulmonary artery aneurysm (PAA) in close proximity to the left coronary artery trunk (LCA).

Cardiac magnetic resonance imaging was performed for better anatomical evaluation, showing: Left Ventricular Ejection Fraction (LVEF) of 65% and Right Ventricular Ejection Fraction (RVEF) of 69%. Cardiac chambers with preserved dimensions. Preserved biventricular systolic function. Absence of myocardial fibrosis. Mild pulmonary valve regurgitation (regurgitant fraction of 18%). Presence of significant aneurysmal dilation of the pulmonary artery, measuring 55 mm in its largest diameter (Figure 2).

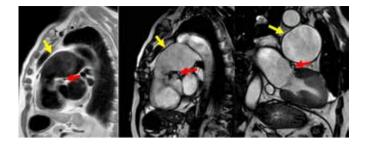


Figure 02. MRI showing pulmonary artery aneurysm (yellow arrow) revealing proximity to the left coronary artery (red arrow).

After discussion with the Heart Team, surgical correction of the aneurysm was indicated. The procedure was performed with a median sternotomy, the heart in sinus rhythm, and an 80mm diameter aneurysm identified. Heparinization followed by aortic and atrial cannulation with initiation of cardiopulmonary bypass (CPB) under hypothermia below 25°. Aortic clamping was performed, and cardioplegia was administered into the aortic root with Custodiol®. The tricuspid valve was calcified. The procedure continued with pulmonary arteriotomy and pulmonary valve commissurotomy, end-to-end anastomosis of a 30 Dacron tube with the left pulmonary artery, side-to-side anastomosis with a tube, and end-to-end anastomosis of the pulmonary origin with the tube. Reinforcement with biological glue and closure of the aneurysm over the tube. Warm-up and air removal maneuvers were performed. Aortic declamping occurred at 44 minutes with return to sinus rhythm after 10J defibrillation. CPB weaning was done at 55 minutes, mediastinal drainage with two Blake drains, two temporary pacemaker wires in the right ventricle, hemostasis revision with biological glue, and pericardium reconstruction with Goretex®. Layer closure and dressing. He had a good postoperative evolution without surgical complications during hospitalization. Twice, after 9 and 24 days post-procedure, he presented acute atrial fibrillation (AF) in the emergency room, returning to sinus rhythm after chemical reversion with amiodarone.

In the post-surgical follow-up appointment, the patient brought an echocardiogram (ECO) and an angioCT of the pulmonary arteries. The ECO showed a diagnosis of mild pulmonary valve stenosis, with a peak gradient of 35 mmHg and mild pericardial effusion. The angioCT revealed a prosthesis in the main pulmonary artery that starts from the valve and extends to the bifurcation of the right and left branches, which has a preserved caliber and is patent. The maximum caliber of the prosthesis measures 30 mm. Correction of the pulmonary artery aneurysm with a prosthesis was successful, without signs of complications.

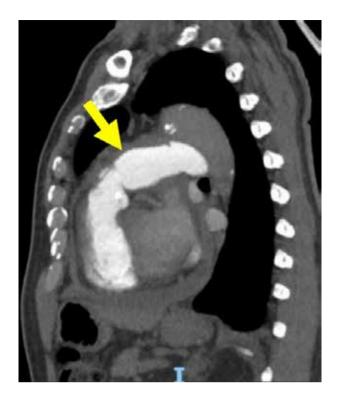


Figure 03. Post-procedure angiotomography showing Dacron tube (yellow arrow) demonstrating correction of the PAA.

The patient is currently using Eliquis, Diovan HCT, Amiodarone, and Dexilant. He is clinically stable, with no new episodes of atrial fibrillation, pre-syncope, or syncope, reporting an improved quality of life without complaints of angina-like chest pain.

#### DISCUSSION

The PAA is a rare entity among pulmonary vascular diseases, but with unfavorable and potentially fatal outcomes. Patients may go for long periods without a diagnosis, as a considerable portion of cases are asymptomatic, with diagnosis often occurring incidentally through imaging exams done for other reasons. Fatality can result from aneurysmal rupture with frank hemoptysis, pulmonary artery dissection, and compression of the coronary artery, leading to acute coronary syndrome.

It presents a varied spectrum of clinical manifestations depending on the size, pressure, growth rate, and contact with adjacent structures. In situations of low pressure and slow growth, patients can remain asymptomatic for years to decades <sup>4</sup>.

Studies unanimously report the difficulty in finding the ideal management after diagnosis, primarily because there are no specific guidelines for PAA to date. Treatment should be tailored to each case, considering the underlying cause, symptoms, risk-benefit of surgical intervention, and comorbidities. In cases where conservative management is chosen, treatment should include managing underlying conditions, pulmonary hypertension if present, and regular imaging follow-up.

When there is dilation of the pulmonary artery (PA), extrinsic compression on the left main coronary artery (LMCA) can occur, as in the case reported. This becomes likely when the diameter exceeds 55 mm or when the ratio between this diameter and the diameter of the aortic root is at least 1.98. Its development depends on a chronic increase in the diameter of the PA, usually associated with pulmonary hypertension (PH). The most common clinical presentation is angina-like chest pain, associated or not with dyspnea. Syncope, arrhythmias, or myocardial infarction may also be present. It has been observed that abnormal flow through the pulmonary valve can cause tension in the vessel wall due to the eccentric jet outflow from the right ventricle, leading to weakness in the vessel wall. A review identified 19 cases due to pulmonary valve abnormalities, with no association with PH. The median age was 50 years, with cases reported up to 79 years, and a similar gender distribution <sup>3</sup>.

After an extensive review inferring the rarity of the diagnosis, we also identified numerous cases discovered in individuals younger than the case in question. The low pressure in the PA explains the long asymptomatic period, with symptoms starting from a significant increase in arterial diameter of 55 mm, which, according to the review, increases the risk of compression of adjacent structures

Some factors increase the risk of PAA rupture, such as chronic PAH, PA pressure greater than 50 mmHg, PA diameter greater than 75 mm, and annual growth exceeding 2 mm. There are several options to reduce the risk of these complications, including addressing the underlying cause, continuous monitoring through imaging studies as already mentioned, and even the need for surgical intervention.

Aneurysmorrhaphy and aneurysmectomy surgeries are well described for the treatment of aneurysms confined to the pulmonary trunk. Early surgery should be considered in patients with reasonable surgical risk to prevent worsening of cardiac function due to diameter progression and difficulty in ventilation due to chronic bronchial obstruction and atelectasis. Procedures in younger patients reduce postoperative morbidity and mortality<sup>2</sup>.

The patient in question had more than one formal indication for surgical intervention, including a diameter of 55 mm, presence of symptoms (chest pain/syncope), and associated pulmonary valve pathology. Due to the clinical benefits of reducing symptoms, preventing progression, and avoiding complications related to the PAA, early surgical treatment, with its proper indications, showed good results and improved the patient's quality of life.

This study, along with the scarce number of published cases, may contribute to advising medical teams on possible interventions for PAA, while awaiting guidelines to guide the ideal management.

- De Souza AH, Petto J, Gardenghi G. Surgical correction of pulmonary artery aneurysm with extrinsic compression of the left main coronary artery: a case report. Journal of Clinical and Translational Research. 2019. DOI: 10.18053/jctres.05.201902.003
- Kreibich M, Siepe M, Kroll J, Hohn R, Grohmann J, Beyersdorf F. Aneurysms of the Pulmonary Artery Circulation. 2015 Jan 20;131(3):310-6. DOI: 10.1161/ circulationaha.114.012907
- Gupta M, Agrawal A, lakovou A, Cohen S, Sha R, Talwar A. Pulmonary artery aneurysm: a review. Pulmonary Circulation [Internet]. 2020 Jan;10(1):1-10. Available https://www.nbci.nlm.nih.gov/pmc/articles/PMC7052473/. DOI: 10.1177/2045894020908780
- Flaifel M, Rohan Suresh Daniel, Nakanishi H, Than CA, Shiakos G, Ioannis Tzanavaros. A Novel Approach for the Treatment of Pulmonary Artery Aneurysm Repais Using Inclusion Technique: A Case Report. Cureus. 2023 Mar 21. DOI: 10.7759/cureus.36456
- Falchetto EB, Saad JA, Almeida FL de, Washizu EKM, Mandil A. Obstrução extrínseca do tronco da coronária esquerda causada por dilatação da artéria pulmonar associada à esquistossomíase. Revista Brasileira de Cardiologia Invasiva. 2015 Apr;23(2):148-51. DOI: 10.1016/j.rbci.2015.12.016

# CASE REPORT: MIRIZZI SYNDROME DIAGNOSED INTRAOPERATIVELY

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#### ABSTRACT

The article in question describes a clinical case of Mirizzi syndrome, a rare condition in which there is compression or obstruction of the common bile duct due to gallstones impacted in the cystic duct or gallbladder. Mirizzi syndrome is a potentially serious complication of cholelithiasis (the presence of stones in the gallbladder). However, the case report details the patient's clinical presentation, which was incredibly asymptomatic. During the surgical procedure (cholecystectomy), surgeons identified anatomical abnormalities and changes typical of Mirizzi syndrome, confirming the intraoperative diagnosis. The objective of the case report is to provide a detailed description of a specific clinical case, demonstrating the clinical presentation, diagnosis, treatment and evolution of this rare condition. Also using the Data Collection method, where the patient's clinical data were collected, including medical history, physical examinations, laboratory test results, diagnostic imaging images (such as ultrasound, computed tomography, etc.) and findings intraoperatively. Finally, in final considerations, the results were analyzed in light of the medical literature and discussed in terms of differential diagnosis, therapeutic management and specific challenges encountered during the patient's treatment.

#### KEYWORDS: MIRIZZI SYNDROME; INTRAOPERATIVE; SURGERY; CASE REPORT

#### **INTRODUCTION**

Mirizzi syndrome is a rare complication of cholelithiasis, characterized by compression or obstruction of the common bile duct due to impaction of gallstones in the cystic duct or gallbladder. Initially described by Pablo Mirizzi in 1948, this condition poses a diagnostic and therapeutic challenge due to its varied clinical presentation and the need for immediate surgical intervention in many cases. Mirizzi syndrome can mimic other biliary pathologies, such as acute cholecystitis, gallbladder carcinoma, and common bile duct stenosis, making preoperative diagnosis often challenging.

The typical clinical presentation includes abdominal pain, jaundice, fever, and abnormalities in liver function tests. The definitive diagnosis of Mirizzi syndrome is often established during surgery, when characteristic intraoperative findings are identified, such as adhesions between the gallbladder and the common hepatic duct, as well as impacted stones in the bile duct<sup>1</sup>.

In this case report, we describe a patient with Mirizzi syndrome whose diagnosis was established during cholecystectomy due to the presence of intraoperative complications. This case highlights the diagnostic and therapeutic challenges associated with this condition, as well as the management strategies adopted to ensure a safe and effective surgical intervention.<sup>2</sup> By reporting this clinical case, we aim to contribute to the understanding and management of this rare condition, providing data and results that may assist in the early identification and appropriate treatment of Mirizzi syndrome.

#### OBJECTIVE

Report the case of a patient describing a case of cholelithiasis with a complication of Mirizzi syndrome, highlighting the clinical presentation, diagnostic findings, treatment and intraoperative findings and discussing the intraoperative considerations and challenges associated with the presence of impacted stones and fistula, as well as the strategies used to manage these complications during laparoscopic cholecystectomy.

#### METHOD

Patient M.R.R, a 26-year-old woman, was selected based on clinical presentation and diagnosis of cholelithiasis with a complication of Mirizzi syndrome. Data were collected from the patient's medical history, including reported symptoms, laboratory test results, imaging findings, and information about the surgical procedure.

The patient's previous exams were reviewed, including results of complete blood count, liver enzymes (GGT, AST, ALT), bilirubin, amylase, and lipase.<sup>3</sup> The clinical case was detailed, including the patient's medical history, physical findings, results of previous exams, and intraoperative findings. The collected data were analyzed in light of rele-

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LEONARDO ETERNO MEDEIROS DOS SANTOS E-mail: leonardmed@icloud.com vant medical literature, discussing the symptoms, diagnosis, treatment, and outcomes of the case in question.

#### CASE REPORT

Patient M.R.R, female, 26 years old, white, was admitted to the Hospital Estadual de Aparecida de Goiânia for elective cholecystectomy after an ultrasound diagnosis of cholelithiasis. The patient reported a 6-month history of epigastric pain, nausea and intolerance to fatty foods. She had been hospitalized 15 days before with refractory pain symptoms in the right hypochondrium, but without reports of cholestasis. On physical examination, the patient did not present jaundice, abdominal pain, distension or palpable masses<sup>4</sup>.

The patient's previous hospitalization exams showed leukocytosis of 13,540 with 79% segmented, GGT of 85, normal bilirubin levels, normal AST and ALT levels, and normal amylase and lipase levels. The patient underwent laparoscopic cholecystectomy, during which impacted stones were identified in the infundibulum of the gallbladder with a fistula to the main bile duct<sup>5</sup>.

Surgical conversion to laparotomic access was performed and intraoperative cholangiography was performed, observing erosion of the lateral wall of the main bile duct in less than a third of its circumference as a result of the impacted stone. The surgical treatment instituted was choledochroplasty with drainage of the cavity<sup>6</sup>.

Given the benign evolution of the case, the patient was discharged on the sixth day after the operation without bile contents externalized in the sentinel drain, which was removed. Upon post-operative return after 15 days, the patient remained asymptomatic.

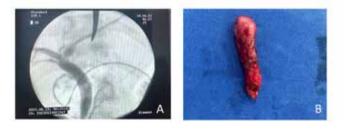


IMAGE 01 - The intraoperative images of the surgical approach show in A an intraoperative cholangiogram, demonstrating erosion of the lateral wall of the common bile duct in less than one-third of its circumference due to the impacted stone, and in B the result of cholecystectomy.

#### DISCUSSION

The Mirizzi syndrome can be challenging to diagnose, especially during surgery, due to its similarity to other conditions that can also cause bile duct obstruction. Discussing strategies to differentiate Mirizzi syndrome from other pathologies during surgery can be crucial for proper management. Once Mirizzi syndrome is diagnosed during surgery, the surgical approach needs to be carefully considered. This may include discussing treatment options such as removing the gallstones, gallbladder resection, or, in more severe cases, biliary reconstruction<sup>7</sup>.

Mirizzi syndrome can be associated with intra- and postoperative complications, such as bile duct injuries, infections, and bile leaks. It is important to discuss strategies for managing these complications, as well as preventing their occurrence during and after surgery. Emphasizing the importance of communication and collaboration between different specialties can be crucial to ensuring the best outcome for the patient<sup>8</sup>.

In conclusion, due to its uncommon occurrence in surgical practice and the potential complications associated with its management, we believe that Mirizzi syndrome should always be included in the differential diagnosis of patients with cholecystolithiasis presenting with clinical or laboratory abnormalities.

- BASTOS, A. P.; MAIA, L. M. de O. Síndrome de Mirizzi tipo V um desafio diagnóstico e de prognóstico alarmante na colelitíase: relato de caso. Revista de Saúde, [S. l.], v. 10, n. 2, p. 56–60, 2019. DOI: 10.21727/rs.v10i2.1706. [Acesso em 4 fev. 2024] Disponível em: http://editora.universidadedevassouras.edu.br /index.php/RS/article/view/1706;
- Nagakawa T, Ohta T, Kayahara M, Ueno K, Konishi I, Sanada H. A new classification of Mirizzi syndrome from diagnostic and therapeutic viewpoints. Hepatogastroenterology 1997;
- DALTRO, Michelli; BATISTA, Alexsandro; FECHTNER, Renata; LEMES, Polyana; MENEGATTI, Pedro; VENDRAMEL, Bruna; AZAMBUJA, Kamylla. Relato de caso: Síndrome de Mirizzi diagnosticada no intra-operatório. COORTE - Revista Científica do Hospital Santa Rosa, [S. I.], n. 08, 2018. [Acesso em: 5 fev. 2024]. Disponível em: https://revistacoorte.com.br/index.php/ coorte/ article/ view/106;
- TORREZ, S. E. P.; MOTA, I. A. C. Síndrome de Mirizzi: revisão de literatura: Mirizzi Syndrome: a review of the literature. Brazilian Journal of Health Review, [S. I.], v. 5, n. 4, p. 16911–16918, 2022;
- Lacerda P.S. et al, Síndrome De Mirizzi: Um Grande Desafio Cirúrgico, ABCD Arq Bras Cir Dig 2014;27[3]:226-226;
- FONSECA-NETO, Olival Cirilo Lucena da; PEDROSA, Maria das Graças Lapenda; MIRANDA, Antonio Lopes. Manejo cirúrgico da síndrome de Mirizzi. Revista Scielo Jun 2008. Disponível em: https://doi.org/10.1590/ S0102-67202008000200002;
- Reverdito, R., et al, Síndrome De Mirizzi Graus III E IV: Tratamento Cirúrgico, Rev. Col. Bras. Cir. 2016; 43[4]: 243-247;
- McSherry CK, Fertenberg H, Virshup M. The Mirizzi syndrome: suggested classification and surgical therapy. Surg Gastroenterol 1982; 1:219-25;
- Morelli A, Narducci F, Ciccone R. Can Mirizzi syndrome be classified into acute and chronic form? An endoscopic retrograde cholangiography (ERC) study. Endoscopy 1978; 10:109-12.

# SINGLE FETAL ABORTION IN TWIN PREGNANCY WITH THE PRESENCE OF A PAPYRACEOUS FETUS: CASE REPORT AND CLINICAL IMPLICATIONS

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#### ABSTRACT

Objectives: To present and analyze a clinical case of fetal abortion in a twin pregnancy with the presence of a papyraceous fetus, with an emphasis on diagnostic evaluation, clinical management and consequences for the coexisting fetus and maternal health. Methods: Work carried out retrospectively and descriptively of a patient's clinical condition. The data were obtained with the patient's consent through medical records, in addition to exams and the pregnant woman's notebook. The remaining information was acquired through a bibliographic search on the Google Scholar, PubMed and Scielo platforms with the descriptors twin pregnancy, papyraceous fetus, fetal abortion, intrauterine death, twins. Discussion: Twin pregnancies are considered risk factors for fetal and maternal deaths. The occurrence of death of only one of the fetuses at the beginning of twin pregnancy, allows the occurrence of a rare event called papyraceous fetus, characterized by intrauterine retention of the deceased fetus, generating its mummification. This situation can generate serious complications for the mother and the viable fetus. Conclusions: It is a rare condition that can pose risks to the health of the mother and the viable fetus. However, in the clinical case described, there was no maternal compromise and the viable fetus showed good viability.

#### KEYWORDS: TWIN PREGNANCY, FETAL ABORTION, PAPYRACEOUS FETUS, INTRAUTERINE DEATH, TWINS.

#### INTRODUCTION

The term "fetus papyraceus" is used to describe a degenerated ovum that does not progress beyond the early stages of embryogenesis. The phenomenon involves the fetal demise of one of the twins in early pregnancy, with the deceased fetus likely compressed due to the growth of the healthy fetus, followed by the intrauterine retention of the deceased fetus for at least 10 weeks<sup>1-3</sup>.

This phenomenon is a rare event that occurs in 0.018-0.02% of multifetal pregnancies. It occurs more specifically in monochorionic twin pregnancies, resulting in the abortion of one of the fetuses. The cause is usually idiopathic but may be related to twin-to-twin transfusion syndrome, inadequate umbilical cord insertion, velamentous insertion, or genetic and chromosomal anomalies<sup>3-5</sup>.

In the context of twin pregnancies, we can predominantly categorize them into two types: monochorionic and dichorionic. Monochorionic pregnancies are notably associated with a significantly higher risk of malformations. Among the most common complications in this clinical scenario is fetal death due to inadequate vascular connections in the shared placenta<sup>2,6</sup>.

The prognosis of pregnancy after the death of one of the twins is mainly influenced by the gestational age at the time of fetal demise and by chorionicity, regardless of amnionicity. When loss occurs in the first trimester, the death of one fetus does not seem to be associated with adverse effects on the development of the survivor, especially in diamniotic dichorionic pregnancies<sup>4</sup>.

It is observed that the later the occurrence of twin fetal death, the greater the complications for both the surviving fetus and the mother. These complications include neural tube disorders, which can result in cerebral palsy, as well as cerebral hypoxia due to the diversion of blood from the healthy fetus to the non-surviving fetus through the placenta, causing ischemic brain damage. Additionally, other areas of the body may also be affected, such as the digestive system, with the occurrence of gastrointestinal tract atresia, and the renal system, with renal agenesis<sup>1,2</sup>.

These complications require careful medical monitoring and appropriate treatment to ensure the safety of both the mother and the healthy fetus being carried. It is

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ADELINO CRISTOVÃO NETO Av. das nações, 616 - bairro vera cruz, aparecida de goiânia - GO CEP: 74976-190. E-mail: dr.adelinocn@hotmail.com ana.p.m.ferreira@aluno.famp.edu.br essential for pregnant women to receive adequate medical care during pregnancy to monitor the health of the fetus and the mother and to take preventive measures when necessary<sup>2</sup>. In this regard, the authors reported a clinical case of a dichorionic and diamniotic pregnancy with the presence of a papyraceous fetus resulting from a 12-week abortion.

#### CASE REPORT

Patient, 24 years old, female, G2P0A, without morbid antecedents, started prenatal care at a Basic Health Unit on January 12, 2023, with an approximate gestational age of 7 weeks.

The patient underwent the first ultrasound (1st trimester) on February 10, 2023, showing a twin pregnancy of 11 weeks and 5 days, dichorionic and diamniotic with the presence of amniotic membrane, yolk sac, embryo in both gestational sacs, normal amount of amniotic fluid in both cavities, and apparently normal fetal anatomies (Figure 1).

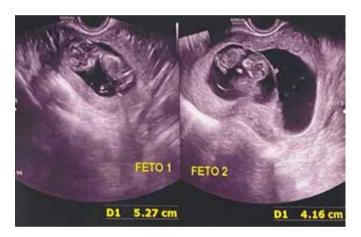


Figure 1: 1st-trimester morphological ultrasound at 11 weeks and 5 days showing dichorionic and diamniotic twin pregnancy, with an embryo in both gestational sacs Source: Shiokawa Clinic

In the second ultrasound performed on March 6, 2023, a twin pregnancy was evident, with fetus 1 showing present, rhythmic heartbeats with good vitality and biometrics compatible with 14 weeks and 3 days (Figure 2). The ultrasound evaluation of fetus 2 showed a present embryo, with a crown-rump length measuring 55.6 cm, absence of fetal movements and heartbeats, irregular gestational sac, signs of fetal resorption, and biometrics compatible with 12 weeks and 1 day (Figure 3).



Figure 2: Obstetric ultrasound showing a living fetus of 14 weeks and 3 days Source: Diagnose Clínica de Ultrassom



Figure 3: Obstetric ultrasound showing a dead fetus, signs of fetal resorption, and biometrics compatible with 12 weeks and 1 day Source: Diagnose Clínica de Ultrassom

The pregnancy was followed up with routine prenatal exams, including serologies, all of which were normal, as well as monthly Doppler ultrasound exams, which showed a normal pattern for the surviving fetus, with no evidence of maternal or fetal risks, allowing for a total of 11 prenatal visits.

At 39 weeks of gestation, on August 22, 2023, an elective cesarean section was performed at Hospital São Silvestre in Goiânia-GO. The procedure was uneventful, with a good final outcome and a male newborn weighing 3590g with Apgar scores of 8/9. After the birth of the viable twin, the dead fetus, with a papyraceous, macerated appearance and undefined sex, was extracted and sent to the pathology department (Figure 4). The newborn and the mother progressed without complications and were discharged from the ward in 2 days.



Figure 4: Papyraceous, macerated fetus with placenta Source: Author's files

### DISCUSSION

The term "fetus papyraceus" is based on the analogy between the fetus and an inanimate substance, attributing to it a solid texture resembling the consistency of stone. This terminology is used to describe a degenerated ovum that does not progress beyond the early stages of embryogenesis. The phenomenon involves the fetal demise of one of the twins in early pregnancy, followed by the intrauterine retention of the deceased fetus for at least 10 weeks. Compression of the deceased fetus occurs due to the growth of the healthy fetus, resulting in a flat and thin appearance, similar to paper<sup>1-3</sup>.

This phenomenon is a rare event that occurs in 0.018-0.02% of multifetal pregnancies. It occurs more specifically in monochorionic twin pregnancies, resulting in the abortion of one of the fetuses. The cause is usually idiopathic; however, it may be related to twin-to-twin transfusion syndrome, which results from discrepancies in blood circulation between the fetuses, or to inadequate umbilical cord insertion, usually occurring in the center of the placental mass, as well as genetic and chromosomal anomalies. An anomalous variant known as velamentous insertion, characterized by blood vessels that connect to the membranes surrounding the placenta instead of inserting directly into its center. These factors can exacerbate fetal loss and contribute to congenital anomalies<sup>3-5</sup>.

In the context of twin pregnancies, we can predominantly categorize them into two types: monochorionic and dichorionic. Monochorionic pregnancies are notably associated with a significantly higher risk of malformations due to intense competition for nutrients and oxygen between fetuses sharing a single placenta. When this competition reaches severe levels, the risks of perinatal mortality and morbidity increase considerably. Among the most common complications in this clinical scenario is fetal death due to inadequate vascular connections in the shared placenta<sup>2,6</sup>.

The prognosis of a pregnancy after the death of one of the twins is influenced mainly by the gestational age at the time of fetal death and by chorionicity, regardless of amnionicity. When loss occurs in the first trimester, the death of one fetus does not seem to be associated with adverse effects on the development of the survivor, especially in diamniotic dichorionic pregnancies. In this case, patients may be asymptomatic or may present with abdominal pain and mild genital bleeding. However, the death of a single fetus after 14 weeks, and especially after the 20th week of gestation, is associated with adverse effects on the surviving fetus, with a higher risk of prematurity (spontaneous or iatrogenic), intrauterine growth restriction, neurological morbidity, preeclampsia, hemorrhage, and sepsis<sup>4</sup>.

The process of fetal death can lead to serious complications for the mother, such as complications during delivery, for example, dystocia. Additionally, fetal death can lead to disorders in intravascular coagulation due to the significant release of thromboplastin from the deceased fetal tissue. This released thromboplastin can enter the maternal circulation, resulting in changes in the coagulation cascade, a critical process in hemostasis<sup>1</sup>. In the described clinical case, there were no complications, most likely due to the gestational age of fetal death being below the 14th week of gestation and because it was a diamniotic dichorionic pregnancy.

Furthermore, it is important to highlight another point regarding the surviving twin. It is observed that the later the occurrence of twin fetal death, the greater the complications for both the surviving fetus and the mother. These complications include neural tube disorders, which can result in cerebral palsy, as well as cerebral hypoxia due to the diversion of blood from the healthy fetus to the non-surviving fetus through the placenta, causing ischemic brain damage. Additionally, other areas of the body may also be affected, such as the digestive system, with the occurrence of gastrointestinal tract atresia, and the renal system, with renal agenesis<sup>1, 2</sup>.

In dichorionic pregnancies, the management is for the pregnancy to continue until at least 38 weeks, as long as both maternal health and fetal well-being are assured, unless there is another obstetric indication for pregnancy termination. In the case of monochorionic pregnancies, conservative management is a viable option, especially before 34 weeks, due to the higher neonatal risks associated with prematurity. In this context, the administration of prenatal corticosteroids should be considered. The patient in question gave birth at 39 weeks through an elective cesarean section without complications, with the birth of a viable twin. For pregnancies managed conservatively, attention should be provided with equipment for monitoring fetal well-being through serial ultrasound scans to monitor fetal growth and amniotic fluid volume. The evaluation of fetal anemia through measurement of the maximum systolic velocity in the middle cerebral artery using Doppler ultrasound is an effective parameter for monitoring the fetal health. For maternal monitoring, blood coagulation tests are recommended.<sup>34</sup>.

Additionally, special attention should be given to blood pressure and proteinuria levels. This is due to the higher risk of hypertensive disorders associated with twin pregnancies, especially those in which one fetus dies<sup>3,4</sup>. In the case of the clinical case patient, complementary exams such as Doppler ultrasound were performed monthly to check the viability of the fetus, as well as laboratory tests (all without alterations) every trimester, as recommended by the Ministry of Health. These complications require careful medical monitoring and appropriate treatment to ensure the safety of both the mother and the healthy fetus being carried. It is essential for pregnant women to receive adequate medical care during pregnancy to monitor the health of the fetus and the mother and take preventive measures when necessary<sup>2</sup>.

#### CONCLUSION

The present case report illustrates a dichorionic and diamniotic twin pregnancy, with the occurrence of a fetus papyraceus, a rare event that occurs in a small percentage of multifetal pregnancies. A patient underwent specific prenatal monitoring, including regular ultrasounds and laboratory tests, which allowed for a comprehensive assessment of fetal and maternal well-being. Early detection of fetal death and appropriate management were crucial to ensuring the safety of both the mother and the surviving fetus. The performance of an elective cesarean section at 39 weeks of gestation resulted in a positive outcome, with the birth of a healthy newborn.

The case underscores the importance of careful monitoring and timely medical intervention in twin pregnancies, especially when complications such as fetus papyraceus occur. Understanding risk factors and implementing monitoring protocols are essential to ensuring a progressive outcome for both mother and child. Additionally, the report highlights the importance of knowledge and the ability to identify less common clinical conditions, such as fetus papyraceus, to ensure the delivery of high-quality healthcare and informed decision-making during twin pregnancies.

#### REFERENCES

- Correia L, Valdoleiros S, Marujo A, Silva A, Simões T. Fetus papyraceous: A rare cause of labor dystocia. Porto Acta Obstet Ginecol. 2014;8(2):189-92.
- Gadre S, Gangatirkar R. Fetus papyraceous in monochorionic diamniotic twins. J Obstet Gynaecol India. 2019;69:40-3.
- 3. Timilsina N, Tamrakar SP, Thapaliya S, Sachdeva C, Tamang A. Fetus papyraceous disguised as compound presentation: a case report. Ann Med Surg

[Internet]. 2022 Sep;81:104481. Available from: https://www.sciencedirect. com/science/article/pii/S2049080122012419 doi:10.1016/j.amsu.2022.104481

- Macêdo Maciel RA, Leal CRV, Fistarol M, Paiva SCP, Pedrosa MS, Maia MLP. Morte fetal e feto papiráceo em gestação gemelar: relato de caso. EMJ Repro Saúde. 2021;7(1):72-6.
- Matovelo D, Ndaboine E. Fetus papyraceus causing dystocia in a rural setting: a case report. J Med Case Rep. 2015 Aug 15;9:178.
- Al Riyami N, Al-Rusheidi A, Al-Khabori M. Perinatal outcome of monochorionic in comparison to dichorionic twin pregnancies. Oman Med J. 2013 May;28(3):173-7.

# **CASE REPORT**

# MINIMALLY INVASIVE CARDIAC SURGERY TO REPLACE TRANSCATHETER IMPLANTED AORTIC BIOPROSTHESIS AFTER EARLY DEGENERATION. CASE REPORT

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#### ABSTRACT

Surgical replacement of a transcatheter implanted aortic valve prosthesis (TAVR) that presented early degeneration (in less than 24 months after its implantation) does not commonly occur in a short period of time and may be related to a worse prognosis and complications. Minimally invasive cardiac surgery can minimize surgical stress in situations where the individual undergoing it is fragile. The purpose of this article is to review the topic and describe a case of a frail 84-year-old patient who presented with valve dysfunction after TAVR and underwent surgical replacement of the bioprosthesis, through minimally invasive surgery.

# KEYWORDS: POSTOPERATIVE COMPLICATIONS; AORTIC VALVE STENOSIS; THORACIC SURGERY; TRANSCATHETER AORTIC VALVE REPLACEMENT.

#### INTRODUCTION

Aortic stenosis is an abnormality characterized by narrowing of the aortic valve orifice, resulting in obstruction of the left ventricular outflow tract. The etiology is related to the age group of the affected population. In individuals over 70 years old, it is related to calcification, and in younger populations, it is related to the bicuspid valve. In terms of prevalence, it is present in 0.5% of the general population.<sup>1</sup>

As aortic stenosis treatment, surgical valve replacement (SVR) is the first-line therapy for patients with valve disease. The dysfunctional native valve is replaced with an artificial one, which can be mechanical or bioprosthetic. Additionally, transcatheter aortic valve replacement (TAVR) has emerged as a valid option for patients with severe symptomatic aortic stenosis who are appropriately selected. TAVR is an alternative to open surgery through sternotomy for patients with severe symptomatic aortic stenosis who are appropriately selected. Several randomized studies have established the superiority of TAVR for treating patients who present a prohibitive and high risk of surgical mortality and as a reasonable alternative for elderly patients with an intermediate risk of surgical mortality. <sup>1, 2, 3, 4</sup>

The durability of TAVR is less well-defined than SAVR.

In the literature, studies on the durability of TAVR extend only to five years, a period shorter than the expected time for deterioration of valves used in SAVR. The durability of TAVR is even less defined in the population of patients with bicuspid aortic valves (BAV). <sup>3</sup> Valve replacement can be with a mechanical or bioprosthetic prosthesis, with each option having advantages and disadvantages in terms of durability and anticoagulation, for example. The main disadvantage of a bioprosthesis is durability, related to structural valve degeneration (SVD), a condition that eventually requires reoperation for valve replacement, a major surgical intervention. The probability of SVD is very low in the first 10 years after valve replacement in the elderly, with a gradual increase in incidence after that period. <sup>2, 3, 5</sup>

In the clinical case reported in this study, the insertion of a bioprosthesis through TAVR was performed on a patient who was appropriately selected according to the criteria found in the literature. The patient presented degeneration of the aortic bioprosthesis in a significantly shorter period than previously reported in the literature. This case report was approved by the Research Ethics Committee of the Hospital de Urgências de Goiás under CAAE: 38630920.7.0000.0033.

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### CASE REPORT

Patient is an 84-year-old female, 1.43 m tall, weighing 55 kg, with a medical history of systemic arterial hypertension, aortic stenosis, left ventricular hypertrophy, diffuse atherosclerosis, pulmonary hypertension, peptic disease (gastritis), intestinal polyp, diverticular disease of the colon, hypothyroidism, neurogenic bladder, and difficult-to-control heart failure (pulmonary congestion and low cardiac output). She is taking Brasart HCT®, Selozok®, Trezor®, AAS®, Marevan®, Euthyrox®, Uniprost®, and Venaflon®. The patient underwent TAVR for implantation of an aortic valve bioprosthesis (Sapien S3® 20 mm) on 03/11/2021, due to severe and symptomatic aortic valve stenosis. On 22/06/2023, the patient was admitted to the emergency department with complaints of asthenia, malaise, abdominal discomfort, exertional dyspnea, upper gastrointestinal bleeding (melena), and normocytic, normochromic anemia without hemodynamic compromise, requiring transfusion. The patient had an elevated INR and Marevan® was suspended upon admission due to upper gastrointestinal bleeding.

The patient had a previous transthoracic echocardiogram performed on 23/05/23, with the following findings: aortic root diameter (Ao): 25 mm, left atrium (LA): 40 mm, right ventricular diameter (RV): 24 mm, left ventricular end-diastolic diameter (LVEDD): 43 mm, left ventricular end-systolic diameter (LVESD): 25 mm. Presence of a biological aortic endoprosthesis, with inadequate visualization of its leaflets on transthoracic examination, but presenting severe stenosis and mild to moderate intraprosthetic regurgitation (peak systolic gradient of 126 mmHg and mean of 78 mmHg, time to peak aortic flow acceleration (TA): 137 ms, TA/Left ventricular ejection time (LVET): 0.37, Doppler velocity index (DVI): 0.23, effective orifice area estimated by the continuity equation at 0.57 cm2, indexed at 0.40; aortic regurgitation parameters: vena contracta: 3.6 mm and half pressure decay time = 309 ms). Mild mitral and tricuspid valve regurgitation. High echocardiographic probability of pulmonary hypertension. On a transesophageal echocardiogram performed on 20/06/2023, the patient presented: Ao: 25 mm, LA: 36 mm, RV: 22 mm, LVEDD: 44 mm, LVESD: 27 mm. Doppler of the biological aortic prosthesis: mean gradient: 72 mmHg, peak gradient: 117 mmHg, peak velocity 5.4 m/s, DVI: 0.21, TA: 124 ms, estimated area 0.51 cm2. Turbulent flow in the right atrium, with peak velocity of 3.3 m/s, turbulent flow in the left ventricular outflow tract consistent with central aortic insufficiency. Moderate left ventricular diastolic dysfunction.

Once the dysfunction of the aortic valve bioprosthesis was evidenced - severe aortic stenosis and mild to moderate intraprosthetic regurgitation secondary to valve degeneration, associated with a history of difficult-to-control heart failure (pulmonary congestion and low cardiac output), the patient was evaluated by the cardiac surgery team and the hemodynamics team. Percutaneous treatment was considered unfeasible, and therefore, open aortic valve replacement surgery was indicated, with a minimally invasive approach (figures 01 and 02).

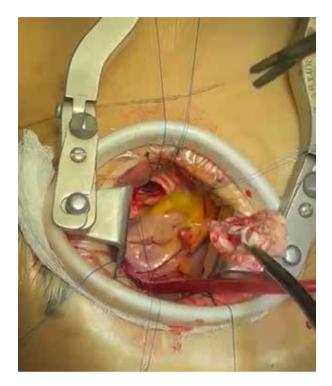


Figure 01. Minimally invasive surgery for removal of the anterior aortic valve bioprosthesis.



Figure 02. Appearance of the degenerated aortic valve bioprosthesis after its extraction.

The patient underwent successful cardiac surgery on 20/07/2023 for the implantation of aortic valve bioprosthesis no. 23 (Crown PRT®), enlargement of the aortic ring and aorta with bovine pericardium, through anterolateral/right paraesternal minithoracotomy, with no complications (figure 03).

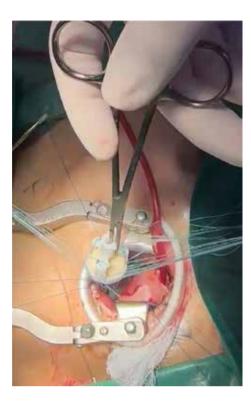


Figure 03. Implantation of the new aortic valve prosthesis through minimally invasive surgery.

The patient was monitored with a cardiac monitor, pulse oximeter, temperature probe, non-invasive blood pressure cuff, capnograph, gas analyzer, and Conox. A radial artery catheter was inserted for invasive blood pressure monitoring, a central venous catheter was placed in the subclavian vein, and a peripheral venous access was established with a 20-gauge catheter. A urinary catheter was also inserted. The patient underwent balanced general anesthesia, with intravenous induction and maintenance with an inhaled anesthetic. She was maintained on controlled mechanical ventilation in a closed circuit system with gas reabsorption.

The duration of cardiopulmonary bypass (CPB) was 156 minutes, and the aortic cross-clamp time was 119 minutes. The patient received a blood transfusion (3 units of packed red blood cells in the operating room and 8 units of cryoprecipitate). An autotransfusion system (Cell Saver) was used. A right-sided pleural effusion was identified, and thoracic drainage was performed. The patient was then transferred to the ICU while still intubated, on mechanical ventilation, hemodynamically stable, and receiving sodium nitroprusside (0.76 mcg/kg/minute). She remained stable, had effective awakening, and was successfully extubated without complications. She continued to be stable, with adjustment of antihypertensive medication, weaning off and discontinuation of sodium nitroprusside. The chest tube was removed. On 21/07/2023, she was discharged from the ICU.

A follow-up transthoracic echocardiogram on 22/07/23

showed a good surgical outcome: mild dilation of the left atrium (indexed volume = 41 ml/m2), normal dimensions of the left ventricle (LV) with preserved systolic function and moderate diastolic dysfunction. The biological prosthesis in the aortic position was functioning normally, without signs of stenosis (mean gradient = 6 mmHg, peak gradient = 11 mmHg), and no regurgitation. The mitral valve showed slight calcification of the annulus and mild regurgitation. Mild tricuspid regurgitation was also present. There was moderate dilation of the aortic root (48 mm, post-surgical enlarge ent).

#### DISCUSSION

Aortic stenosis is characterized by the narrowing of the aortic valve, which obstructs the blood flow from the left ventricle of the heart to the aorta. Symptoms of aortic stenosis include fatigue, heart murmur, chest pain or tightness, heart palpitations, shortness of breath, and feeling faint or dizzy with exertion. Complications of aortic stenosis can include heart failure, stroke, blood clots, endocarditis, and sudden death. <sup>6</sup>

In terms of pathophysiology, aortic stenosis appears to be mediated by an inflammatory process, similar to atherosclerosis, and calcification deposition can occur in the final stage of the scarring process, similar to coronary atheroma. As a result of the deposition and thickening of the valve, obstruction of the left ventricular outflow tract occurs, leading to wall hypertrophy. The myocardium becomes less compliant due to the increased diastolic pressure in the left ventricle and impaired relaxation. Pre-syncope and syncope can occur in situations of high cardiac demand, vasodilation, and arrhythmia. In severe cases, angina can occur due to increased left ventricular mass, poor coronary filling, and reduced coronary flow reserve. The risk of sudden cardiac death is proportional to the severity of the disease. <sup>1,6</sup>

The use of bioprosthetic valves has been steadily increasing in the last decade, surpassing mechanical valves, with aortic valve prostheses being the most common. This trend is likely multifactorial, explained by the better hemodynamic performance of aortic bioprostheses, patient lifestyle preferences, the absence of a need for prolonged systemic anticoagulation, and the aging of the target population. <sup>3, 4, 5</sup> TAVI is a minimally invasive procedure in which a prosthetic valve, which will replace the damaged valve, is inserted through a catheter via access routes such as the femoral artery, subclavian artery, or common carotid artery. <sup>6,7</sup>

The guidelines for reporting the outcomes of bioprosthetic valves classify the related factors affecting durability and promote valve dysfunction into BVD and non-BVD. The pathophysiology corresponds to calcific degeneration as a result of repetitive mechanical stresses. There is significant variability in the definition of BVD, still lacking a universal definition. <sup>3, 4</sup> Non-BVD refers to secondary processes involving the valve, such as patient-prosthesis mismatch, valve leaflet thrombosis, endocarditis, pannus ingrowth, or paravalvular leak. BVD and non-BVD are not exclusive processes. Non-BVD mechanisms, such as patient-prosthesis mismatch, leaflet thrombosis, and paravalvular regurgitation, have been associated with accelerated BVD due to valvular hemodynamic alteration and mechanical stress. <sup>4</sup>

Bioprosthetic Valve Degeneration (BVD) is defined as the degeneration or intrinsic dysfunction of prosthetic valve materials. Previous studies have defined BVD as the need for reoperation due to the absence of careful and regular echocardiographic follow-up, but they do not provide specific criteria to define BVD and/or the indication for reoperation. Based on changes in transprosthetic gradients and the severity of regurgitation on echocardiography, the term "hemodynamic deterioration of the valve" has been introduced. There are several proposed definitions for bioprosthetic valve degeneration according to echocardiographic criteria, including a progression of the transprosthetic aortic gradient, leading to a mean gradient of  $\geq$ 30 mm Hg associated with a reduced effective orifice area to  $\leq$ 1 cm2 or intraprosthetic aortic insufficiency grade  $\geq$ 3.<sup>2-4</sup>

There are several limitations regarding the assessment of transcatheter valve durability and the incidence of BVD in the surgical literature. This is because the absence of valve reintervention is a common clinical outcome, which underestimates the true incidence of BVD, as reoperation may not be offered to all patients and some may die before echocardiographic detection of BVD.<sup>3,4</sup>

A patient reported in this article underwent SAVR via minimally invasive surgery. Minimally invasive surgery has proven to be an excellent option for treating atrioventricular valve diseases. Surgeons specifically trained in this approach have achieved excellent results compared to those obtained by sternotomy, but with some advantages over conventional techniques such as: better pain control, shorter hospital stay, faster recovery time, less need for blood transfusion, lower rates of perioperative infection, less need for imaging and laboratory tests, lower rate of readmission in the first year postoperatively, better aesthetic outcome, and lower overall cost. <sup>8,9-11</sup>

#### REFERENCES

- Clayton B, Morgan-Hughes G, Roobottom C. Transcatheter aortic valve insertion (TAVI): a review. Br J Radiol. 2014;87(1033):20130595. doi:10.1259/ bjr.20130595
- Rodriguez-Gabella T, Voisine P, Puri R, Pibarot P, Rodés-Cabau J. Aortic Bioprosthetic Valve Durability: Incidence, Mechanisms, Predictors, and Management of Surgical and Transcatheter Valve Degeneration. J Am Coll Cardiol. 2017;70(8):1013-1028. doi:10.1016/j.jacc.2017.07.715
- Kataruka A, Otto CM. Valve durability after transcatheter aortic valve implantation. J Thorac Dis. 2018;10(Suppl 30):S3629-S3636. doi:10.21037/ itd.2018.07.38
- Mack MJ, Leon MB, Thourani VH, Pibarot P, Hahn RT, Genereux P, Kodali SK, Kapadia SR, Cohen DJ, Pocock SJ, Lu M, White R, Szerlip M, Ternacle J, Malaisrie SC, Herrmann HC, Szeto WY, Russo MJ, Babaliaros V, Smith

CR, Blanke P, Webb JG, Makkar R; PARTNER 3 Investigators. Transcatheter Aortic-Valve Replacement in Low-Risk Patients at Five Years. N Engl J Med. 2023 Nov 23;389(21):1949-1960.

- DOI: 10.1056/NEJMoa2307447
- Kostyunin AE, Yuzhalin AE, Rezvova MA, Ovcharenko EA, Glushkova TV, Kutikhin AG. Degeneration of Bioprosthetic Heart Valves: Update 2020. J Am Heart Assoc. 2020 Oct 20;9(19):e018506. doi: 10.1161/JAHA.120.018506. Epub 2020 Sep 21. PMID: 32954917; PMCID: PMC7792365.
- Young C, Horton J. Transcatheter Aortic Valve Implantation for Patients With Severe Symptomatic Aortic Stenosis [Internet]. Ottawa (ON): Canadian Agency for Drugs and Technologies in Health; 2021 Jul. PMID: 36173871. https://doi.org/10.2217/fca-2017-0056
- Mariathas M, Rawlins J, Curzen N. Transcatheter aortic valve implantation: where are we now? Future Cardiol. 2017 Nov;13(6):551-566. doi: 10.2217/fca-2017-0056. Epub 2017 Oct 24. PMID: 29064293.
- Ko K, de Kroon TL, Post MC, Kelder JC, Schut KF, Saouti N, van Putte BP. Minimally invasive mitral valve surgery: a systematic safety analysis. Open Heart. 2020 Oct;7(2):e001393. doi: 10.1136/openhrt-2020-001393.
- Vervoort D, Nguyen DH, Nguyen TC. When Culture Dictates Practice: Adoption of Minimally Invasive Mitral Valve Surgery. Innovations (Phila). 2020 Sep/ Oct;15(5):406-409. doi: 10.1177/1556984520948644.
- Paparella D, Fattouch K, Moscarelli M, Santarpino G, Nasso G, Guida P, Margari V, Martinelli L, Coppola R, Albertini A, Del Giglio M, Gregorini R, Speziale G. Current trends in mitral valve surgery: A multicenter national comparison between full-sternotomy and minimally-invasive approach. Int J Cardiol. 2020 May 1;306:147-151. doi: 10.1016/j.ijcard.2019.11.137.
- Nakayama T, Nakamura Y, Kanamori K, Hirano T, Kuroda M, Nishijima S, Ito Y, Tsuruta R, Hori T. Early and midterm results of minimally invasive aortic and mitral valve surgery via right mini-thoracotomy. J Card Surg. 2020 Jan;35(1):35-39. doi: 10.1111/jocs.14313.

# **REVIEW ARTICLE**

# CHALLENGES IN THE EMERGENCY: MANAGEMENT OF ANORECTAL ABSCESS IN CRYPTOGLANDULAR DISEASE: LITERATURE REVIEW

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#### ABSTRACT

This scientific article addresses cryptoglandular disease, with a specific focus on anorectal abscess, a common and potentially serious condition that requires urgent attention and appropriate medical intervention. Cryptoglandular disease refers to an infectious process that develops in the anorectal glands, generally resulting in an abscess and, if left untreated, a fistula. Anorectal abscess is an acute complication of cryptoglandular disease, described by inflammation and accumulation of pus in the tissues around the anus and rectum. This article highlights the importance of early recognition of the signs and symptoms of anorectal abscess, which include intense, severe pain, swelling, and fever. Initial treatment usually consists of surgical abscess surgery, which can be performed on an outpatient basis or in a hospital setting, depending on the severity and extent of the infection. The importance of appropriate post-drainage management, including local wound care, specific analgesia and possibly antibiotic therapy, especially in complicated cases, is emphasized. Furthermore, the article highlights the need for careful monitoring to avoid long-term complications such as the formation of anorectal fistulas. The article in question is used from the Theoretical Reference, through a bibliographical review on the subject, collecting as much data as possible, from a general perspective, research on the topic was carried out through studies of articles, literary works and an analysis of research and facts. Finally, the work provides a comprehensive overview of cryptoglandular disease in the emergency department, with a focus on anorectal abscess, highlighting the importance of early identification, accurate diagnosis and appropriate management to improve clinical outcomes and reduce the risk of complications.

## KEYWORDS: CRYPTOGRANDULAR DISEASE; TREATMENT; CONSEQUENCES; PREVENTION.

### INTRODUCTION

Cryptoglandular disease and anorectal abscess represent one of the most common and challenging conditions encountered in proctological and surgical clinical practice. This condition, although often underestimated, can cause significant discomfort and compromise patients' quality of life, in addition to potential serious complications if not treated appropriately. Cryptoglandular disease is characterized by an infection of the anorectal glands located close to the anal canal, triggering an inflammatory process that can culminate in the formation of anorectal abscesses.

These abscesses, when not diagnosed early and treated effectively, can evolve into anorectal fistulas, further complicating the clinical picture and therapeutic management. Management of cryptoglandular disease and anorectal abscess in medical emergencies requires a comprehensive understanding of the underlying pathophysiology, characteristic signs and symptoms, and available diagnostic and therapeutic strategies.

Early identification, adequate intervention and meticulous monitoring are essential to minimize complications, prevent recurrences and promote complete recovery of patients. In this context, this article proposes to explore in detail cryptoglandular disease in emergency care, with a special focus on anorectal abscess. Relevant aspects of the definition, pathophysiology, clinical presentation, diagnosis, treatment and prognosis of this condition will be discussed, as well as strategies to optimize the management of these challenging cases in contemporary clinical practice <sup>1</sup>.

By offering a comprehensive and up-to-date analysis, this article aims to provide valuable insights to healthcare professionals involved in the care of patients with cryptoglandular disease in the emergency department.

# DEFINITION AND PATHOPHYSIOLOGY OF CRYPTO-GLANDULAR DISEASE

Cryptoglandular disease, specifically related to anorectal abscess, is a condition characterized by inflammation of the anorectal glands located in the region of the anus and rectum. These glands produce mucus to lubricate the anal canal during the passage of feces. When an obstruction or infection of these glands occurs, an anorectal abscess can

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develop. Cryptoglandular disease refers to infection of one or more of the anorectal glands <sup>2</sup>.

The word "crypt" comes from Greek and means "hidden" or "inaccessible," indicating that these glands are located in the innermost part of the anal canal, near the anus, and can be difficult to access directly. The pathophysiology of cryptoglandular disease and anorectal abscess involves a sequential process: Obstruction of the Anorectal Glands, which is often the first step in pathogenesis. This can occur due to various reasons, including chronic constipation, local trauma, increased intra-rectal pressure, or local irritation.

Bacterial Overgrowth: When the glands are obstructed, the mucous produced cannot be secreted normally to the surface of the anal canal. This creates an environment conducive to bacterial overgrowth within the obstructed glands. Inflammation and Abscess Formation: The presence of bacteria within the obstructed glands triggers a local inflammatory response. This leads to the formation of an anorectal abscess, which is a collection of pus and inflammatory tissue around the infected anorectal glands <sup>3</sup>.

Clinical Presentation: An anorectal abscess can present with symptoms such as severe pain, swelling, localized redness around the anus, and fever, depending on the extent and severity of the infection. Progression to Fistula (in some cases): If the anorectal abscess is not properly treated, there is a risk of developing an anal fistula. The fistula is an abnormal connection between the anal canal and the skin around the anus, which can lead to additional complications and recurrences.

The process of forming an anorectal abscess is often rapid and can result in acute and significant symptoms for the patient. Treatment typically involves surgical drainage of the abscess to relieve pressure, remove purulent material, and facilitate healing of the affected tissues. If not treated properly, the abscess can progress to more serious complications, such as the formation of anorectal fistulas. Therefore, early and appropriate intervention is essential to avoid complications and promote patient recovery <sup>4</sup>.

# EPIDEMIOLOGY AND RISK FACTORS

The epidemiology of cryptoglandular disease and anorectal abscess reveals a relatively common condition, although the exact incidence may vary according to different populations and studies. The prevalence of cryptoglandular disease and anorectal abscess varies in different population groups, but it is estimated to affect a significant portion of the general population. Epidemiological studies suggest that anorectal abscess is more common in young and middle-aged adults, although it can occur at any age.

The exact prevalence may be underestimated due to patients' reluctance to seek treatment due to the delicate nature of the condition. As for Risk Factors, Chronic constipation can lead to excessive straining during bowel movements, increasing the risk of trauma and obstruction of the anal glands. Chronic diarrhea, if frequent, can result in irritation and inflammation of the lining of the anal canal, predisposing to infection <sup>5</sup>.

Inflammatory bowel diseases, such as Crohn's disease and ulcerative colitis, can increase the risk of developing anorectal abscesses due to chronic inflammation in the gastrointestinal tract. Although the condition can affect both sexes, men appear to have a slightly higher incidence of anorectal abscesses. Traditionally, cultures taken from an inflamed site are usually obtained to determine the appropriate antibiotic treatment, which is not the case for most anorectal abscesses.

Diabetes mellitus can compromise immune function, making patients more susceptible to infections, including anorectal abscesses. Finally, obesity is associated with increased intra-abdominal pressure, which can contribute to obstruction of the anorectal glands <sup>6</sup>.

These risk factors can increase the likelihood of developing anorectal abscesses in the general population. However, it is important to recognize that cryptoglandular disease and anorectal abscess can occur in individuals without apparent risk factors, highlighting the importance of awareness, prevention, and proper management of this condition in clinical practice.

# SYMPTOMS AND DIAGNOSIS

The clinical presentation of anorectal abscess is a crucial aspect to understand in clinical practice, as it guides the diagnosis and effective management of this condition. Anorectal abscess is an acute complication of cryptoglandular disease, resulting from infection of the anal glands and subsequent formation of pus and inflammatory tissue around the anus and rectum <sup>7</sup>. The clinical presentation of anorectal abscess can vary in terms of severity, location, and extent of infection, but generally includes the following signs and symptoms:

• Intense Anal Pain: The pain is often described as sharp, stabbing, and intense. Patients report pain around the anus, which may worsen with movement, sitting, defecation, or even with light touch to the affected area.

• Swelling and Tenderness: There is often visible swelling and tenderness around the anus. The swelling may be palpable and may be associated with localized warmth.

• Localized Redness and Heat: The affected region may experience redness and increased temperature due to underlying inflammation. These signs indicate the body's inflammatory response to the infection.

• Pus Secretion: In more advanced cases, there may be pus secretion from the anus. The presence of pus may be evident in the patient's underwear or during anal hygiene.

Fever and Malaise: In more severe cases, the pa-

tient may present a low to moderate fever due to the systemic inflammatory response. General malaise, fatigue and loss of appetite may also be present in some cases.

• Difficulty Defecating: Severe pain around the anus can make it difficult to pass stool, leading to discomfort and pain during defecation.

It is essential to recognize the characteristic signs and symptoms of anorectal abscess for early diagnosis and effective treatment. A detailed clinical evaluation, including medical history, physical examination, and possibly additional tests such as ultrasound or magnetic resonance imaging, may be necessary to confirm the diagnosis and determine the extent of the infection <sup>8</sup>.

In the physical examination, anal inspection shows ervthema, edema, and often a fluctuant area when the infection is more superficial. When the infection is deep, no signs are evident. In these cases, digital rectal examination, when possible, or imaging studies are important aids in diagnosis. Anorectal magnetic resonance imaging has 95% sensitivity, 80% specificity, and 97% positive predictive value for the diagnosis of abscesses and fistulas, especially chronic and complex ones. Transperineal ultrasound can be extremely useful for detecting abscesses, with many authors considering it as a first-line imaging study due to its non-invasive nature, low cost, and availability, although this method has not gained widespread popularity. Digital rectal examination and anoscopy are often not performed due to the patient's intense pain. When the diagnosis is uncertain, an examination under anesthesia is necessary (Botelho, A., & Carlos, L. 2023).

The differentiation between anorectal abscess and other anorectal conditions may require the use of imaging tests such as ultrasound, computed tomography, or magnetic resonance imaging to assist in diagnosis and planning appropriate treatment. Early recognition and effective management of anorectal abscess are essential to alleviate the patient's symptoms, prevent complications, and promote full recovery.

### INITIAL TREATMENT

The initial treatment of anorectal abscess plays a crucial role in the effective management of this painful and potentially debilitating condition. Understanding the available clinical and surgical approaches is essential for relieving the patient's pain, preventing complications, and promoting proper recovery. Clinical strategies begin with Analgesia, pain relief is a priority in the initial treatment of anorectal abscess, oral analgesics such as paracetamol or nonsteroidal anti-inflammatory drugs (NSAIDs) are often prescribed to control acute pain and improve patient comfort <sup>9</sup>.

Antibiotic therapy, considering that antimicrobial therapy may be indicated in certain cases, especially if there are signs of perianal cellulitis or evidence of systemic spread of infection. Broad-spectrum antibiotics, such as ciprofloxacin or metronidazole, may be prescribed empirically until culture results and sensitivity tests are available.

Furthermore, local care where adequate local hygiene plays an important role in preventing bacterial contamination and promoting wound healing. Warm sitz baths or moist compresses can help relieve discomfort and promote drainage of the abscess. In the background, surgical strategies, which begin with Incisional Drainage, are often necessary for anorectal abscesses that do not respond to conservative treatment or that show signs of complications, such as perianal cellulitis <sup>10</sup>.

The procedure involves making an incision in the affected area to allow drainage of the accumulated pus and facilitate healing. In some selected cases, Percutaneous Drainage, guided by ultrasound or computed tomography, may be an option for deeply located abscesses or in patients with multiple comorbidities who are not surgical candidates. During anorectal abscess drainage, it is important to assess for associated fistulas. Early identification and appropriate treatment of fistulas are crucial to prevent recurrence and long-term complications.

In immunocompromised patients, the elderly, or those with multiple comorbidities, the therapeutic approach may need to be adapted to meet the individual needs of the patient. Careful monitoring of the patient after initial treatment is essential to monitor treatment response, detect complications early, and adjust therapy as necessary. In conclusion, the initial treatment of anorectal abscess requires a comprehensive approach that incorporates clinical and surgical strategies to relieve patient symptoms and promote effective recovery.

# FOLLOW-UP AND PROGNOSIS

After initial treatment, it is essential to schedule follow-up appointments to assess wound healing, symptom resolution, and the presence of complications. During the post-treatment evaluation, the physician conducts a detailed physical examination and discusses any concerns or persistent symptoms reported by the patient. The patient should be instructed to monitor their symptoms, such as pain, swelling, discharge, or rectal bleeding, and report any changes to the physician immediately.

Monitoring symptoms is essential for detecting abscess recurrence, fistula formation, or additional complications. In some cases, additional tests such as ultrasound, computed tomography, or magnetic resonance imaging may be necessary to assess the extent of healing, identify residual fistulas, or diagnose hidden complications. The choice of follow-up tests depends on the individual clinical evaluation of the patient and suspicion of complications.

Regular follow-up allows for early detection of abscess recurrence, fistula formation, or complications such as perianal cellulitis. Early intervention is essential to prevent serious complications and ensure long-term treatment success. The prognosis for anorectal abscess treatment is generally good, especially with timely diagnosis and treatment.

Still, most patients experience significant symptom relief after initial treatment and achieve complete wound healing. However, the prognosis can vary depending on factors such as the extent of the infection, presence of fistulas, underlying medical conditions, and adherence to the treatment plan.

### FINAL CONSIDERATIONS

Cryptoglandular disease, especially when manifested in the form of anorectal abscess, can represent a significant source of physical, emotional, and social discomfort for patients. In addition to the intense pain and anal dysfunction associated with the abscess, the possibility of recurrences and complications, such as the formation of anorectal fistulas, can profoundly impact the quality of life and mental health of affected individuals.

In this context, a holistic and patient-centered approach is crucial, which not only treats the acute symptoms of the abscess but also takes into account individual risk factors, underlying conditions, and psychosocial aspects that can influence the patient's experience and the course of the disease. Patient education plays a fundamental role in empowering individuals to understand their condition, recognize signs of recurrence and complications, and adopt preventive measures to minimize the risk of future episodes.

This may include guidance on proper anal hygiene, healthy diet and lifestyle, and strategies for coping with the stress and anxiety associated with the disease. Additionally, psychosocial support and ongoing follow-up by the medical team are essential to provide emotional support, clarify doubts and concerns, and promote the patient's active engagement in their own care.

As we advance in understanding cryptoglandular disease and its clinical implications, it is essential for healthcare professionals to continue to seek innovative, evidence-based approaches for the diagnosis, treatment, and follow-up of this condition. This may involve the development of new drainage techniques, adjuvant therapies, and strategies for preventing recurrences, as well as promoting greater public awareness about the condition.

Ultimately, by adopting a comprehensive and compassionate approach to managing cryptoglandular disease, we can help patients achieve a better quality of life, minimize the impact of the condition on their well-being, and promote complete and lasting recovery.

#### REFERENCES

- Assis, L. M. de, Tafner, P. F. D. A., Formiga, F. B., Pompeu, B. F., & Paes Leme, L. F. (2022). Tratamento cirúrgico dos abscessos anorretais em hospital de urgência e emergência. Journal of Coloproctology, 42(S 01), S1-S219. DOI: 10.1055/s-0043-1764605. [Licença CC BY-NC-ND 4.0];
- Ansari, P. (2023, janeiro). Abscesso anorretal. Em Manual MSD Versão saúde para a família. Hofstra Northwell-Lenox Hill Hospital, New York;
- 3. Nadal SR, Manzione CR, Horta SHC, Galvão VM. Comparação das doenças

perianais nos doentes HIV positivos antes e depois da introdução dos inibidores da protease. Revista Brasileira de Coloproctologia. 2001;21(1):5-8;

- MAGALHÃES, Maria Iranilda Silva et al. DOENÇA INFLAMATÓRIA IN-TESTINAL: ABORDAGEM E TRATAMENTO. Revista Ibero-Americana de Humanidades, Ciências e Educação, [S. I.], v. 9, n. 4, p. 941–950, 2023. DOI: 10.51891/rease.v9i4.9280. Disponível em: https://periodicorease.pro.br/ rease/article/view/9280. Acesso em: 5 fev. 2024;
- Raman SP, Chen Y, Fishman EK. Evolution of imaging in rectal cancer: multimodality imaging with MDCT, MRI, and PET. J Gastrointest Oncol. 2015 Apr;6(2):172-84. doi: 10.3978/j.issn.2078- 6891.2014.108;
- Araújo, S. E. A., Marcente, M. T., Mendes, C. R. S., Bertoncini, A. B., Seid, V. E., Horcel, L. A., Perez, R. O., & Klajner, S. (2017). Ligadura interesfincteriana do trato fistuloso (LIFT) para fístulas anais: uma experiência brasileira bi-institucional [Interesfincterial ligation of fistula tract (LIFT) for patients with anal fistulas: a Brazilian bi-institutional experience]. ABCD. Arquivos Brasileiros de Cirurgia Digestiva (São Paulo), 30(4), 235-238. DOI: 10.1590/0102-6720201700040002;
- Garcia-Aguilar J, Patil S, Kim JK, Yuval JB, Thompson H, Verheij F, et al. Preliminary results of the organ preservation of rectal adenocarcinoma (OPRA) trial. J Clin Oncol. 2020 [citado em 2022 Out 12]; 38(suppl 15):4008. Disponível em: https://ascopubs.org/doi/10.1200/JCO.2020.38.15\_ suppl.4008;
- Gotfried, J. (2022, janeiro). Diarreia em adultos. Lewis Katz School of Medicine at Temple University. Em Manual Merck (MSD). Recuperado de https:// www.msdmanuals.com/pt-br/casa/dist%C3%BArbios-digestivos/sintomas-de-dist%C3%BArbios-digestivos/diarreia-em-adultos;
- Botelho, A., & Carlos, L. (2023). Rotinas em Coloproctologia. Editora Unoesc. Disponível em: https://galeria.unoesc.edu.br/Nunca%20apagar/Rotinas\_ em\_coloproctologia.pdf;
- Nelson RL, Prasad L, Abcarian H. Anal carcinoma presenting as perirectal abscess or fistula. Arch Surg 1985; 120:632–635;

# **REVIEW ARTICLE**

# BURNOUT SYNDROME IN DOCTORS, A NEGLECTED PROBLEM

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#### ABSTRACT

The individual's relationship with his work and experiences in the work environment results in satisfaction and personal improvement and, at the same time, can be a source of stress and professional exhaustion. Burnout syndrome (BS) is a psychological syndrome that arises from an ongoing response to chronic interpersonal stressors at work. Professions that require a high level of stress on a daily basis are more susceptible to BS, especially in healthcare professionals. The prevalence of this syndrome in medical specialties is well documented and shows impressive rates. It is a public health problem that can lead to medical leave, increasing the costs to the employers, in addition to affecting the quality of the service offered with potentially serious consequences for caregivers, patients and health institutions, including the risk of medical malpractice, depression, and adverse effects on patient safety. The long-term effects of BS on healthcare workers, especially after the COVID-19 pandemic, must be a significant concern for governments, medical entities, hospitals and physicians themselves. Addressing medical burnout should be seen as a shared responsibility between health systems, organizations, institutions and individual physicians.

#### KEYWORDS: BURNOUT SYNDROME; QUALITY OF LIFE: MEDICAL RESIDENCY.

#### INTRODUCTION

The individual's relationship with their work and the experiences lived in the work environment result in satisfaction and personal improvement, and at the same time, can be a source of stress <sup>1</sup>. In the 1970s, some doctors working in free public healthcare began to speak of a concept of professional burnout. The term "burn-out," from the English "to burn oneself out, to consume oneself," was first used in 1974 by the psychoanalyst Herbert Freudenberger<sup>2</sup> when he observed that his work did not bring him the same pleasure as before, linking the feeling of burnout to the lack of stimulation originating from a scarcity of emotional energy. In addition to these symptoms, Freudenberger included fatigue, depression, irritation, and inflexibility as belonging to the symptomatic framework of Burnout Syndrome (BS). In the 1980s, Christina Maslach and Susan Jackson placed burnout in the perspective of intense and continuous stress caused by work <sup>3</sup>, In 1999, Christina Maslach and Michael Leiter gave BS its final definition and characterization: a syndrome composed of the three pillars of emotional exhaustion, depersonalization, and lack of professional achievement 4.

The Burnout Syndrome (BS) is a psychological syndrome that arises from a continuous response to chronic interpersonal stressors during work <sup>5</sup>. According to Maslach <sup>3</sup>, burnout begins when "energy turns into exhaustion, involvement turns into cynicism, and effectiveness turns into inefficiency."

The prevalence of this syndrome in medical specialties is well documented and presents impressive rates <sup>6,</sup> <sup>7</sup>. Its prevalence in studies varies from 14.7% to 76.0% <sup>8,9</sup>. The etiology is complex and multifactorial, some intrinsic - for example, perfectionism, altruism, or excessive empathy - and others extrinsic - largely beyond the individual's control, such as organizational culture, balance between personal and professional life, and catastrophic events like the global pandemic <sup>7</sup>.

#### Burnout syndrome in doctors.

Professions that require a high level of daily stress are more susceptible to BS, especially in healthcare professionals <sup>10</sup>. Specifically in the field of Medicine, the presence of this syndrome is critical. Doctors need to navigate skillfully in a medical minefield with the acuity of a chess champion, the hands of a concert violinist, the patience of a monk, the communication skills of a diplomat, and the endurance of a marathon runner <sup>11</sup>.

BS is a public health issue that can lead to absenteeism and sick leave, generating expenses for the employing organization, as well as affecting the quality of service offered, productivity, and profit <sup>12</sup>. Worldwide, it affects 1 in every 2 doctors; one-third of them are significantly affected, and one-tenth experience severe effects

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that are irreversible <sup>13, 14</sup>. According to the Brazilian Federal Council of Medicine, 23.1% of doctors in a sample of 7,700 professionals from all states experienced high levels of burnout <sup>15</sup>. The Covid-19 pandemic may have worsened this problem <sup>7, 11</sup>. In the US, nearly 40% of doctors exhibited at least one symptom of burnout in 2020, and another 40% say they are too overloaded with work to seek help <sup>11</sup>.

High-quality medical care has a significant impact on the ability to maintain or return to a healthy life, while clinical lapses in treatment can lead to frustrating or even catastrophic outcomes <sup>6, 11</sup>. The consequences of burnout are potentially serious for caregivers, patients, and healthcare institutions, and include the risk of medical errors, depression, and adverse effects on patient safety <sup>16</sup>.

A systematic review, including 20 articles, suggests that BS affects primary healthcare professionals (mainly nurses, but also doctors and pharmacists) and leads to high job stress, intense time pressure and workload, as well as lack of organizational support <sup>17</sup>. Another systematic review shows that cardiovascular diseases, musculoskeletal pain, depressive symptoms, psychotropic and antidepressant treatment, job dissatisfaction, and absenteeism are consistent effects of BS. Conflicting findings were observed for headache and insomnia. The individual and social impacts of burnout highlight the need for preventive interventions and early identification of this health condition in the workplace <sup>18</sup>.

Cross-sectional studies have associated physician burnout with suboptimal patient care practices <sup>6</sup>, as well as doubling the risk of medical error <sup>19</sup> and being associated with a 17% higher likelihood of being involved in a malpractice lawsuit 20. The longitudinal study Internal Medicine Resident Well-Being (IMWELL) found that higher levels of burnout were associated with greater odds of reporting a major medical error in the subsequent 3 months. Self-perceived serious medical errors were also associated with worsening burnout, depressive symptoms, and decreased quality of life, suggesting a bidirectional relationship between medical errors and distress <sup>21</sup>. Increasing levels of emotional exhaustion among physicians working in intensive care units are associated with higher standardized patient mortality rates and lower perceived quality of interpersonal teamwork 6, 22,

The syndrome also affects patient satisfaction and, along with personal suffering, has been linked to self-reported suboptimal care practices among residents in various medical specialties <sup>8, 18</sup>. Cross-sectional studies report significant correlations between physician burnout and job satisfaction <sup>23, 24</sup> and patient satisfaction with their hospital care <sup>25</sup>. This suggests a potential impact of burnout on patient satisfaction and physician-patient relationships, with concomitant effects on health outcomes <sup>6</sup>. Burnout syndrome and medical residency.

Medical residency is a form of postgraduate education for physicians, in the form of a specialization course, characterized by on-the-job training in a full-time dedication regime. It was instituted by the federal government in September 1977 with Decree No. 80,281. Although considered the "gold standard" of medical specialization, it is also the most difficult and demanding, both physically and emotionally, part of the specialization process <sup>26, 27</sup>, characterized by a high level of stress since the training physician is under constant pressure, experiencing fatigue, exhaustion, and fear of making mistakes <sup>28</sup>.

During this period, marked by extensive personal development, the traditional long working hours, night shifts, and psychological pressure to apply the knowledge acquired in undergraduate studies make the routine of young doctors very exhausting. This demands a change in lifestyle, which often leads to daytime sleepiness, resulting in physical and mental health impairment of the physician and, ultimately, also affecting their quality of life <sup>27</sup>.

Medical residency, due to its risk factors - intense workload, sleep deprivation, daily contact with severely ill patients, social isolation, financial concerns, lack of adequate working/learning conditions - seems to be the peak moment of distress among physicians and becomes highly conducive to the development of BS <sup>26</sup>. However, in addition to BS, depressive symptoms and recent suicidal ideation are also common in the early years of medical practice <sup>29</sup>. These issues not only predispose these professionals to illness but also increase the risk of suicide.

An initial attempt to systematize the psychological and psychiatric morbidity in residents related to their activity was made by Small <sup>30</sup>, who described a syndromic picture in training physicians, which he called the resident stress syndrome. According to this author, the syndrome presents the following characteristics: episodic cognitive disturbances, chronic anger, skepticism, family discord, abusive drug use, depression, suicidal ideation and behavior <sup>30</sup>. Still, in BS, which also considers other factors such as the individual's social and private life, the higher observed incidence of suicide-related phenomena may be influenced by the latter <sup>26, 31</sup>. In the study by Soares et al. <sup>26</sup>, the majority of individuals who reported having had suicidal thoughts also presented factors related to burnout, indicating a probable correlation between the two events <sup>26</sup>.

### Prevention measures.

According to Grosse <sup>11</sup>, in the USA, almost half of physicians are throwing up their hands and saying enough is enough. This is likely a major reason why 70% of physicians are not recommending their profession to their own children, which, in turn, makes it harder to address the projected shortage of 124,000 physicians in the US in the next decade <sup>11</sup>. The negative effects of BS on the well-being of healthcare professionals and its consequent implications for patient care highlight the urgent need for interventions aimed at this population. These interventions should aim to reduce levels of occupational stress, increase self-esteem, promote self-care, and create a healthy work environment <sup>32</sup>. Individual, organizational, and combined interventions can be implemented to prevent it by reducing occupational stress <sup>32</sup>.

Individual interventions are focused on learning adaptive coping strategies in the face of stressful agents <sup>32</sup>. A healthy diet, regular exercise, and good sleep hygiene are vital starting points. Time off work to recharge is also important <sup>33</sup>. Training in coping behavioral and cognitive skills, meditation, physical activity <sup>34</sup>, as well as self-care practices such as ensuring adequate rest, balancing work and other life dimensions, and engaging in a hobby <sup>35, 36</sup>. Creating balance in your life is essential, especially the balance between home and work life <sup>33</sup>.

There are anonymous online tools that allow individual doctors to privately assess their level of burnout <sup>37, 38</sup>. These tools have been shown to stimulate reflection and potential action steps to address burnout in large groups of physicians <sup>38</sup>.

Organizational strategies have proven more challenging to study so far <sup>6</sup>. These refer to modifying the situation in which work activities are carried out, aiming to improve communication and teamwork. They include training employees, restructuring tasks, and changing physical-environmental conditions, such as flexible hours, participation in decision-making, career planning, and job autonomy <sup>39, 40</sup>. Restrictions on resident work hours have reduced burnout rates. Reducing working hours has also benefited physicians in intensive care units and in teaching rotations <sup>6</sup>.

Given the context presented, it is noted that both are necessary for the prevention of BS, as it is triggered by a combination of environmental, social, and individual factors <sup>32</sup>. For this reason, the use of combined interventions is recommended, which combine two or more types of interventions aiming to integratedly modify working conditions, the worker's perception, and coping mechanisms in the face of stressful situations <sup>39, 40, 41</sup>.

Solutions for physician burnout will require coordinated efforts at a national and potentially international level <sup>6</sup>. Fortunately, an increasing number of leaders in the healthcare sector are aware of burnout and are taking steps to address it. The American College of Emergency Physicians recently called on healthcare organizations to promote clinician "well-being," while the American Medical Association has developed tools to help physicians measure and monitor burnout symptoms. Additionally, more attention is now being paid to how physicians spend their time during a typical exhausting workday <sup>11</sup>.

In the UK, the British Medical Association (BMA) of-

fers well-being support services open to all doctors with professional 24/7 telephone support. The Royal Medical Benevolent Fund (RBMF) provides financial assistance when burnout affects work capacity. DocHealth, a joint project of RBMF and BMA, offers confidential psychotherapy to all UK doctors, accessed through self-referral without the need for specific referrals. Additionally, there are also non-governmental initiatives, such as Burnout UK, which offers online courses for doctors to overcome and prevent burnout <sup>33</sup>.

#### CONCLUSION

Physician burnout is prevalent internationally and negatively affects doctors, patients, healthcare organizations, and systems. The triggering factors of this true epidemic are deeply rooted in organizations and healthcare systems, although individual factors also play a role. The longterm effects of BS on workers' health, especially after the COVID-19 pandemic, should be a significant concern for governments, medical entities, hospitals, and physicians themselves. It is important to recognize that each physician faces different challenges and needs different types of support; the most important thing is to recognize when you need that support and reach out. Caring for caregivers is everyone's responsibility and should be seen as a shared responsibility among healthcare systems, organizations, institutions, and individual physicians.

#### REFERENCES

- 1- Oliveira PRC, Melo TG, Lopes A, et al Frequência da Síndrome de Burnout em médicos residentes. Residência Pediátrica 2019;9(2):91-96.
- 2- Freudenberger HJ. Staff Burn-Out. J Soc Issues. 1974; 90(1):159-65.
- Maslach CJS, Jackson SE The measurement of experienced burnout. Journal of Occupational Behaviour. 1981; 2:99-113.
- 4- Maslach CJS, Leiter MP. Maslach Burnout Inventory, 3rd ed. Mountainview, CA: Consulting Psychologists Press; 1996.
- Maslach C, Leiter MP. Understanding the burnout experience: Recent research and its implications for psychiatry. World Psychiatry. 2016; 15(2):103-11.
- 6- West CP, Dyrbye LN, Shanafelt TD. (Mayo Clinic, Rochester, MN; and Stanford University Medical Center, Stanford, CA, USA). Physician burnout: contributors, consequences and solutions (Review). J Intern Med 2018; 283: 516–529.
- 7- McClafferty HH, Hubbard DK, Foradori D, et al; AAP Section on Integrative Medicine. Physician Health and Wellness. Pediatrics. 2022;150(5):e2022059665.
- Meira-Silva VST, Freire ACTN, Zinezzi DP, et al. Burnout syndrome in healthcare workers during the COVID-19 pandemic: a systematic review. Rev Bras Med Trab. 2022; 20(1):122-131. http://dx.doi.org/10.47626/1679-4435-2022-849.
- 9- Lim R, Van Aarsen K, Gray S, et al Emergency medicine physician burnout and wellness in Canada before COVID19: A national survey. CJEM 2020:1–5 DOI 10.1017/cem.2020.431.
- Ferrari R,França FM de,Magalhães J. Avaliação da síndrome de burnout em profissionais de saúde: uma Revisão integrativa da literatura. Revista Eletrônica Gestão & Saúde. 2012; 3:1150-165.
- Grosse E 'Pajama Time': Unraveling The 'Whys' Behind Physician Burnout. 2022. https://www.forbes.com/sites/forbestechcouncil/2022/12/05/ pajama-time-unraveling-the-whys-behind-physician-burnout/?sh=16df-09165cb6
- Moreira HA, Souza KN, Yamaguchi MU Síndrome de Burnout em médicos: uma revisão sistemática. Rev Bras Saude Ocup. 2018;43:e3. http://dx.doi. org?10.1590/2317-6369000013316.
- 13- González RP, González JF. Prevalencia del síndrome de burnout o desgaste profesional en los médicos de atención primaria. Aten Primaria. 1998;22(9):580-4.
- 14- Tucunduva LT, Garcia AP, Prudente FV, Centofanti G, Souza CM, Monteiro TA,

et al. A síndrome da estafa profissional em médicos cancerologistas brasileiros. Rev Assoc Med Bras. 2006;52(2):108-12.

- Barbosa GA. A saúde dos médicos no Brasil. Brasília: Conselho Federal de Medicina; 2007.
- 16- Moreira WC, Sousa AR, Nóbrega MPSS. Adoecimento mental na população geral e em profissionais de saúde durante a COVID-19: scoping review. Texto Contexto Enferm. 2020;29:e20200215.
- 17- Dewa CS, Loong D, Bonato S, Trojanowski L, Rea M. The relationship between resident burnout and safety-related and acceptability-related quality of healthcare: a systematic literature review. BMC Med Educ. 2017;17(1):195.
- 18- Salvagioni DAJ, Melanda FN, Mesas AE, Gonza'lez AD, Gabani FL, Andrade SMd (2017) Physical, psychological and occupational consequences of job burnout: A systematic review of prospective studies. PLoS ONE 12(10): e0185781. https://doi.org/10.1371/journal. pone.0185781
- 19- Shanafelt TD, Balch CM, Bechamps G et al. Burnout and medical errors among American surgeons. Ann Surg 2010; 251: 995–1000.
- Balch CM, Oreskovich MR, Dyrbye LN et al. Personal consequences of malpractice lawsuits on American surgeons. J Am Coll Surg 2011; 213: 657–67.
- West CP, Tan AD, Habermann TM, Sloan JA, Shanafelt TD. Association of resident fatigue and distress with perceived medical errors. JAMA 2009; 302: 1294–300.
- 22- Welp A, Meier LL, Manser T. The interplay between teamwork, clinicians' emotional exhaustion, and clinician-rated patient safety: a longitudinal study. Crit Care 2016; 20: 110.
- Sharma A, Sharp DM, Walker LG, Monson JR. Stress and burnout in colorectal and vascular surgical consultants working in the UK National Health Service. Psychooncology 2008; 17: 570–6.
- 24- Shanafelt TD, Balch CM, Bechamps GJ et al. Burnout and career satisfaction among American surgeons. Ann Surg 2009; 250: 463–71.
- 25- Halbesleben JR, Rathert C. Linking physician burnout and patient outcomes: exploring the dyadic relationship between physicians and patients. Health Care Manage Rev 2008; 33: 29–39.
- 26- Soares LR, Lopes TMO, Silva MAO, et al Burnout e Pensamentos Suicidas em Médicos Residentes de Hospital Universitário. Revista Brasileira de Educação Médica.2012; 36(1): 77-82.
- 27- Asaiag PE, Perotta B, Martins MA, Tempsk P. Avaliação da qualidade de vida, sonolência diurna e burnout em médicos residentes. Rev Bras Educ Med. 2010;34(3):422-9.
- Martins LAN. Natureza e magnitude do estresse na residência médica. Rev Assoc Med Bras. 1998;44(1):28-34.
- 29- Dyrbye LN, West CP, Satele D, et al Burnout Among U.S. Medical Students, Residents, and Early Career Physicians Relative to the General U.S. Population. Acad Med. 2014;89:443–451.
- 30- Small GW. House officer stress syndrome. Psychosomatics 1981; 22: 860-9.
- Nogueira-Martins LA, Jorge MR. Natureza e magnitude do estresse na residência médica. Rev Assoc Med Bras. 1998; 44(1):28-34.
- 32- Perniciotti P, Serrano Júnior CV, Guarita RV, et al. Síndrome de Burnout nos profissionais de saúde: atualização sobre definições, fatores de risco e estratégias de prevenção . Rev. SBPH. 2020; 23:35-52.
- 33- Siobhan Harris. Am I Burned Out? Medscape 27 December 2022.
- 34- Moreno FN, Gil GP, Haddad MCL, et al. Estratégias e intervenções no enfrentamento da síndrome de burnout. Revista enfermagem UERJ. 2011;19:140-5.
- 35- Moss M, Good VS, Gozal D, et al. An official critical care societies collaborative statement: burnout syndrome in critical care healthcare professionals: a call for action. American Journal of Critical Care. 2016; 44(7), 1414–1421. Recuperado de https:// doi.org/10.1097/CCM.000000000001885
- 36- Swensen S, Strongwater S, Mohta NS. Leadership Survey: Immunization Against Burnout Insights Report. New England Journal of Medicine Catalyst. 2018. Recuperado de https:// catalyst.nejm.org/survey-immunization-clinician-burnout/
- 37- Dyrbye LN, Satele D, Sloan J, Shanafelt TD. Utility of a brief screening tool to identify physicians in distress. J Gen Intern Med, 2013; 28: 421–7.
- 38- Shanafelt TD, Kaups KL, Nelson H et al. An interactive individualized intervention to promote behavioral change to increase personal well-being in US surgeons. Ann Surg 2014; 259: 82–8.
- 39- Melo, L. P.de, & Carlotto, M. S. (2017). Programa de prevenção para manejo de estresse e Síndrome de Burnout para bombeiros: Relato de experiência de uma intervenção. Estudos de Psicologia (Natal), 22(1), 99-108. Recuperado de https://dx.doi.org/10.22491/1678- 4669.20170011
- 40- Garrosa, H.E., Benevides, P.A.M.T., Moreno, J.B., &Gozalez, J.L. (2002). Prevenção e intervenção na síndrome de burnout: como prevenir (ou remediar) o processo deburnout. In:Benevides, P.A.M.T. (org). Burnout: quando o trabalho ameaça o bem-estar do trabalhador (pp. 224- 267). São Paulo: Editora Casa do Psicólogo.

 Murta,S.G. & Tróccoli, B.T. (2007). Stress ocupacional em bombeiros: efeitos de intervenção baseada em avaliação de necessidades. Estudos de Psicologia (Campinas), 24(1), 41-51.



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