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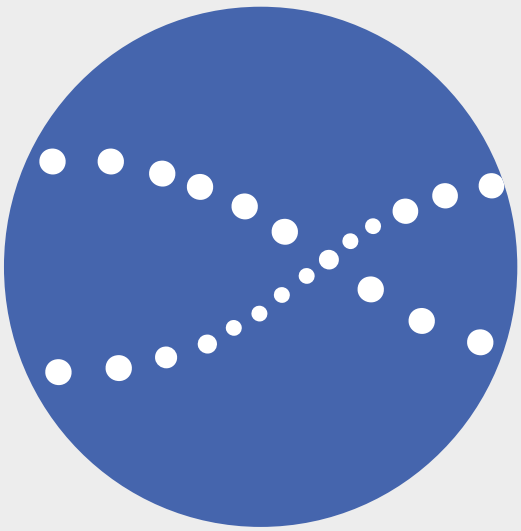

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Scientific Journal of the Paraguayan Society of Surgery (SOPACI)

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Nueva plataforma de la Revista Cirugía Paraguaya

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Starting with this issue, the Cirugía Paraguaya Journal offers a new format for the journal, both on the website and in the print design of the articles.

The current website is www.cirugia.org.py, where the Open Journal Systems platform is hosted. Usernames and passwords, as well as all submitted manuscripts, have been preserved and backed up from the previous site. One of the goals was to offer greater security.

We apologize for the delay in publishing this issue.

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Original article
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Video-assisted retroperitoneal necrosectomy on acute pancreatitis. Itauguá National Hospital, period 2015-2021

Necrosectomía retroperitoneal videoasastida en la pancreatitis aguda. Hospital Nacional de Itauguá, periodo 2015-2021

Carlos Dario Yegros Ortiz*, Daisy Analía González Ayala*, Dennis Cabral**

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ABSTRACT

Introduction: Acute pancreatitis on its necrotic form presents an estimated mortality of 50 % in cases with surgery and up to 100 % without surgery. **Materials:** Observational, descriptive, retrospective, transversal study of patients with a complicated acute pancreatitis diagnosis who had a video-assisted necrosectomy performed through retroperitoneal approach on the Itauguá National Hospital, years 2015 to 2021. **Results:** 35 patients with an average age of 57 years old, 60 % were women, 57.1 % were admitted with a severe pancreatitis diagnosis, and 42.9 % with moderated pancreatitis. The percutaneous drainage was placed first in 29 cases, the patients were directly intervened with a video-assisted necrosectomy in 6 cases. The time between performing the draining and the debridement was 65.5 % between the first and third subsequent week. Two thirds of the patients needed an additional surgical procedure, such as a second video-assisted debridement, cholecystectomy or open necrosectomy. A mortality of 11.4 % was observed. **Conclusion:** Necrotizing pancreatitis' treatment has now averted from open surgical debridement to a more conservative treatment and minimally invasive approaches. The video-assisted retroperitoneal debridement yielded relatively good results as a previous step to open surgery, hence avoiding complications befitting of a laparotomy.

Key words: acute pancreatitis, retroperitoneal debridement, necrosectomy.

RESUMEN

Introducción: La pancreatitis aguda en su forma necrótica presenta una mortalidad estimada en 50 % de los casos con cirugía y hasta 100 % sin cirugía. **MATERIALES:** Estudio observacional, descriptivo, retrospectivo, transversal de pacientes con diagnóstico de pancreatitis aguda complicada en quienes se realizó necrosectomía videoasistida por vía retroperitoneal en el Hospital Nacional de Itauguá, periodo 2015 a 2021. **Resultados:** 35 pacientes con una edad promedio e de 57 años, 60 % fueron mujeres, 57,1 % fue admitido con el diagnóstico de pancreatitis grave y 42,9 % pancreatitis moderada. En 29 casos se realizó la colocación de drenaje percutáneo en primer lugar, en 6 casos los pacientes fueron intervenidos directamente con necrosectomía videoasistida. El tiempo entre la realización del drenaje y el debridamiento fue 65,5% entre la primera y tercera semana posterior. Dos tercios de los pacienter necesitaron un procedimiento quirúrgico adicional, como un segundo debridamiento videoasistido, colecistectomía o necrosectomía abierta. Se observó una mortalidad de 11,4 %. **Conclusión:** El tratamiento de la pancreatitis necrotizante se ha alejado ahora del desbridamiento quirúrgico abierto a un tratamiento más conservador y enfoques mínimamente invasivos. El debridamiento retroperitoneal videoasistido arrojo relativos buenos resultados como escalón previo a la cirugía abierta, evitando así las complicaciones propias de una laparotomía.

Palabras claves: Pancreatitis aguda, debridamiento retroperitoneal, necrosectomía.

INTRODUCTION

One of the most frequent pancreatic diseases in the world is acute pancreatitis (AP). It has an incidence from 5 to 80 cases for every 100,000 people, which variates according to different geographical regions, depending on alcohol consumption and the prevalence of gallstones. [1] AP is characterized by the activation of pancreatic enzymes and release of cytokines. The 20 % of them evolves to become more severe, bringing forth complications such as pancreatic necrosis and sepsis, and finally multiorgan failure. [2]

Currently, the necrosis-infected AP is handled in a minimally invasive manner through the *"step-up approach"*. This methodology consists in the percutaneous or endoscopic drainage with antibiotics, followed by retroperitoneal debridement in case of the former's failure, leaving open surgical drainage as the last resort. [3]

In AP, the main cause of death is infection of the necrotic tissue, which is associated with an improper diagnosis: mortality is approximately up 30 to 39 % in those with infected necrosis (which occurs at some point during the clinical process in approximately a third of the necrosis patients). An intervention in

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the case of an infected pancreatic necrosis is generally required and, with less frequency, in patients with sterile necrosis that are symptomatic (especially in the case of biliary obstruction or gastric or duodenal outlet). Additional treatment has been open surgical necrosectomy: provides ample access to the infected necrosis, but it's highly invasive and associated with reported morbidity rates of 34 % to 95 % and mortality rate of 11% to 39 %, due to the physiological stress of the laparotomic debridement. [4]

The present study is dedicated to evaluating the characteristics of the procedure known as VARD (video-assisted retroperitoneal debridement) on the Itauguá National Hospital during the years 2015 to 2021.

MATERIALS AND METHODS

An observational, descriptive, retrospective, crosscut study was performed.

The sample were patients with a complicated AP diagnosis with infected necrosis intervened through VARD in the Itauguá National Hospital during the years 2015 to 2021.

As for the inclusion criteria, patients intervened through the VARD technique, that have had or not a previous drainage (percutaneous or endoscopic) were selected. As for exclusion criteria, patients intervened through other methods were not selected.

For the case selection, the surgical procedures' register book of the General Surgery Services was accessed, to classify the AP patients on which VARD was performed, requesting clinical files to the Itauguá National Hospital's statistical service afterwards for the recollection of variables of interest.

RESULTS

35 AP and infected necrosis patients were included, on which a VARD was performed. The average age was 57 years old, with a minimal age of 24 and a maximum of 78 years old. The 60 % were of the feminine sex, while the 40 % were masculine.

Regarding comorbidities, 77.1 % reported high blood pressure, 45 % obesity and Mellitus diabetes ***(see Table 1)***.

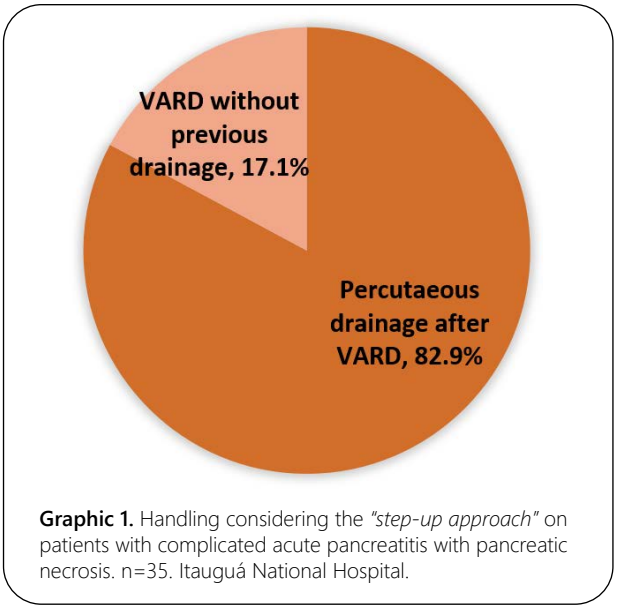
Table 1. Acute pancreatitis patients' comorbidities intervened through VARD.

Comorbidity	n	%
HBP	27	77.1%
Obesity	16	45.7%
Mellitus diabetes	16	45.7%
COPD	4	11.4%
CKD	1	2.8%

*VARD: video-assisted retroperitoneal debridement; HBP: high blood pressure; COPD: chronic obstructive pulmonary disease; CRK: chronic kidney disease

Table 2. Time between the percutaneous drainage's placement and the VARD's execution.

Time between percutaneous drainage and VARD	n	%
1 to 3 weeks	19	65.5%
4 to 6 weeks	8	27.5%
7 to 9 weeks	2	7.0%
Total	29	100%



Graphic 1. Handling considering the *"step-up approach"* on patients with complicated acute pancreatitis with pancreatic necrosis. n=35. Itauguá National Hospital.

Table 3. Surgical interventions after the first VARD.

Surgical intervention after VARD	n	%
Second VARD	12	50%
Cholecystectomy	7	29.1%
Open necrosectomy	5	20.8%
Total	24	100%

Regarding the admission diagnosis, 57.1% were severe AP and 42.9 % moderate AP. As for etiology, 31 patients were of lithiasis cause, 2 alcoholic, and 2 hyperlipidemic.

Of the cited 35 patients on which a VARD was performed, 29 of them (82.9 %) followed a *"step-up approach"* scheme with first a percutaneous drainage placement, and in 6 cases (17.1 %) the patients were directly intervened through VARD ***(see Graphic 1)***.

Time within the drainage's placement and the VARD's execution was 65.5 % between the first and third post drainage weeks, 27.5 % between the fourth and sixth weeks, and 7% between the seventh and nine weeks ***(see Table 2)***.

After the first intervention through VARD, another surgical intervention was performed on 24 of the 35 patients (68.6 %): second VARD on 12 patients (50 %), cholecystectomy on 7 (29.1 %), and open necrosectomy on 5 (20.8 %) ***(see Table 3)***.

The mortality of VARD-treated patients in this case was of 11.4 % (4 patients).

DISCUSSION

AP yet constitutes a severe problem for current-day surgery, despite registered advances in its proper clinical identification, causes, production mechanisms, imagery diagnosis elements, and treatment guidelines. According to statistics, 80 % of the total patients will present a tame form of the disease, associated to an interstitial edematous pancreatic tissue or around the gland, and generally resolved in a week of medical treatment. However, the necrotizing variant of the condition, present on the resulting 20 %, constitutes the most severe outcome, being characterized by pancreatic or peri-pancreatic necrosis, and becoming infected in a third of the cases, associating as well to sepsis statuses, severe sepsis, or simple or multiple organ failure, assessing an

estimated mortality of 50 % of the operated patient total and almost 100 % of the non-operated ones. [5]

On the Itauguá National Hospital between January 2015 and November 2021, 35 patients with infected necrosis AP were intervened through the VARD technique. The average age was 57 years old, with a female predominance of 60 %. These findings coincide with the study performed by Bang et al. where a female predominance of 75 % was found. The cited study reports an age predominance between 18 to 35 years old reaching the 50 %. This age range differs from the range found in our study, in which the patients presented an older age average. [6]

This study is based on patients who received VARD as treatment. The van Santvoort at al. study of the year 2010 showed that close to 35% of *"step-up approach"* patients did not require a subsequent necrosectomy [7]. The minimally invasive handling looks to reduce the surgical stress and associated complications to conventional interventions. Video-assisted handling of pancreatic necrosis improved the morbidity rates of patients submitted to surgical necrosectomy. [8]

It is crucial to find a proper window for performing a VARD. The point of entry must be comfortable for the patient, the trajectory must be as direct as possible and not compromise any organs or vital structures. [9]

Open necrosectomy is associated to a high mortality (approx. 40 %) and morbidity (more than 95 %) including bleeding, gastrointestinal fistulas, and pancreatic insufficiency. In the Wrońsky et al. study, the "*step-up approach*", including the VARD was the superior approach in terms of results when compared to open necrosectomy. The number of patients with complications was significantly greater in patients submitted to a laparotomy compared to those who received VARD. [10]

The dilemma with the percutaneous approach is to define the moment in which the method fails and if the patient requires a necrosectomy, whether video-assisted or through conventional surgery. Early surgery, without a doubt has a higher morbimortality, but a great delay in the surgical indication is also accompanied by an important mortality. Exclusive percutaneous drainage treatment doesn't always achieve a sepsis control on these patients, making the video-assisted surgical handling through retroperitoneal way necessary. [11]

CONCLUSION

The average age of AP patients in this work was 57 years old, predominately of the female sex. In 88.5 % of cases the etiology was lithiasic.

In this study, 82.9 % of patients followed the "step-up approach" scheme, percutaneous drainage and then VARD. Inn 17.1 % a VARD was performed as a first procedure. The time between the percutaneous drainage' performance and the VARD's was of 1-3 weeks (65.5 %).

Approximately two thirds of the patients (68.6 %) submitted to VARD required an additional surgical procedure: 50 % of patients needed a second debridement (VARD) and 20.8 % an open necrosectomy. Mortality was of 11.4 %.

Conflict of interest
Authors declare no conflict of interests.

Author's contribution
All authors participated on the information search, data recollection, draft's redaction, critical revision of the manuscript and final approval of it.

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Ethical considerations
All data were treated confidentially, equally, and justly, respecting the Helsinki principles.

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Comparison of characteristics and morbility in colorectal oncological surgeries with primary anastomosis in elderly patients according to the approach route

Comparación de las características y morbilidad en cirugías oncológicas colorrectales con anastomosis primarias en pacientes de edad avanzada según vía de abordaje.

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ABSTRACT

Introduction: Colorectal cancer is common and has more morbimortality in the elderly. Laparoscopic surgery is used to treat it, showing benefits such as less bleeding and hospitalization time. Despite their vulnerability, the elderly can be benefited from minimally invasive surgeries. **Materials and Methods:** Observational, descriptive, and retrospective study to compare the characteristics and morbimortality of colorectal surgeries with programmed primary anastomosis according to the handling way in 65-year-old or older patients in the General Surgery Services of the Institute of Social Prevision's General Hospital between December 2019 and August 2020. **Results:** Of the 26 patients, 30.7 % was submitted to laparoscopic surgery and 69.2 % to conventional surgery. Most of them were right colectomies. The anastomotic configuration did not significantly differ. Stapler use was 87.5% in laparoscopic surgery and 55.6 % in conventional. There were anastomosis leaks in both groups (27.8 % in conventional). Complications were classified according to Clavien-Dindo observing that more than a third were type 1 and 2. **Conclusions:** Most patients were operated by conventional surgery. Right colectomy was the most performed procedure, and the anastomosis leak was observed in both groups, although with a higher conventional surgery rate. Most complications were mild. More studies with bigger samples are required to evaluate the relationship between surgical technique, surgeon's experience, and morbimortality.

Key words: Colorectal surgery, colorectal cancer, postoperative complications.

RESUMEN

Introducción: El cáncer colorrectal es común y tiene mayor morbimortalidad en los ancianos. La cirugía laparoscópica se usa para tratarlo, mostrando beneficios como menor sangrado y tiempo de hospitalización. A pesar de su vulnerabilidad, los ancianos pueden beneficiarse de técnicas mínimamente invasivas. **Materiales y Métodos:** Estudio observacional, descriptivo y retrospectivo para comparar las características y morbimortalidad de cirugías colorrectales con anastomosis primaria programada según la vía de abordaje en pacientes de 65 años o más en el Servicio de Cirugía General del Hospital Central del Instituto de Previsión Social entre diciembre 2019 y agosto 2020. **Resultados:** De los 26 pacientes, el 30,7 % se sometió a cirugía laparoscópica y el 69,2 % a cirugía convencional. La mayoría fueron colectomías derechas. La configuración anastomótica no difirió significativamente. El uso de grapadora fue de 87,5 % en cirugía laparoscópica y 55,6 % en convencional. Hubo fugas de anastomosis en ambos grupos (27,8 % en convencional). Las complicaciones se clasificaron según Clavien-Dindo observándose que más de los 2/3 fueron tipo 1 y 2. **Conclusiones:** La mayoría de los pacientes fue operado por cirugía convencional. La colectomía derecha fue el procedimiento más realizado y la fuga de anastomosis se observó en ambos grupos, aunque con una tasa mayor en cirugía convencional. La mayoría de las complicaciones fueron leves. Se requieren más estudios con mayor muestra para evaluar relación entre técnica quirúrgica, experiencia del cirujano y morbimortalidad.

Palabras clave: Cirugía colorrectal, cáncer colorrectal, complicaciones postoperatorias.

INTRODUCTION

Colorectal resections can be performed through malignant or benign pathologies, with anastomosis or ostomy confection. The anastomosis types include the term-terminal, term-lateral, or lateral-lateral type, with mechanical (staple) or manual suture. The surgical options include ileus-colic resection, right, transverse, or left colectomies, sigmoidectomy, and protocolectomy, among others. Conventional or minimally invasive handlings can be used, and in the latter group the manually or robotically assisted laparoscopic handling are included. [1]

The colorectal carcinoma is the most common neoplasia of the digestive tube, and its treatment has surgical, curative or pal-

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liative indication, even with the existence of metastasis, [2] because it decreases the tumoral load and prevents possible complications.[3] Among the most important risk factors ageing is included, given that 90% of patients who suffer from it are older than 50 years old, however, it's also prevalent in patients younger than 50 years old hence a colonoscopy is key for diagnosing.[4] The surgery's individualization in elderly patients looks to reduce complications. Minimally invasive surgery, even ambulatory, is studied for better results. [5] Postsurgical complications can be evaluated according to the Clavien-Dindo classification.[6]

In the present article, morbidity and colorectal surgeries with primary anastomosis' characteristics were compared in patients older than 65 years old, according to the handling way, in the Institute of Social Prevision's Central Hospital (December 2019 – August 2020).

MATERIALS Y METHODS

An observational, descriptive, and retrospective study was performed. The data was collected through the selection of clinical files and procurement of a database. The inclusion criteria were colorectal cancer patients of 65 or more years of age, submitted to colorectal surgery with programmed primary anastomosis, admitted to General Surgery Services of the Institute of Social Prevision's Central Hospital, during the December 2019 to August 2020 period. Among the exclusion criteria are patients whose initial surgery was performed in a different facility and patients with an incomplete clinical history or medical file.

The surgical handling method was determined and compared to the anastomosis' configuration, technique, drainage utilization, morbimortality according to the Clavien-Dindo classification, anastomosis leak and inpatient stay. Once the information was compiled, a database processed through an Excel® sheet was designed.

RESULTS

26 patients of 65 years old or older submitted to colorectal surgeries with primary anastomosis were evaluated, of which 30.7 % were performed by laparoscopic way, and 69.2 % by conventional way.

Base pathologies were found present on all patients operated through laparoscopic way (8/8), and in 72.2 % (13/18) of those operated by conventional way.

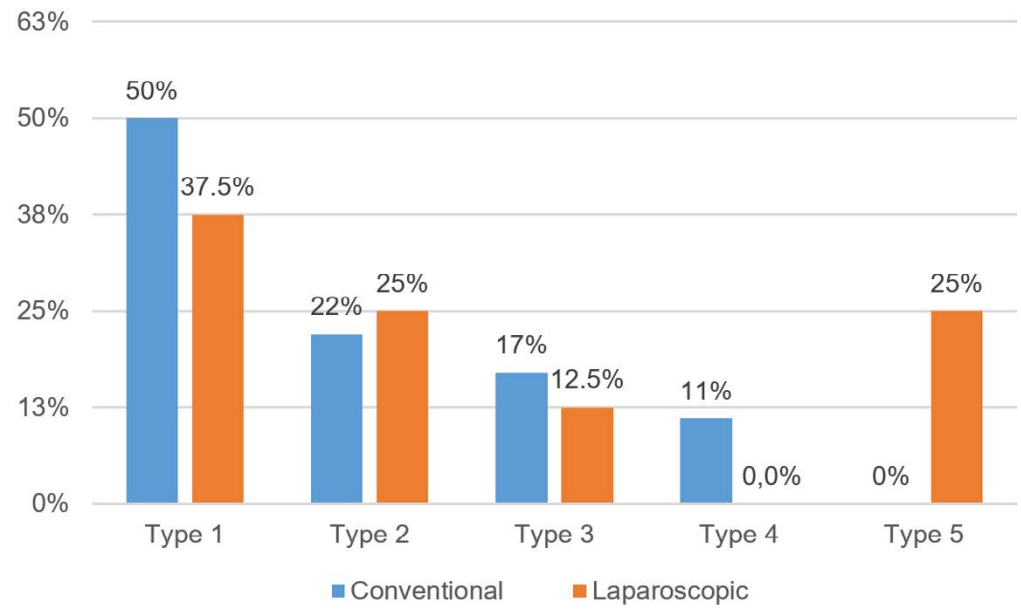
Within the group submitted to laparoscopic-way surgery, 62.5 % were right colectomies, 25 % left colectomies, and 12.5 % sigmoidectomies. As for the group submitted to conventional-way surgery, 61.1% were right colectomies, 16.6 % sigmoidectomies, 11.1 % lower anterior resections, 5.5 % left colectomies, and 5.5 % transversectomies.

Anastomotic configuration in the group submitted to laparoscopic-way surgery was 37.5 % term-terminal, 37.5 % lateral-lateral, and 25% term-lateral. Through conventional way 38.8 % were lateral-lateral, 33.3% were term-lateral, and 27.7 % were term-terminal. Manual suturing was used in 12.5 % of laparoscopic approaches and in 44.4 % of conventional-way approaches. Mechanical suturing was used in 87.5 % of laparoscopic approaches and in 55.5 % of conventional approaches. ***(see Table 1)***

Regarding anastomosis' complications, 12.5 % presented anastomosis leak through laparoscopic way and 27.7 % through conventional way. 87.5 % of patients operated through laparoscopic way had a tubular drainage placement, and 88.8 % through conventional way did so as well. ***(see Table 1)***

According to the Clavien-Dindo classification, the most frequent complication amongst both groups was type 1. ***(see Table 1, Graphic 1)***

The average inpatient stay of patients operated through laparoscopic way was 6.75 days, and 9.2 days through conventional way.



Graphic 1. Complications' classification according to the Clavien-Dindo classification

Table 1. Characteristics of patients over 65 years of age with colorectal surgery and primary anastomosis.

		Conventional (open)		Laparoscopic		ji2	p value
		n	%	n	%		
Anastomosis configuration	Termino-terminal	5	27.8	3	37.5	0.2979	0.8616
	Termino-lateral	6	33.3	2	25.0		
	Latero-lateral	7	38.9	3	37.5		
Anastomosis technique	Mechanical	10	55.6	7	87.5	2.4971	0.1141
	Manual	8	44.4	1	12.5		
Dreainage	With drainage	16	88.9	7	87.5	0.0105	0.9185
	Without drainage	2	11.1	1	12.5		
Complication (Clavien-DIndo)	Type 1	9	50.0	3	37.5	—	—
	Type 2	4	22.2	2	25.0		
	Type 3	3	16.7	1	12.5		
	Type 4	2	11.1	0	0.0		
	Type 5	0	0.0	2	25.0		
Anastomosis leakage	Present	5	27.8	1	12.5	0.7282	0.3935
	Absent	13	72.2	7	87.5		

DISCUSSION

A revision of 26 colorectal cancer patients who were submitted to colorectal surgeries with primary anastomosis was performed. The most common surgical approach was conventional-way and although it was the most frequent, laparoscopic-way has been the protagonist in our hospital center. In our current day, the laparoscopic way has become the standard method for colon surgeries, [7] the use of laparoscopic surgery on geriatric patients with colorectal cancer should increase due to the better long-term results. [8]

No significative difference between the different anastomosis configurations was observed, noting a similar frequency among term-terminal, lateral-lateral, and term-lateral anastomosis for both approach ways. This is also subjected to the patient's localization and anatomy, the ileocolic anastomosis configuration's impact that might have in the intestinal transit's restitution after a right hemicolectomy. It's necessary to perform a randomized essay comparing isoperistaltic and antiperistaltic modalities, [9] ileocolic, isoperistaltic, and antiperistaltic presented similar results in terms of performance, security, and functionality. [10]

Regarding the anastomosis technique, the use of mechanical stapler for anastomosis was predominant in both groups. It's worth mentioning that the use of stapler with manual suture backup after the rectum cancer's laparoscopic radical resection is convenient and effective, [11] making the standardized use of laparoscopic way and stapler for rectum cancer evident. A group of collaborators from the European Coloproctology Society informed increased use of the stapler technique rather than the manual one, alike our study, however, the most used surgical approach was laparoscopic way, which does not match the data of our study. [12]

Anastomosis leak was mostly present with conventional way and in an increased rate. Luglio and Corcione informed lesser anastomosis leak with the stapler's use, however, they did not report the surgical approach way and specifically studied ileocolonic anastomosis. [13] Nordholom et al however reports a times two risk of anastomosis leak with the stapler's use for ileocolic anastomosis, although it does not specify the surgical approach. [14] The collaborators' group from the European Coloproctology Society studied the predictive factors of anastomosis leak in right colon cancer, where the surgery's duration, conventional approach and stapler's use was associated with a higher risk of anastomosis leakage. [12] There are studies that report that anastomosis leak us related to the deteriorated general survival and high local recurrency. [15,16]

In our study, abdominal drainage was left in almost every case, regardless of surgical approach. Some authors are not in favor of the rutinary use of drainages after colon and rectal anastomosis, voicing that it's associated with an increase in injury's infection rate which can be up to a 25 %. [17]

For the morbimortality assessment the Clavien-Dindo classification was used. We observed that the most frequent complication was type 1, for laparoscopic approach as much as for conventional, although no type 5 complications were observed for the conventional-way submitted group, which were present in 25% of the laparoscopic-way submitted patients. The fact that most patients presented type 1 complications is encouraging given that studies report that the complications' gravity is related to the general survivability. [18,19]

The shorter inpatient stay was for laparoscopic way, which matches with other studies with similar results. [20]

This article may be biased due to the low quantity of studied patients and the fact that the study was performed in a specialized hospital.

CONCLUSION

Of the patient total (26), most (69.2 %) was submitted to conventional surgery. The most common resection type was right colectomy. Anastomosis configuration and the use of stapler did not show variations amongst the conventional and laparoscopic surgery groups. Anastomosis leaks were registered in both groups, with a higher rate in the conventional surgery group (27.7 %). Complications are categorized according to the Clavien-Dindo classification, without significant discrepancies.

CONFLICT OF INTEREST

Authors declare no conflict of interests.

AUTHOR'S CONTRIBUTION

Dr. Pablo Schaerer, Dr. Marcelo Samudio, Dr. Michelle Feltes, Dr. Mónica Martínez, Dr. Monserrat Riquelme, Dr. Gabriela Sanabria, and Resident Giulianna Benedetti conceived the idea, elaborated the manuscript, bibliographic research, and final revision.

ETHICAL CONSIDERATIONS

All principles of bioethics were upheld. Confidentiality of the results was kept under the use of a numeric code that identified each file, data only being known by the authors.

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Complications of total laryngectomy in the Department of Otorhinolaryngology of the Hospital de Clínicas from 2015-2022

Complicaciones de la laringectomía total en la Catedra de Otorrinolaringología del Hospital de Clínicas de 2015-2022

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ABSTRACT

Introduction: Laryngeal cancer is the most common malignant neoplasia of the aerodigestive tract, representing 4.5 % of the organism's cancers and the 30% of the head and neck cancers. Post-operatory complications of the total laryngectomy are frequent and predisposing factors for them are known. Knowing the prevalence of total laryngectomy's complications, performed means for its treatment, anatomical site, presence or absence of previous adjuvant treatments were set as objectives. **Materials and methods:** A descriptive and retrospective study was conducted in the Clinical Hospital, Otorhinolaryngology Area, from 2015 to 2022. The study was constituted by a total of 46 patients admitted with the laryngeal cancer diagnosis, 21.7 % (10) of which had a total laryngectomy performed on them. Other surgical techniques were excluded. **Results:** 10 patients submitted to a total laryngectomy within an age range of 31-70 years old, all males. Supraglottic compromise was of 30 %. 70 % presented complications. The most frequent complication was pharyngocutaneous fistula, others were immediate post-operatory bleeding and surgical site's infection. 71.4 % of patients with fistula were resolved only with conservative means, 2 required reintervention for the defect's closure. **Conclusion:** Total laryngectomy complications are present very frequently, affecting the quality of life, evolution, and post-surgical recovery of the patients.

Keywords: pharyngocutaneous fistula, laryngectomy, laryngeal cancer.

RESUMEN

Introducción: El cáncer de laringe es la neoplasia maligna más común de las vías aerodigestivas, representa el 4,5% de los cánceres del organismo y el 30% del cáncer de cabeza y cuello. Las complicaciones postoperatorias de la laringectomía total son frecuentes y se conocen factores predisponentes para ello. Se estableció como objetivo conocer la prevalencia de las complicaciones de la laringectomía total, las medidas realizadas para su tratamiento, el sitio anatómico, la presencia o ausencia de tratamiento adyuvante previo. **Materiales y métodos:** Se realizó un estudio descriptivo y retrospectivo en el Hospital de Clínicas, Cátedra de Otorrinolaringología, del 2015 al 2022. El universo estuvo constituido por un total de 46 pacientes ingresados con el diagnóstico de cáncer de laringe, 21,7% (10) de los cuales se les realizó laringectomía total. Se excluyeron otras técnicas quirúrgicas. **Resultados:** 10 pacientes sometidos a laringectomía total con un rango de edad de 31-70 años, todos de sexo masculino. El compromiso de la supraglotis fue del 30%. El 70% presentó complicaciones. La complicación más frecuente fue fistula faringocutánea, otras fueron sangrado en el postoperatorio inmediato e infección del sitio quirúrgico. El 71,4% de los pacientes con fistula se resolvieron únicamente con medidas conservadoras, 2 requirieron reintervención para el cierre del defecto. **Conclusiones:** Las complicaciones de la laringectomía total se presentan con mucha frecuencia, afectando a la calidad de vida, la evolución y recuperación post quirúrgica del paciente.

Palabras clave: fistula faringocutanea, laringectomía, cáncer de laringe.

INTRODUCTION

Laryngeal cancer is the most common malignant neoplasia of the aerodigestive tract, representing 4.5% of the organism's cancers and 30% of the head and neck cancers. Several risk factors have been implicated in laryngeal cancer's pathogenesis. The most significant of these is alcohol and tobacco consumption [1, 2, 3]. It has been proved that the use of tobacco has a lineal association with the development of laryngeal cancer, with a risk for smokers that is 10 to 15 times greater than it is for non-smokers. It has also been proven that alcohol and tobacco have a multiplying effect on the risk of laryngeal cancer [1, 2, 4]. It's believed that exposure to several other ambiental factors potentially increase the risk of larynx's squamous cell carcinoma, such as asbestos,

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polycyclic aromatic hydrocarbons, and textile powder [1].

The clinical presentation of laryngeal cancer is variable and depends on the anatomical localization and size of the tumor. Tumors that sit on the glottis typically produce an early symptomatology, presenting dysphonia, which leads to consultation and earlier diagnosis. It's posed that every patient with a dysphonia longer than 15 days of evolution with risk factors of developing a laryngeal cancer must be evaluated by an otorhinolaryngologist. Cancers that sit on the supraglottic region are typically of later diagnosis given that they initially present vague symptoms such as sore throat, laryngeal strange body sensation, occasional dysphagia and later dysphonia, stridor, dysphagia. Subglottic tumors are less frequent, and symptoms are dyspnea or a lower cervical mass [1, 5].

The most important adverse forecast factors for laryngeal cancer include increasing the T stage and the N stage. Other forecast factors can include sex, age, functional state, and a variety of the tumor's pathological characteristics, including the degree and depth of the invasion [3, 4, 6, 7]. Total laryngectomy is indicated on advanced injuries with intrinsic muscular invasion of the vocal cord, thyroid cartilage's compromise, exophytic lesions which compromise both commissures and arytenoids or in patients with subglottic or supraglottic invasion above the epiglottis' borders and aryepiglottic folds, and in failure of radiant treatment in T1 and T2 of the larynx, in which a conservative surgery is not possible [3, 4, 7, 8]. Post-operative complications of the total laryngectomy are frequent. Local-regional complications appear to be the most common ones such as surgical site's infection and hematoma, necrosis of the flap used in the neopharynx's closure, tracheostomy, pharyngostoma complications, lymphorrhea or vascular rupture [3, 5, 8, 9].

Multiple favoring factors have been studied, amongst them malnutrition and previous radiotherapy are the most significant ones. Other favoring factors exist in several studies like anemia, preexisting tracheostomy, lymph node dissection, and lesser surgical experience [7, 8, 10]. In the past few years, systemic hematological markers have become increasingly more renown as results' forecasts of malignant neoplasia. Recently, the RDW (red blood cell distribution width) has proven to be of forecasting utility in patients with different malignant neoplasia. A previous study yielded that RDW could predict the survivability of laryngeal cancer patients, an extracapsular extension, that affects the neck and results of the surgical treatment. These factors are associated with the disease's ailing; little is known about forecast factors not related to the disease [11, 12, 13].

The present article's objective is to describe the post-surgical complications' type and quantity of patients who've had a total laryngectomy, performed measures for its treatment, neoplasia's most frequent anatomical site, presence of absence of adjuvant treatment before surgical treatment.

MATERIALS AND METHODS

A revision of total laryngectomy's complications was performed in the Clinical Hospital of the Medical Sciences' Faculty's Otorhinolaryngology Services during a period of 7 years, comparing our results with the results of international actors and national statistics. An observational, descriptive, cross-cut retrospective investigation was performed. The sampling was of consecutive cases by convenience. The data was obtained from the patient's clinical files and registered in a Microsoft Excel® 2016 spreadsheet and descriptive statistics were done in the SPSS® Statistics 28 software. A characterization of the patients was performed according to the following variables.

- Age.
- Sex.
- Complication.
- Complication's treatment.
- Lesion's anatomical site.
- Adjuvating treatment.
- Histological differentiation.
- RDW.
- Symptoms' evolutive time.
- Follow-up time.

The target population were post-operative total laryngectomy patients by larynx's neoplasia within our services from the year 2015 to 2022 from general otorhinolaryngology consultations or redirected from other centers that fit the inclusion criteria.

Inclusion criteria: Patients older than 18 years old, total laryngectomy post-operative patients with larynx's neoplasia anatomopathological diagnosis.

Exclusion criteria: Patients that haven't been submitted to a surgical procedure, patients with an incomplete file, patients that abandoned follow-up.

RESULTS

Within the studied series, 46 files from patients who consulted through our service from 2015 to 2022 were revised, with the larynx's neoplasia diagnosis, of which (10) 21.7 % of patients had a total laryngectomy performed ***(see Image 1)***.

The average age of the 10 patients was of 56,3 ± 10.2 years-old, in an age range from 31 to 70 years old, all males. 90 % of patients presented risk factors related to laryngeal cancer. 30 % of patients had a supraglottic affectation, 70 % of them only had a glottic-region affectation. Half of the patients had received preoperatory radiotherapy ***(see Table 1)***.

70 % of operated patients presented a pharyngocutaneous fistula as a complication, furthermore this group of patients presented post-operative bleeding (which required reintervention) and a patient presented a surgical site infection ***(see Table 1)***.

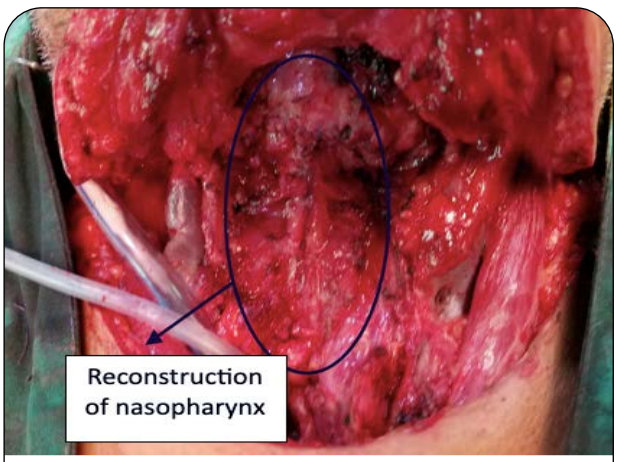

Figure 1. Neopharynx's reconstruction before a total laryngectomy.

Table 1. Demographic data and clinical characteristics

Clinical-demographical characteristics	
Average age	56.3 years-old ± 10.2 (31-70)
Sex	
Male	10 (100%)
Female	0 (0)
Previous radiotherapy	
Yes	5 (50%)
No	5 (50%)
Anatomical location	
Glottis	7 (70%)
Glottis + supraglottis	3 (30%)
Post-operative complications	
Pharyngocutaneous fistula	7
Bleeding	1
Surgical site's infection	1
Patients with post-operative complications' total	7 (70%)
Complication's treatment	
Compressive bandaging	5 (71.4%)
Reintervention: pectoral flap	2 (28.6%)
Antibiotic therapy	1 (14.3%)
Reintervention due to bleeding	1 (14.3%)
RDW value in complications	
<13%	4 (57.1%)
13-14.3%	2 (28.6%)
>14,3%	1 (14.3%)

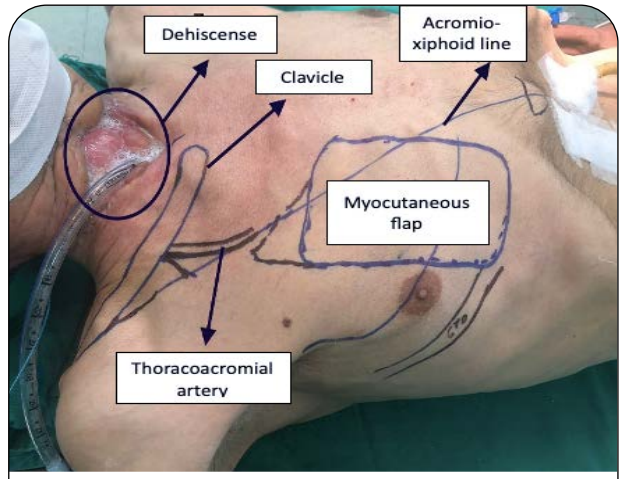

Image 2. Pectoral flap's design for the pharyngocutaneous fistula's closure.

Of the 7 pharyngocutaneous fistula patients, 5 (71.4 %) received compressive bandaging as therapeutic measure, only 2 patients (28.6 %) required a second intervention for the defect's closure consisting of a rotatory pectoral flap ***(see Image 2).***

Of the patients that presented complications, 4 (57.1 %) received radiotherapy before surgical treatment. 57.1 % presented a RWD lesser than 13 %, and only one presented a RWD greater than 14.3 %.

All patients had a squamous cell carcinoma preoperatory anatomopathological diagnosis. Afterwards the most frequently histological differentiation found in 7 patients was grade II-III, a patient presented a grade I-II differentiation.

The evolutive time of the disease before the laryngectomy was of 10.4 months (±7.7). The average follow-up time was of 7.3 months (±5.1).

DISCUSSION

Almost all the patients included in our study were adult males, justified by the fact that being a neoplastic pathology means that the patients are usually older adults, and matching Gülpembe Bozkurt's research in which they obtained similar results with an average age of 59.7 ± 9.4[8, 11].

All patients were of the masculine sex, matching with the study done by María Fernández-Prada, in which 98.6 % were males. This finding could be due to the social environment with more risk factors associated to the masculine sex, likewise Valero Ruiz reported the highest incidence during the 60's and it affected men 10 times more than women [14, 15].

Of the sample total only 21.7 % was submitted to a total laryngectomy, this due to the greater radiotherapy availability and good results of it, hence many of our patients choose a conservative treatment. Despite this, total laryngectomy is still the first line in some patients with infiltrating carcinomas in stages III and IV, where conservative isn't entirely effective.

Of the patients included in our study a lesion sitting on the glottis was mostly discovered, and only 30 % had lesions that also covered the supraglottic region, in a previous study of our service a supraglottic compromise was found in 50 % of the cases. Other authors speak of a greater prevalence of supraglottic lesions found in up to 66 %. As far as primary lesion's localization concerns, supraglottic tumors are associated in several series with a higher risk of fistula formation, given that their location requires greater pharyngeal mucosa resection and with it a greater tension of the suturing during the defect's closure [7, 8, 9, 12, 18].

Of the 10 patients submitted to a total laryngectomy, 90 % presented risk factors related to laryngeal cancer, which relates to María de los Ángeles Reynaldo González's study where 91.3 % of patients had some risk factor [3]. Most of the patients with a larynx squamous cell carcinoma were or are heavy smokers. Often with high alcohol consumption [2, 3, 8].

Pharyngocutaneous fistula is the most frequent post-operatory complication after a total laryngectomy. Pharyngocutaneous fistula's prevalence in our sampling was 70 % of patients submitted to a laryngectomy, this being greater than the results found by María Fernández-Prada who reports a 48.6 % incidence, other authors report different values such as 49,6 % Aires et al., 34.5 % Sarra et al. In a similar study within our services, a 50 % prevalence was found in 2012 [3, 8, 9].

It was also found that a patients presented a surgical site infection, as a complication besides pharyngocutaneous fistula, in similar studies this complication was reported in 67.53 % in the

study done by María Antonieta Álvarez Urbay et al. A previous study within our services yielded an 8,3 % prevalence, very similar to the one found in the current data. Acevedo Ortiz et al. published a surgical site infection rate of 14.5 %, similar to the data found in our sampling [9, 10, 11, 16].

The 10 %, a patient, presented surgical site bleeding which required a reintervention to contain the bleeding, while in the reports this complication was found with 11.3 % prevalence in the study by Laura Acevedo Ortiz et al [11, 16]. We must highlight that some patients met many of the complications, which are often cause and effect of each other.

71.4 % of post-laryngectomy pharyngocutaneous fistula were resolved by conservative measures without the requirement of other types of interventions for the defect's closure, this matches the work of Víctor Palomar-Asenjo et al. who reported that 72.7 % of patients were resolved spontaneously with conservative measures [6]. Only 2 patients required the defect's closure with a reintervention, both of which gad a pectoral rotatory flap performed for the closure ***(see Figure 2)***.

There are many authors who point out that patients who have received adjuvant treatment before surgery have greater pharyngocutaneous fistula incidence. In our patients, a greater tendency towards pharyngocutaneous fistula was observed in previously irradiated patients. Treatment with radiotherapy before the laryngectomy has been said to, not just increase the risk of fistula's incidence but also increase in-patient stay and the need for surgical treatment [19, 20].

RDW is a simple marker of easy systemic inflammatory response's attainment, and it was recently reported that it negatively affects clinical results of several types of cancer. However, there are few studies related to head and neck cancers. There are authors who report a great chance of suffering distance metastasis or a greater prevalence of non-surgical systemic complications, such as deep vein thrombosis, pneumonia, cardiovascular events, and difficulty to disconnect from mechanical ventilation with RWD values greater than 14.4 [11, 12].

CONCLUSION

All patients were of the masculine sex, with an average age of 56.3 years old and a previous evolutive time of 10.4 months.

Of the 10 patients who had a total laryngectomy, 7 presented complications afterwards. The most frequent complication and present in all 7 patients was pharyngocutaneous fistula, which was mostly handled conservatively. Only two patients required a major pectoral muscle flap. Neoplastic lesions were mainly found in the glottic region and half of the sample had received radiotherapy before the surgical procedure. A patient presented immediate post-operatory bleeding which required an emergency intervention. The RDW value in complicated patients was generally less than 13 %.

Author's contribution

All authors have equally contributed to the following aspects: a. Creation and design of the work/idea; b. Data recollection and obtaining results, c. Data analysis and interpretation; d. Writing of the draft; e. Draft's revision; f. Approval of the final draft; g. Patients or study material contribution.

Conflict of interest

There are no factors that can lead to a conflict of interest between the work's authors.

Ethical considerations

The study had the permission of the Otorhinolaryngology Services and all people involved, respecting the principles of: beneficence, non-maleficence, autonomy, and justice.

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Experience in the treatment of complex perianal fistulas in a public hospital. 2018-2022

Experiencia en el tratamiento de las fístulas perianales complejas en un hospital público. 2018-2022

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ABSTRACT

Introduction: Complex perianal fistula treatment is controversial; it requires a precise anatomical and auxiliary diagnosis methods' knowledge to offer the best result possible for the patient. **Objective:** To describe the experience of complex perianal fistula treatment within the Public Hospital during the 2018 – 2022 period. **Materials and Methods:** Observational, descriptive, cross sectional, non-probabilistic study of consecutive patients with a complex perianal fistula diagnosis admitted to coloproctology services in the Itauguá National Hospital during the January 2018 – December 2022 period, of which 21 fit the complex perianal fistula criteria. **Results:** Of the 21 patients, 76% were male, with an average age of 35 ± 10 years old, the main consultation reason was anal secretion in 19 patients. Regarding the fistula type, an 81% high transsphincteric fistula was identified, as for the treatment, in 48% a fistulectomy was performed, and regarding complications, only 10% of patients suffered from fistula recurrence which was treated through a fistulectomy with seton placement. **Conclusion:** Complex perianal fistula is an infrequent periorificial pathology, the data obtained from physical examination and the possibility to effectuate complementary studies with advanced technological resources are fundamental to a satisfactory resolution.

Keywords: perianal fistula, complex fistula, fistulectomy, endorectal ultrasound, nuclear magnetic resonance.

RESUMEN

Introducción: El tratamiento de la fistula perianal compleja es controvertido, requiere de un conocimiento anatómico preciso y de métodos auxiliares de diagnóstico para ofrecer el mejor resultado posible al paciente. **Objetivos:** Describir la experiencia en el tratamiento de las Fístulas Perianales complejas en un Hospital Público en el periodo 2018 – 2022. **Material y Métodos:** Estudio observacional descriptivo de corte transversal, no probabilístico de casos consecutivos de pacientes con diagnóstico de fistula perianal compleja ingresados en el servicio de coloproctología del Hospital Nacional de Itauguá en el periodo de enero de 2018 hasta diciembre de 2022, de los cuales 21 cumplen con los criterios de inclusión de fistula perianal compleja. **Resultados:** De los 21 pacientes, 76% fueron hombres, con una media de edad de 35 años ± 10, el principal motivo de consulta fue secreción anal en 19 pacientes. En cuanto al tipo de fístula se identificó un 81% de fístula transesfinteriana alta, en cuanto al tratamiento, se realizó en un 48% fistulectomìas, en cuanto a las complicaciones, solo en un 10 % de los pacientes se constató recidiva de la fístula que fue tratada con fistulotomía con colocación sedal. **Conclusión:** La fístula perianal compleja es una patología periorificial poco frecuente, son fundamentales los datos obtenidos por el examen físico y la posibilidad de efectuar estudios complementarios con recursos tecnológicos avanzados para su resolución satisfactoria.

Palabras claves: fístula perianal, fístula compleja, fistulectomía, ecografía endorrectal, resonancia magnética nuclear.

INTRODUCTION

Complex perianal fistula treatment is controversial; it requires a precise anatomical and auxiliary diagnosis methods' knowledge to offer the best result possible for the patient. A complex fistula on the anus is difficult to diagnose and treat, requiring careful approaches due to the high risk of complications and recurrences. [1]

In over 90% of cases, fistulas are caused by a cryptoglandular-level infection, from there, it spreads from the intersphincteric space to perirectal anatomical spaces, allowing several morphological variables which define the complexity of this pathology. On the remaining 10% its origin can be secondary to Crohn's disease, radiotherapy, tuberculosis, concussion, iatrogenic, among others. Not all perianal orifices correspond to perianal fistula. The tract between the intersphincteric space and external orifice normally defines the type of fistula. Its frequency is greater in men than in women. [1-2]

Regarding classification, Parks suggested a classification based on the relation between the fistula and external anal sphincter. Four types were described: Intersphincteric (the tract goes through only the internal sphincter, most common, represents around 60 % of cases), Transsphincteric (the tract goes through both sphincters: constitutes around 25 % of cases), Suprasphincteric (the tract goes above the external sphincter, in the puborectalis, representing less than 5 %), Extrasphincteric (the tract goes outside the sphincters: does not origin within the crypt. Are caused by concussions, inflammatory bowel disease, or pelvic sepsis. Constitute less than 5 % of cases) [2].

Generally, anal fistulas are simple, which is to say, present an external orifice, evident internal orifice, and a unique, rectilinear, and low tract between the two. The surgical solution is easy and involves a complete plane laying, many times in ambulatory

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surgery regimen and with low anal incontinence risk. When the described situation varies, a complex anal fistula must be taken as a possibility. The complexity criteria are dictated by the fistula's anatomical characteristics or by the surgical intervention's complexity which is crucial to cure and preserve continence. [3] ***(see Table 1)***

Table 1. Complex Fistula Characteristics

Complex Fistula Characteristics
Suprasphincteric, high transsphincteric, and some middle transsphincteric fistulas.
Anterior face fistula on woman.
Extrasphincteric fistulas.
Secondary tract and intermediate cavity fistulas.
Relapsed fistulas.
Fistulas without internal orifice.
Horseshoe fistula.
Fistulas with several internal orifices or orifices situated on top of the pectineal line.
Rectovaginal fistulas, the ones after radiotherapy and patients with an inflammatory bowel disease (IBD), especially Crohn's disease.

DIAGNOSTIC

Diagnostic is based on anamnesis and physical exploration, occasionally including an anoscopy. A well-trained finger is still an exceptionally useful tool for the study of anal fistula. At first the external orifice is studied and additional orifices and debridement or previous intervention scars are investigated. Next, the subcutaneous induration which goes from the external orifice to the anus' deepness, is palpated. This maneuver is not only useful for evaluating the tract's direction, but it even helps to identify additional trajectories or indurations. Afterwards, a rectal feel to identify the internal orifice is performed. This is identified as a small protuberance or depression usually situated on the dented line in high correlation to the traditional Goodsall rule. [4]

Endoanal ultrasound with rotating sound is an exploration that offers high-definition real-time images, with focal distance of up to 6 cm across the entire rectum and anal canal's perimeter, which allows the study of most fistulas. The oxygenated water instillation through the external orifice increases the precision to study the tract and localize the internal orifice. [5]

MRIs also provides very exact information about the anal fistula's morphology, especially when an endoanal coil is used. It's a more sophisticated and costly exploration which requires adequate infrastructure and a radio-diagnostic specialist with deep knowledge in anorectal anatomy and data of interest for the surgeon. It's very useful to value extrasphincteric plans, but more difficultly differentiates sphincteric muscles. [6]

Fistulography has been displaced by ultrasound and MRI. Most likely, its use must be restricted nowadays in cases in which these two explorations don't contribute useful data. CT scans, especially with the introduction of fistulous orifice contrast, can complement fistulography indications. [7]

Surgical options for implementation include ***Fistulotomy (laid plan):*** consists of the full fistulous tract's longitudinal opening. Usually associated with a marsupialization of the fistula's edges, this procedure is used on simple fistulas, however, it can also be used in complex fistulas. [3]

Fistulectomy: it's the extraction of the fistulous tract, a type of technique to preserve the sphincter, procedure which allows precise resection of the fistula's tract, hence reducing the chance of losing a secondary tract. [8]

Advancement endorectal flap: Probably the most used method for high fistula treatment. Total or partial removal of the internal orifice and fistulous tract, a flap is designed which includes mucous and part of the internal anal sphincter's fibers that displaces in craniocaudal-wise to completely cover the fistula's previous orifice and creates a barrier against the rectum's pressure, bringing healthy and vascularized tissue, which favors definitive cicatrization. [9]

Sealant or fibrin glue: This technique, described several years ago, consist of scraping and washing the fistulous tract with oxygenated water, and introducing a sound, a combination of fibrinogen and thrombin (fibrin), to seal the entire tract and favor its cicatrization. It's indicated in fistulas which present only a single tract, without intermediate cavities nor infection, and well-defined internal and external orifices. Healing indexes are varied, ranging from 14 to 85 %, taking into account what its failure does not prevent the use of any other posterior method. [9]

Collagen plug: consists of a porcine small intestine's acellular derivative of extracellular matrix, which allows the inclusion and remodeling of scar tissue. [9]

The seton: the internal and external orifice are linked and knotted on the exterior, presenting several uses such as, *cutting* seton, which is progressively tightened until the sphincteric muscles are sectioned. *Drainage* seton, to trigger the formation of a fibrous tract when there's an active fistula infection. [10]

LIFT (Ligation of the Intersphincteric Fistula Tract), consists of the fistulous tract's ligation to the intersphincteric plan's level, fully preserving the sphincters. It has a healing rate ranging from 68 % to 83 %, with an average healing time of 6 to 7 weeks. [11]

TROPIS (*Transanal opening of intersphincteric space*), like LIFT, where the fistula's tract is linked and cut on the intersphincteric plan, while in TROPIS, the intersphincteric portion of the fistula is opened (the roof is removed) on the anal canal. The roof removal is done for the intersphincteric portion of the tract is completely drained. [12]

PERFACT (*Proximal superficial cauterization, emptying regularly fistula tracts and curettage of tracts*) is a useful method for complex anal fistulas, even in abscess-associated fistulas, supralevator fistulas and when the internal opening cannot be located. The PERFACT procedure involves two steps: superficial cauterization of the mucous in and around the internal opening and maintenance of all clean tracts. [13]

***VAAFT*:** (*Video-assisted anal fistula treatment*), it's a procedure in which a rigid endoscope is used, where curettage, fistulous tract cauterization and closure of internal orifice are performed, one of its limitations being that there must be an external orifice. [14]

The objective of the present work was to describe the experience in complex perianal fistula treatment in a Public Hospital within the 2018 – 2022 period. We consider the knowledge and handling of this pathology important, even if infrequent, as in untrained hands it has the potential to leave aftermaths such as incontinence and recurrence.

MATERIALS AND METHODS

An observational descriptive, cross sectional, non-probabilistic study of consecutive cases of patients with a complex perianal fistula diagnosis admitted to coloproctology services within the Itauguá National Hospital during the January 2018 through December 2022 period was performed. Inclusion criteria were as followed: adult patients of both sexes with complex perianal fistula diagnosis (according to Table 1) who have complete clinical files, of which 21 fulfill the complex perianal fistula inclusion criteria. The data was harvested into Excel spreadsheets. Confidentiality of individual data was kept, and the result exposition has been guaranteed to be collectively, or in the case that it's shown individually the privacy of the patient is maintained. The limitations were the small size of the sample group, a few incomplete files, the handling criteria of the specialists involved in surgical procedures and finally, despite there being trained professionals in endorectal ultrasound, the cost isn't accessible for all patients and the Itauguá National Hospital doesn't provide said study nor nuclear magnetic resonance.

RESULTS

99 clinical backgrounds were revised from perianal fistula patients, who were admitted into coloproctology services, during the January 2018 through December 2022 period, of which 21 fulfill the complex perianal fistula criteria, which represents the 21.2 % of the total clinical backgrounds revised. In the present revision, 16 patients were male (76 %) and 5 were female (24 %), 3:1 ratio.

The most frequent age range was between 31 to 40 years old. The main consultation reason was anal secretion in 19 patients and anal pain in 2 patients.

As for the fistula type, 17 patients (81 %) presented high transsphincteric fistula, 3 patients (14 %) horseshoe fistula, 1 patient (5 %) extrasphincteric fistula ***(see Graphic 1)***.

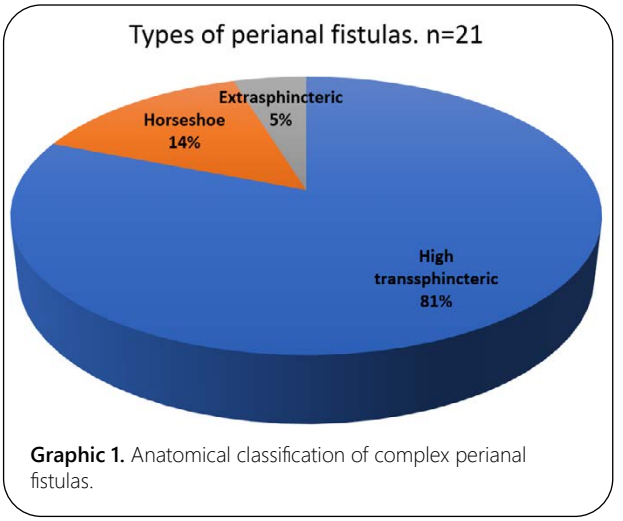
Only in 6 patients a preoperatory colonoscopy was performed.

As for treatment, 10 fistulectomies (48 %), 6 fistulotomy with seton placement (28 %) were performed, and in 5 patients (24 %) only a fistulotomy. All procedures were performed with spinal anesthesia.

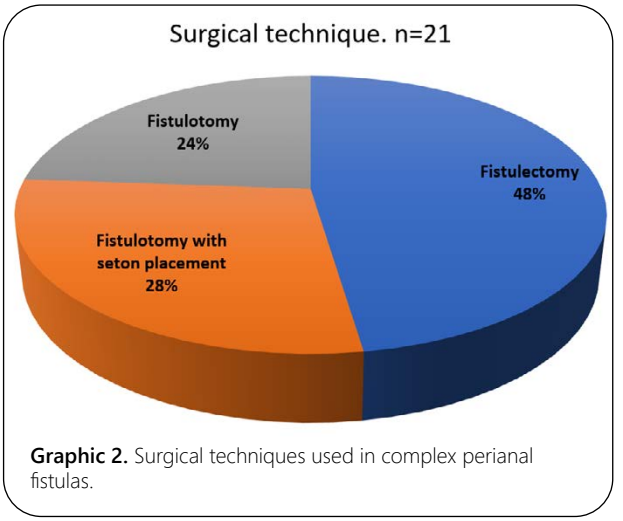
In patients which the seton was left, the average adjustment period was 7 days and the seton fall time was 21 days. The average inpatient time was 3 days (67 %) and 5 days (33 %). Spinal anesthesia was performed on all patients. Sphincter preservation technique was not performed on any patients.

As for complications, only in 2 patients (10 %) fistula recurrence was found, which was treated with seton placement fistulotomy, yielding proper evolution in both cases. Patient follow-up of up to 6 months after surgery was performed, observing only the aforementioned complications (2 recurrences).

Routine anal continence assessment nor incontinence grade scoring were performed on any patients during preoperatory nor postoperatory, continence evaluation was performed alongside interrogation and physical exam within checks up to 6 months after the surgical event, no patient reported gas nor feces incontinence. No mortality cases.



Graphic 1. Anatomical classification of complex perianal fistulas.



Graphic 2. Surgical techniques used in complex perianal fistulas.

DISCUSSION

Perianal fistulas have been a common pathology, but problematic at the same time. Within the studied period regarding sex distribution, it was recorded that 16 patients were male and 5 were female, Carr and col. found an increase prevalence in men rather than women, with a rate of 12.3 cases for every 100.000 and 5,6 cases for every 100.000, respectively. These authors also found that the average age at the moment of diagnosis was of 38 years old, similar data to the present study. [15]

As for consultation reason, the most frequent one was anal secretion, followed up by anal pain, Machain and col. found that the most frequent symptoms were paint, pruritus and secretions. Of them pain represented the main motive of consultation with 41 %; (n=46) followed by secretions with a 32 % of the total (n=36). These authors also found, with relation to the most frequent fistulous tracts were the lower transsphincteric with 46.4 % (n=52) and intersphincteric with 36.6 % (n=41), followed by high transsphincteric with 11.6% (n=13), extrasphincteric and horseshoe were both found in 2.68 % each (n=3). Unlike our series, in which 81 % of patients presented high transsphincteric fistulas, 14 % of patients presented horseshoe fistulas and 5% extrasphincteric fistulas. [16]

Regarding the surgical technique found in the present series, even though the literature is very clear in relation to complex fistulas, in which the sphincter preservation techniques are recommended (advance flap, LIFT, VAAFT, etc.), leaving fistulotomies and/or fistulectomies for simple fistulas, just as Charalampopoulos and col recommend, our series found that all cases were treated with fistulectomies (48 %), fistulotomies with seton

placement (28 %) and only fistulotomy in (24 %). [17]

Subhas and col. in a revision of literature related to the use of setons in anal fistula treatment, found that the cutting seton falls after 30 days, with adjustments every 7 days, unlike our present series, in which it falls after 21 days, with a similar adjustment every 7 days. [18]

In another series Chuang y col., in a study about 112 patients with complex perianal fistula diagnosis, treated through setons, proved that the use of elastic band, adjusted at weekly intervals is safe and effective, with a shorter recovery time for the wound, lower recurrence and less continence disorders. [19]

CONCLUSION

As conclusion we can say that the complex fistula percentage was of 21.2 %. The average age was 35 years old. As for gender the most prominent one was the male sex. The main consultation reason was anal secretion.

Regarding treatment, 10 fistulectomies, 6 fistulotomies with seton placement and in 5 patients only a fistulotomy was performed. Fistula recurrence was reported in 2 patients, which were treated with seton placement fistulotomy. In patients in which the seton was left, the average adjustment time was 7 days and the seton fall time was 21 days. The average inpatient time was 3 and 5 days.

Spinal anesthesia was performed on all patients. Sphincteric preservation techniques were not performed on any patients. No mortality was present in the series. Patient follow-up of up to 6 months was performed after the surgery.

Conflict of interest
Author declares no conflict of interests and respects ethical conducts and proper publication practices.

Author's contribution
Author participated in the creation and design of the work; bibliographic search, data recollection, writing and critical revision in search of important intellectual content, critical revision, and final approval; and agrees to the responsibility of all aspects of the work to guarantee that matters related to the precision or integrity of any part of the work is researched and resolved adequately.

Ethical considerations
Confidentiality of individual data was kept and has been guaranteed that the results will be exposed collectively, or in case of being individual maintaining said privacy.

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Amyand's hernia in a patient with diagnosis of inguinoescrotal hernia

Hernia de Amyand en paciente con diagnóstico de hernia inguino escrotal

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ABSTRACT

Hernia repair in the inguinocrural region is one of the most frequent operations in surgical practice. The cecal appendix may be found contained in the inguinal or crural hernial sac, which is called *Amyand* and *Garengeot* hernias, respectively.

Amyand's hernia constitutes a type of inguinal hernia in which its content is the vermiform appendix. Given the rarity of the condition, it is difficult to unify criteria on the management of this pathology.

Key words: Amyand's hernia; Garengeot's hernia; Appendix; Hernioplasty.

RESUMEN

La reparación de la hernia en la región inguinocrural es una de las operaciones más frecuentes en la práctica quirúrgica. Pueden encontrarse el apéndice cecal contenido en el saco herniario inguinal o crural, lo que se denomina hernias de *Amyand* y *Garengeot*, respectivamente.

La hernia de Amyand constituye un tipo de herniación inguinal en la que su contenido es el apéndice vermiforme, dada la rareza del cuadro resulta complicado unificar criterios sobre el manejo de esta patología.

Palabras claves: Hernia de Amyand; Hernia de Garengeot; Apendice cecal; Hernioplastia.

INTRODUCTION

Inguinal hernia repair is one of the most frequent procedures in surgical practice. However, it can always constitute a technical dilemma for the surgeon, even for those with plenty of experience. They can be met with unusual findings, such as cecal appendix partially or completely contained within the hernial sac, inflamed or not, or present other complications [1-2].

The presence of the vermiform appendix in the interior of an inguinal hernial sac is denominated as an Amyand's hernia. Inguinal hernia contained within the vermiform appendix's incidence is about 0.28-1 %. The presence of an appendicitis within an inguinal hernia's interior is even less infrequent, with a 0.07-0.13 % incidence; and performing a preoperatory diagnosis is exceptional [3].

Historically, Claudius Amyand described in 1735 the presence of a punctured appendix inside an incarcerated inguinal hernial sac; and it was Rene Jacques Croissant de Garengeot who described in 1731 the first femoral hernia intervention containing the non-inflamed appendix. The first appendicitis on a femoral hernia, something even more infrequent, was intervened by Hevin in 1785. Hence the appendix can be found in this type of hernia without alterations, different appendicitis or congestion by incarceration grades, having to use the eponym "Amyand's hernia" to qualify an appendix within an irreducible inguinal hernia and "Garengeot's" to describe the appendix's incarceration within a femoral sac [4-5].

There is no standard protocol for this disorder's management. However, Losanoff and Basson published a guide for the management of Amyand's hernia, which is summarized in Table 1.[6] Factors such as the presence of an inflamed appendix, surgical field contamination, patient's age and anatomical findings of the tissues are important determinators for a proper surgery.[6]

Given that these hernias constitute a historical diagnosis and their confirmation in most cases are stablished during the surgical act, the subject's bibliography is revised with the objective of developing a support material for professionals involved with these rare surgical entities.

CLINICAL CASE'S PRESENTATION

70-year-old male, reported episodes of pain in the inguinal region on several occasions to the emergency services, last one being 5 days ago in the right inguinal region, abrupt origin piercing-type of moderate intensity which irradiates to the same scrotal region, becomes exasperated with physical effort, and yields with rest. Also, a tumor on the scrotal region of insidious origin is added, low and progressive growth, denies fever sensation, feces and gas detention, or other accompanying symptoms.

Refers similar episodes approximately 4 years ago. The physical exam shows a 6 cm tumor on the right scrotal region, painless to palpation with soft, elastic border consistency, non-reductible.

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Table 1. Losanoff and Basson classification for Amyand's hernia management (6)

Classification	Description	Hernia's treatment
Type 1	Normal appendix	Appendix reduction, or Appendectomy. Hernioplasty with mesh
Type 2	Acute appendicitis without peritonitis	Appendectomy through hernia. Repair without mesh
Type 3	Acute appendicitis with peritonitis	Appendectomy through laparotomy. Repair without mesh
Type 4	Acute appendicitis associated to another pathology	Appendectomy. Perform other procedures if necessary. Repair with or without mesh

Blood analysis yield no altered parameters, ultrasound reveals protrusion of abdominal content (adipose tissue with intestinal folds) in the right inguinal canal after Valsalva maneuver, partially reducible, outside the inferior epigastric vessels through wall's continuity solution of 21 mm diameter.

With the right indirect inguinal hernia L3P scrotal variety diagnosis according to EHS classification [5], a hernial sac of 8 cm is found during the surgical act, proceeding to dissection its opening yields cecum slip and no signs of cecal appendix inflammation, the latter being fixed on the sac's edge, a classical Appendectomy is performed, followed by a inguinal hernioplasty through Liechtenstein technique, without additional pathological findings. Postoperative was without incidents.

Pathological anatomy report: hernial sac of fibroadipose tissue with hyalinization sectors. The longitudinal external muscular and circular internal wall, with the central area occupied by fibrosis and adipose tissue. By its morphology this structure is compatible with extreme distal end of cecal appendix with luminal fibrous obliteration. Atypia absence.

DISCUSSION

Amyand's hernia presents usually presents itself as a sensitive, tense, and irreducible mass in the inguinal region, accompanied by several grades of abdominal pain and vomiting. The cecal appendix within the hernial sac's finding is greatly casual in percentage, and from there is when the correct decision must be made about the treatment, based on clinical, laboratorial, and imagery parameters and having the Losanoff classification as backup. Within the present case the Appendectomy was decided due to the appendix's adherence to the hernial sac.

Figure 1. **Panel A:** patient with right L3P inguinal hernia according to EHS classification can be seen. **Panel B:** after appendectomy the adherence to the sac can be observed. **Panel C:** cecal appendix freed from the sac without apparent inflammatory signs.

Conflict of interest
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Author's contributions
All authors contributed to the accomplishment of the present article equally.

Ethical considerations
Taking into consideration the research's ethical principles, no experiments in animals or human beings were done, authors have followed their work center's protocol regarding the publication of the patients' data who have participated and remain anonymous in consideration of confidentiality.

Informed consent was obtained from the patient referenced in this study; this document is held by the work's author.

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Heyde Syndrome: surgical treatment of aortic stenosis associated with lower gastrointestinal bleeding

Síndrome de Heyde: tratamiento quirúrgico de la estenosis aortica asociada a hemorragia digestiva baja

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ABSTRACT

In many cases, the origin of lower gastrointestinal bleeding is not completely elucidated. The highest percentage of cases have their origin in the colon, with diverticula and angiodysplasias being the most common causes. Heyde syndrome is a rare diagnosis that consists of the association of GI bleeding due to angiodysplasia and aortic stenosis, due to a degenerative process and an acquired deficiency of von Willebrand factor type IIA. The objective of this work is to present a rare case of HDB, whose treatment consists of surgery.

Keywords: Lower gastrointestinal bleeding, Heyde syndrome, Von Willebrand factor, Aortic stenosis, Angiodysplasia, intestinal bleeding, Aortic valve replacement.

RESUMEN

En muchas ocasiones, el origen del sangrado gastrointestinal bajo no se dilucida completamente. El mayor porcentaje de casos tiene su origen en el colon, siendo los divertículos y las angiodisplasias las causas más comunes. El síndrome de Heyde es un diagnóstico poco frecuente que consiste en la asociación de sangrado gastrointestinal por angiodisplasia y estenosis aórtica, debido a un proceso degenerativo y un déficit adquirido de factor von Willebrand tipo IIA. El objetivo de ese trabajo es presentar un caso poco común de HDB, cuyo tratamiento consiste en cirugía.

Palabras clave: Hemorragia digestiva baja. Síndrome de Heyde. Factor de Von Willebrand. Estenosis aortica. Angiodisplasia. Sangrado intestinal. Reemplazo valvular aórtico.

INTRODUCTION

Heyde syndrome is an uncommon association amongst aortic stenosis and low intestinal bleeding, which has been a study objective in the past few years, with major advances in its physiological comprehension and treatment.1 In this work, we present a clinical case of Heyde syndrome and its evolution.

CLINICAL CASE

64-year-old male patient reports lipothymia and marked paleness to a low-complexity assistance center's ambulatory consultation, where an acute anemia diagnosis is performed. Requested laboratory studies yield 6.9g/dL hemoglobin (Hb) and 20.7 % hematocrit. Due to the results, a 2-volume concentrated red blood cells transfusion is decided and the patient is discharged with a stool guaiac test order. The following month to said consultation, the patient reports to our emergency services due to a 4-day evolution of several instances of bloody stools of moderate amounts. Does not present lipothymia, weakness, dizziness nor other accompanying symptoms. The physical exam presents vital signs within normal parameters, pale skin and mucous, without systemic repercussions. The exam also presents non-painful soft and depressible abdomen, no defenses nor irritation. Rectal examination yields hematochezia on the glove's fingertip. The rest of the physical exam's data does not provide valuable.

Laboratory studies are requested upon admission, yielding 8.9g/dL Hb, 27 % hematocrit, 8,580/mm3 white blood cells, 66% neutrophils, 270,000/mm3 platelets, 100 % prothrombin time, 0.8mg/dL creatinine, and 20mg/dL urea. The electrocardiogram upon admission yields: R precordial amplitude increase on sinus rhythm, suggesting a left ventricular hypertrophy, no ischemic changes.

An upper gastrointestinal endoscopy is performed which yields a hiatal hernia, chronic gastritis, and erosive duodenitis, while the colonoscopy shows grade II internal hemorrhoids. Upon uncertain diagnosis, a double abdomen and pelvis contrast CT scan is requested, which yields bowel wall thickening of the ascending colon and cecum, suggesting guided study assessment. ***(see Figure 1)***

The patient is discharged stable and in good general state without signs of bleeding, 10g/dL Hb, 32.5 % hematocrit, in plans of having an enteroscopy and an ambulatory-type follow-up.

The patient is readmitted 6 weeks afterwards due to a 3-day evolution of hematochezia in sizable quantities, without systemic repercussions nor other accompanying symptoms. Presents 9.0g/dL Hb and 27 % hematocrit upon laboratory admission.

During inpatient stay, the patient presents bloody stool persistency in great quantities (hematochezia) hence an emergency

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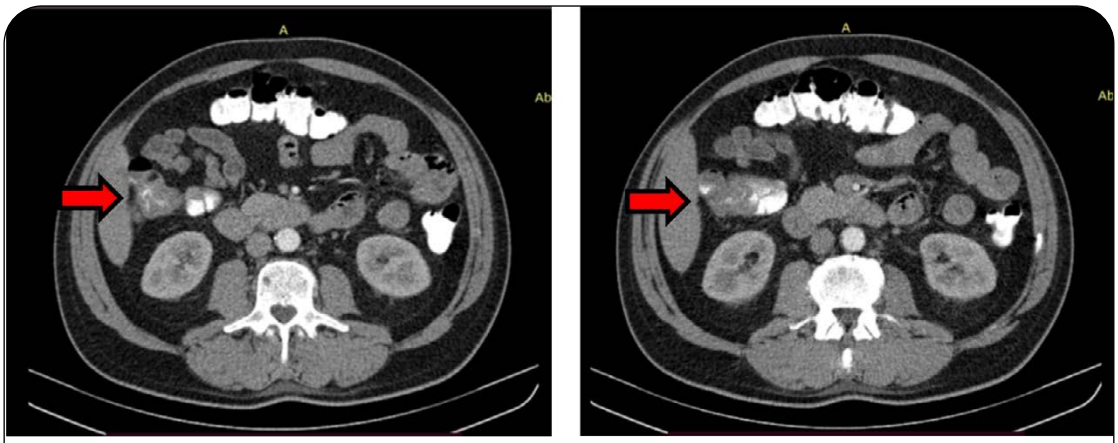

Figure 1. Double abdomen and pelvis contrast CT scan. Focal, asymmetric bowel wall thickening is observed, which compromises the ascending colon.

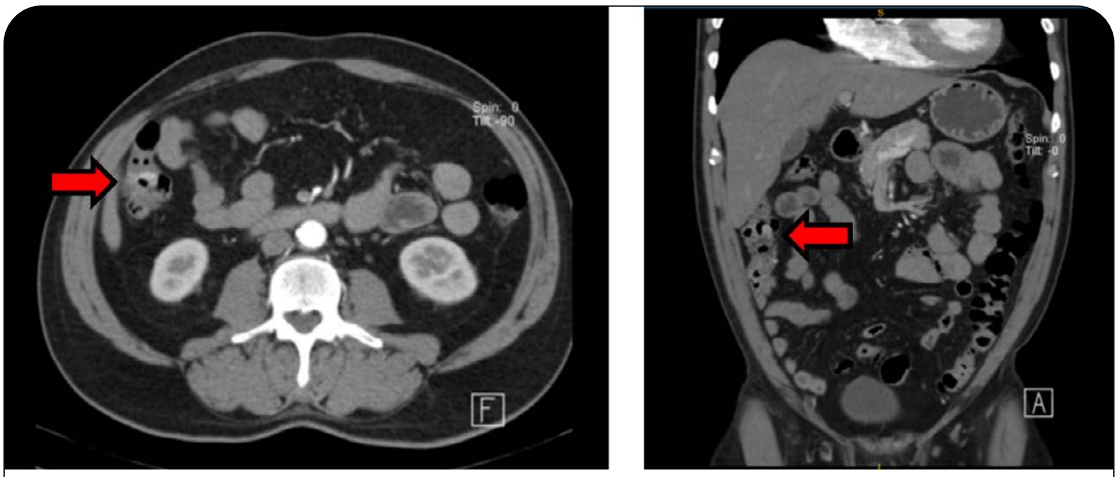

Figure 2. Abdomen angiotomography, arterial phase. Visible focal bowel wall embossment, underneath the colon's hepatic angle. (Left: axial cut / Right: coronal cut)

upper gastrointestinal endoscopy is performed, which did not yield valuable data. The patient remains with hidden digestive bleeding, in plans of having an angiotomography and enteroscopy.

The abdomen angiotomography yields focal bowel wall embossment of the ascending colon underneath the hepatic angle compatible with the first hypothesis through an angiodysplasia. ***(see Figure 2)***

With the probable diagnosis of angiodysplasia and due to the lower digestive bleeding episodes on several occasions, it is decided to prepare the patient for a total colectomy. Amongst presurgical studies, an echocardiogram is requested, which yielded a degenerative aortic valve with severe stenosis, mitral leakage, and slight tricuspid regurgitation.

Due to the gastrointestinal tract angiodysplasia's relation with aortic stenosis, a Heyde syndrome diagnosis is reached, and aortic valve replacement is proposed as definitive treatment. Amongst requested analysis, vWF 160UI/dl antigen (regular value of 50-150UI/dl) and vWF 108UI/dl activity (50-150UI/dl) are highlighted.

The patient was treated with aortic valve replacement surgery 4 months after the diagnosis. Currently showing proper evolution without recurring bleeding to this date.

DISCUSSION

Heyde syndrome was described for the first time in the year 1958 by Dr. Edward Heyde, an intern in Vancouver. It's an association between aortic stenosis and bleeding by intestinal angiodysplasia, the result of an acquired deficit of the von Willebrand factor (vWF) type IIA. Angiodysplasia is a degenerative disease of the intestinal mucous which predisposes gastrointestinal bleeding. Its association with aortic stenosis is well known but Heyde syndrome diagnosis is a rare case. 1 Aortic stenosis usually generates hemorrhagic diatheses due to high-molecular-weight multimers' selective deficit of the vWF. [2]

Heyde syndrome's pathophysiology involves a loss of said multimers with an essential function in hemostasis and an altered platelet function, which favors angiodysplasia bleeding.

In its mature form, the vWF stores itself in Weibel-Palade bodies inside endothelial cells and are gradually freed into the blood flow. It serves a central function in platelet adhesion and aggression, acting as an endothelial damage detector. Likewise, it plays a crucial role in coagulation as it joins and stabilizes the VII factor (F-VII), protecting it from proteolysis and taking it to the injury's location. The proportion in which the different vWF multimers circulate is crucial to maintain a normal hemostasis. The bigger-sized multimers have more proaggregative activity

than the lesser-sized ones, being especially important in high-flux and tangential stress vascular areas, such as angiodysplasias.

From a structural perspective, angiodysplasias are characterized by being arterial or venous capillaries which are dilated and distorted, reaching up to 5mm diameter. These anomalies are located in the gastrointestinal tract's submucous. These malformations' peculiarity is that they lack a middle layer, which results in increased permeability and a notable predisposition to recurring bleeding. Angiodysplasias tend to predominate on the cecum and ascending colon, being able to expand from the cardia to the anal sphincter.

Both the aortic stenosis and angiodysplasias tend to increase their prevalence as age advances. There is proof that suggests that valve disease frequently triggers a hemorrhagic predisposition due to a high-molecular-weight multimers' selective deficit of the vWF [3]. This deficit, known as acquired type IIA von Willebrand disease, favors angiodysplasias' bleeding which coexist within the patient.

Hence the Heyde syndrome's pathophysiology is concisely summarized.

These has been proved in various studies where, after aortic valve replacement, these multimers' levels and platelet functions went back to normal values.

When there is suspicion of Heyde syndrome diagnosis, it's crucial to investigate other possible sources of gastrointestinal bleeding, such as duodenal ulcer, diverticulosis, colon cancer and inflammatory intestinal diseases, among others. In every patient that presents gastrointestinal bleeding, it's recommended to perform a colonoscopy to get a full and precise diagnosis.

When the presence of angiodysplasia and gastrointestinal bleeding is detected, it's important to consider the possibility of an aortic stenosis and actively search this diagnosis to be able to reach a Heyde syndrome conclusion. This diagnosis is confirmed through specific laboratory methods, which allows a precise definition of the disease.

In our case, along with the Heyde syndrome's probable diagnosis a dosage and von Willebrand factor activity study is requested, yielding slightly elevated vWF Ag (160UI/dl of NV of 50-150UI/dl) and vWF Normal activity (108 UI/dl of NV of 50-150 UI/dl).

It's important to highlight that in type IIA von Willebrand disease's case, the vWF Ag dosage tends to fluctuate, it can be normal, slightly decreased or increased. In our patient's case it was slightly increased. This could be explained by the aortic stenosis which causes a blood flow decrease and damage of the vascular endothelium, triggering the release of vWF stored in the endothelial cells, resulting in an increase in vWF Ag levels within the blood flow. Besides, intestinal angiodysplasia, which is a common characteristic in Heyde syndrome, can cause a chronic gastrointestinal hemorrhage. This hemorrhage can lead to a compensatory response from the hemostatic system, including the release of vWF, which can also contribute to the increase on vWF Ag levels. It's important to highlight that high levels of vWF Ag in Heyde syndrome tend to be the result of a physiological response to underlying conditions, meaning, aortic stenosis and intestinal angiodysplasia. However, interpreting these levels must be done with the complete clinical context of the patient and other pertinent diagnostic tests. The decisive analysis for the diagnosis is vWF multimer analysis through electrophoresis.

The most appropriate treatment is valve replacement. [4]

Heyde syndrome is a rare pathology but an entity that we have to be aware of when it comes to evaluate a patient's history of bleeding or anemia, especially when bleeding's origin is not visible on the complementary studies. It's indispensable for the adequate detection and interpretation of its manifestations and appropriate handling of the valve disease, in a multidisciplinary environment.

The most appropriate treatment is the resolution of the aortic valve obstruction. The valve replacement reverts the basic pathophysiological disorder, reversing the subsequential platelet disfunction and angiodysplasias' bleeding coinciding or favored by the aortic stenosis itself.

Ethical considerations
There were no identified ethical issues in the handling of the clinical case. Informed consent was obtained from the patient for the publishing of their medical history.

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Author's contributions
All authors participated on the patient's treatment and elaboration of the manuscript.

Conflict of interests
Authors have declared to not have any conflict of interests related to the present work.

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Cirugía Paraguaya

Generalized peritonitis of appendicular origin with intraoperative finding of situs inversus abdominalis

Peritonitis generalizada de origen apendicular con hallazgo intraoperatorio de situs inversus abdominalis.

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ABSTRACT

Situs inversus is a very rare congenital disease that affects one in 10,000 people. The revelation of *situs inversus* due to a peritoneal syndrome is an extremely rare event. Patients with *situs inversus* have a 0.016-0.024% incidence of acute appendicitis and generally present with some complication. We present the case of a patient with generalized peritonitis of appendiceal origin with a casual intraoperative finding of *situs inversus abdominalis*.

Key Words: Appendicular peritonitis, *situs inversus*.

RESUMEN

El *situs inversus* es una enfermedad congénita muy rara que afecta a una de cada 10 000 personas. La revelación de *situs inversus* por un síndrome peritoneal es un evento extremadamente raro. Los pacientes con *situs inversus* tienen una incidencia de 0,016-0,024 % de sufrir apendicitis aguda y generalmente se presentan con alguna complicación. Se presenta el caso de una paciente con peritonitis generalizada de origen apendicular con hallazgo intraoperatorio casual de *situs inversus abdominalis.*

Palabras Claves: Peritonitis Apendicular, *situs inversus.*

INTRODUCTION

Situs inversus is a rare congenital disease that affects one in every 10,000 people. It's characterized by specular image transposition of the abdominal and thoracic organs. The *situs inversus* diagnosis is generally performed incidentally while researching a non-related medical problem. [1]

Situs inversus' revelation through a peritoneal syndrome is an extremely rare event. *Situs inversus*' patients have a 0.016-0.024% tendency to suffer acute appendicitis and it generally presents itself with some complication. This condition can be completed when both the thoracic and abdominal organs are transposed, or partially when only one of these cavities is affected.

Acute peritonitis in *situs inversus* is a diagnostical dilemma due to the appendix's abnormal position. It's assumed that, even if the viscera is transposed, the nervous system may not show the corresponding transposition, which leads to confusions regarding vitals and symptoms. Hence, regarding the 18.4 to 31% of *situs inversus*' patients, the pain caused by the left acute appendicitis has been reported on the lower right quadrant. [2]

In general, surgical diseases' diagnosis in these patients is delayed due to low clinical suspiciousness, hence why usually these patients are diagnosed in later stages of the disease. In these types of patients, the diagnosis can be based on the physical exam, X-ray, and ultrasound, although CT scans have proved to have increased sensibility and specialty and has become an essential tool to handle these patients. With a male-to-female 3:2 proportion, it has an autosomal recessive presentation, but its precise genetical mechanism is yet to be identified. [3]

The following clinical case is presented with the objective of making the main characteristics of this unpredictable finding known in most of its presentations.

CASE PRESENTATION

43-year-old female patient reports to the emergency services with a 7-day history of evolutive abdominal pain of sudden origin to the left iliac fossa level, stinging and continuous which partially yields with common self-administered oral analgesics and covers the entire abdomen after a few days, also accompanied by vomiting on several occasions and diarrheal stools. Forty-eight hours after their admission the patient presents feces and gas detention. Physical exam yields abdominal distension, little depressiveness, very painful in all quadrants, with peritoneal defense and irritation, increased sonority, and absence of hydro-aerial sounds.

Laboratorial studies yield 10.4gr/dl hemoglobin, 29.8 % hematocrit, 15,000/mm3 leukocytosis, and 88 % neutrophilia. Posteroanterior thoracic and standing abdominal X-rays are requested, which yield hydro-aerial levels and great dilation of the left colonic framework. The thoracic X-rays does not yield pneumothorax, heart in its normal location ***(see image 1 and 2)***.

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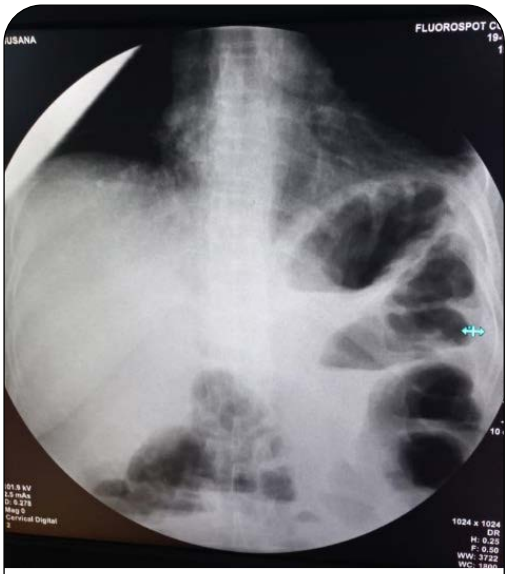

Image 1. Standing abdominal X-ray where hydroaerial levels and colonic dilation of the left side can be observed.

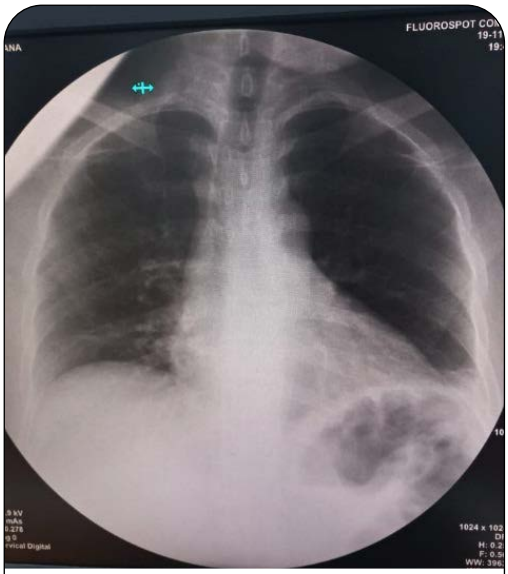

Image 2. Posteroanterior thoracic X-ray with the heart on its normal location (levocardia).

There are no other preoperatory image methods.

An emergency exploratory laparotomy is decided upon, with suprainfraumbilical handling, abundant fetid purulent liquid release is presented immediately after incision, approximately 700 cc suctioned, cavity exploration yields *situs inversus abdominalis*, liver and gallbladder occupying left hypochondrium and stomach and right-side vessel ***(see image 3 and 4)***. Colon exploration yields cecum in the left iliac fossa with cecal appendix characteristically gangrenous, punctured in the middle third and healthy base ***(see image 5)***. Appendectomy and profuse cavity cleanse are performed.

The patient presented proper postsurgical evolution, with antibiotic treatment, being discharged after the fourth postoperative day.

Subsequent anatomopathological report shows cecal appendix with necrosis on both its distal thirds compatible with inflammatory process in all of its extension.

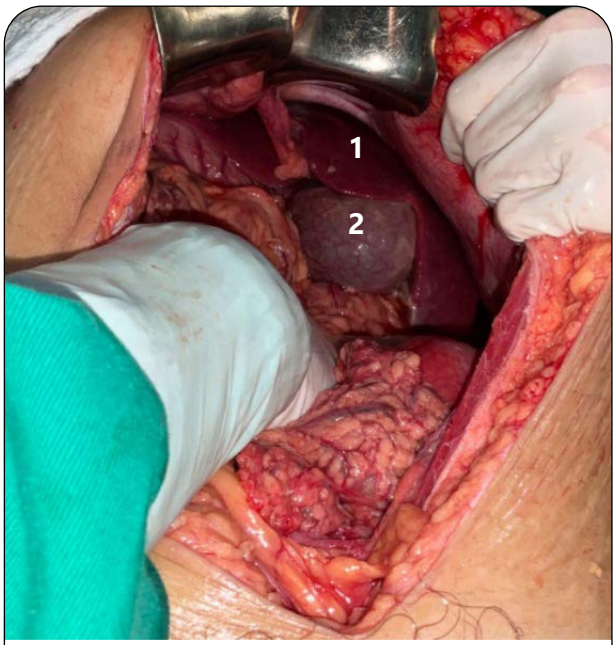

Image 3. Liver (1) and gallbladder (2) occupying the left hypochondrium.

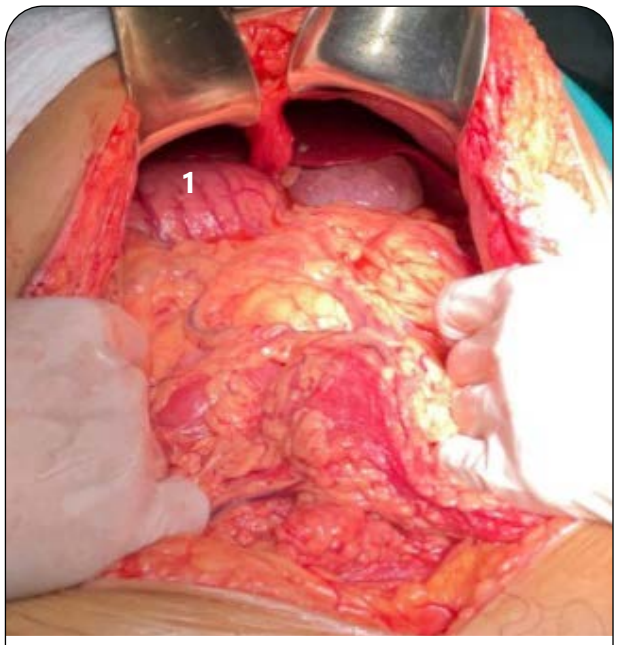

Image 4. Stomach (1) occupying epigastrium and right hypochondrium.

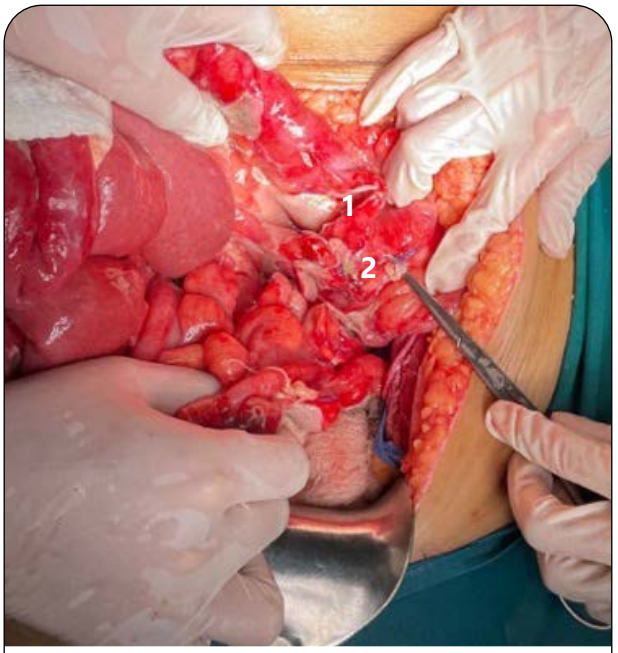

Image 5. *Cecum* (1) occupying left iliac fossa with healthy *appendicular base* (2) prior to appendectomy.

DISCUSSION

Acute inflammatory process' association with *situs inversis abdominalis* is extremely rare. These cases are intraoperative surprises with an 0.001% incidence in operated patients for presumed acute appendicitis. The clinical diagnosis of this type of pathology is hard to establish in absence of a previous image, especially if the symptoms are atypically located. (4)

Currently, the increased availability of imagery methods have proved that *situs inversus* are not exclusive to pediatric age and neither are always associated to comorbidity, on the contrary, they can be presented in adults and as an incidental finding.(5)

Even though in the presented case imagery studies such as ultrasound and CT scan were not performed, the handling and treatment is the same, hence counting on them would serve more as a presurgical guide facilitating the *situs inversus*' diagnosis and diagnostical suspiciousness.

Author's contribution
The author handled the study's design, data collection, draft redaction and subsequent modification and approval of the manuscript.

Conflict of interest
The author denies any conflict of interest regarding the present article.

Ethical considerations
Ethical considerations regarding the presentation of a clinical case were respected.

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Ludwig's angina asociate with acute mediastinitis as a consequence of a non odontogenic infection. Case report. Instituto de Prevision Social

Angina de Ludwig complicada con mediastinitis aguda de origen no odontogénico reporte de caso. Instituto de Previsión Social

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ABSTRACT
The 70-90 % of Ludwig's angina cases are due to dental origin, a lower percentage correspond to cases of sialadenitis, peritonsillar abscesses, and mandibular trauma. There are few reports of non-dental origin cases, making this cause a rare condition. The treatment is similar in both cases: with early diagnosis, directed antibiotic therapy and a good surgical drainage.
Keywords: Submaxilitis, angina, surgical drainage.

RESUMEN
El 70-90 % de los casos de angina de Ludwig se deben a procesos infecciosos de origen dental. Un menor porcentaje se deben a casos como: sialoadenitis, abscesos amigdalinos o trauma maxilofacial. Existen pocos reportes de casos de orígenes no odontogénicos, constituyendo este origen como raro. El tratamiento es similar en ambos casos, teniendo como claves: el diagnóstico temprano, antibioticoterapia dirigida y drenaje quirúrgico oportuno.
Palabras claves: submaxilitis, angina, drenaje quirúrgico.

INTRODUCTION

Ludwig's angina was described in 1836 by Doctor Wilhelm Friedrich Von Ludwig. This being a septic and generally severe process if the mouth's soft tissue which progresses rapidly towards the neck If this infectious process cannot find an exterior drainage, it will indefectibly advance towards the thorax. It's associated with diseases or conditions that affected the organism's defenses such as alcoholism, malnutrition, and organ transplant. But they can be present in patients with no such conditions.[1]

Its diagnosis is especially based on clinical and imagery criteria. Clinical manifestations must be rapidly identified, occurrence of pain in the area, along with lockjaw and tongue elevation, which alerts an early diagnosis.[1]

The infection's advance towards submandibular, sublingual, and submental spaces consequentially causes the collapse of the oropharynx and hypopharynx.[2]

Ludwig's angina is of odontogenic origin in most cases (70-90 %). Dental cavities or necrotizing ulcerative gingivitis are the main causes. But the non-odontogenic causes, such as submaxilitis, peritonsillar abscess or maxillary sinusitis, as well as maxillofacial trauma, represent the other 10 %. Ludwig's angina may bring forth a glottis oedema with necrotizing fasciitis and necrotizing descending mediastinitis as it progresses on, which has an increased mortality rate.[2]

The non-odontogenic origin triggers a similar syndrome, making the diagnosis more difficult and time consuming. The oral cavity's inspection must be thorough and regulated. The infection starts underneath the mylohyoid muscle's crest and progresses to deeper spaces of the neck. The retropharyngeal space, known dangerous space, works as an easy way for propagation to the mediastinum. Mediastinitis is a rare complication which results in the infection's propagation, first to the parapharyngeal space and from there to the retropharyngeal space and upper mdiastinum.[3]

The patient reports painful and septic-type typical facies, with local swelling and breathing difficulty, absence of lymphadenopathy and lack of frank collections are telling of this disease. Indurations with subcutaneous emphysema alongside the neck can be palped. The characteristic thickened neck can also be seen, commonly referred to as "bull neck".[4]

TREATMENT

The treatment is directed to stop the infection's progression with systemic antibiotics of ample specter, covering polymicrobial agents, and after the patient and air way's stabilization, the possibility of surgical drainage is evaluated.[5]

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The surgical treatment's goal is to ultimately drain the collections. Whichever the handling method is, thorough drainage of the collections must be assured. Inadequate access to the neck or lack of deep spaces' drainage would compromise the response to the surgical treatment. It's also important to identify and treat the causing agent of Ludwig's angina. Eliminating the causing factor on the first surgery, is the key to the initial treatment.[5]

Microbiology

The most commonly identified organisms in these types of origin-independent infections are the viridans streptococci. Being these agents, typical on the oral cavity. Negative gram aerobes can also be isolated, such as staphylococcus aureus, including the methicillin-resisting ones which also contribute to the deep neck space infections, especially in children, immunocompromised and other at-risk patients.[5]

The streptococcus anginosus (streptococcus milleri) are a particularly virulent viridans group. Many odontogenic infections may carry oral anaerobes with them such as species of Peptostreptococcus, Fusobacterium nucleatum or pigmented bacteroids (Prevotella melaninogenica and Porphyromonas spp).[6]

The following is a case of a patient who developed Ludwig's angina of non-odontogenic origin.

CASE PRESENTATION

We present the case of a male patient, 67 years old, from the Department of Paraguarí, businessman. Reports arterial hypertension currently in treatment with oral antihypertensives, non-diabetic, neither reports any other systemic pathologies. Former smoker for more than 20 years and moderate alcohol drinker.

Has an 8-day evolution history, localized stinging-type pain in the right submandibular region, which exacerbates and radiates the neck area with the passing of days. Local swelling with temperature changes and local erythema. Reports progressive dysphagia to solid food. The patient explains that the pain increased in intensity, manifesting a lack of response to domestic treatment with common analgesics and warm compresses. Furthermore, shows a decrease of the general state with feverish feeling and progressive dysphagia. The patient reports swelling under the tongue with halitosis, not related to any dental piece.

Arrives at the Hospital of the Social Prevision Institute's emergency department 8 days after the symptoms began, in a normal general state with tachycardia, fever, evident swelling of the submandibular region, and postural respiratory distress ***(see figure 1 and 2)***.

Inspection of the oral cavity yields opening distress (lockjaw) along with white tongue. Dental pieces of the upper and lower maxillary show chronic periodontitis, but with no associated local acute process. Inspection of the mouth's floor yields evident swelling, painful and erythematous, along with fetid and purulent fibrinous exudate over the mucous surface.

Laboratory tests upon admission corroborate a sepsis state with: 26,500/mm3 leukocytosis, 89 % neutrophilia, 13.1g/dL hemoglobin, 37.6 % hematocrit, 235,000/mm3 platelets, 5.58mg/dl total bilirubin, 61 mg/dl urea, 1.45mg/dl creatinine, 136mEq sodium, 4.2mEq potassium, 77 % prothrombin time, PCR +.

Thorax X-ray upon admission: light widening of the upper mediastinum, right pleural effusion with densification of the cardiac area. No air present within the subcutaneous cellular tissue neither within the mediastinum.

CT scans were simple at 24 hours from admission to emergency services. Densification of soft tissue with air within the subcutaneous cellular tissue (submandibular, cervical, and me-

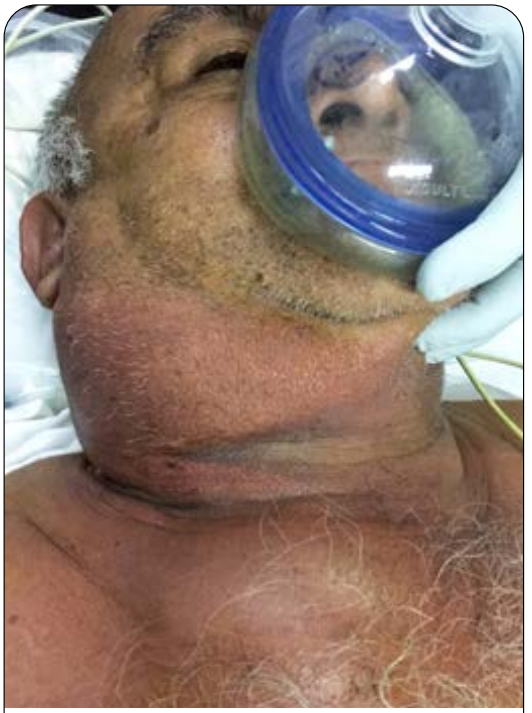

Figure 1. inflammatory signs on the submandibular region and crackles. Patient with respiratory distress and laryngeal stridor.

Figure 2. indurated submandibular area. Collection on the anterior thoracic wall region is observed.

diastinum) was recorded. Deep neck spaces and the thorax's anterior wall with local edemas of the soft tissues were observed to be compromised. Some liquid collections within the submandibular space. Right pleural effusion with densification of the hilar tract, and pneumomediastinum is recorded ***(see figure 3 y 4)***.

A cervical ultrasound is performed, which does not reveal collections, only yields air within the subcutaneous cellular tissue.

The patient is admitted into the intensive care unit where antibiotic therapy with ampicillin plus 3-gram sulbactam every 8 hours begins.

Enters the operating room approximately 24 hours after being admitted into emergency services, where a fibrolaryngoscopy reveals an important glottis enema, with tracheal deviation. Orotracheal intubation is successful.

We perform a classic lateral cervicotomy over the inner edge of the sternocleidomastoid muscle, finding ample muscular and soft tissue necrosis ***(see figure 5)***. We broaden the incision on both directions due to the infection's extension, besides performing a tracheostomy.

We perform a debridement of all affected tissues. In the submaxillary cell we record multiple collections but partitioned in less than 1cm3. Necrosis of the glandular tissue with loss of its architecture. The authors attribute this as the possible origin due to the lack of other possible ones. Despite an exhaustive search of some dental and/or gingival infectious process in regard to the maxillofacial surgery team, no attributing origins to any dental process were found.

Due to the infection's depth and compromising of the scalene muscles, we decided to extract fibers from the brachial plexus. We also recorded necrosis and great affection of the sternocleidomastoid, mylohyoid muscle which we redried. The main stem of the spinal nerve was sectioned and cauterized due to its viability being compromised. We recorded a septic thrombophlebitis of the inner jugular vein confirming Lemierre's triad, whose literature cites a syndrome characterized by bacteremia, thrombophlebitis, and oropharyngeal infection. We proceeded to open the vein through a thrombectomy and double ligature of its edges.

We performed a cleanse of the surgical wound with an iodine solution and left chlorhexidine and oxygenated water com-

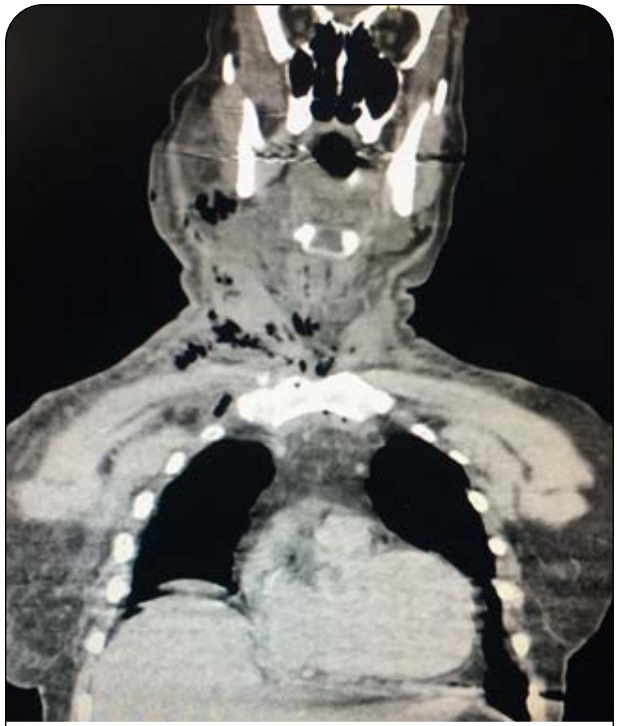

Figure 4. Extension of the infection to neck spaces with anterior pneumomediastinum.

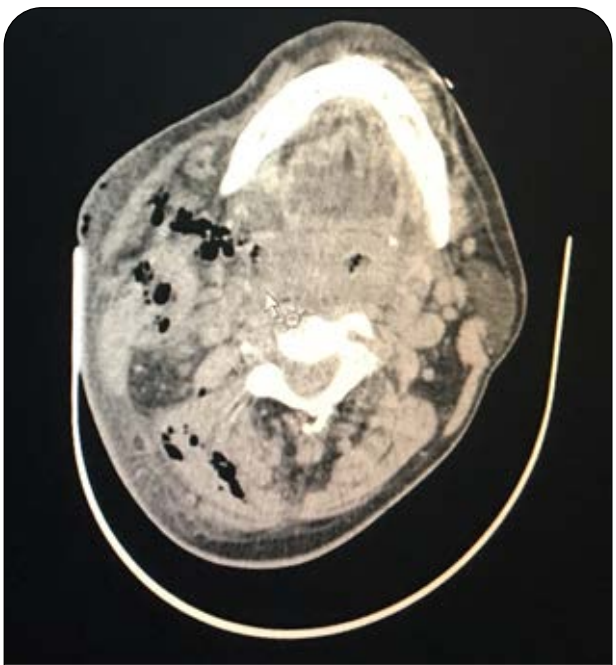

Figure 3. Air bubbles on the simple CT scan showing dissection of the planes and masseteric, pre-vertebral, and vascular spaces' invasion.

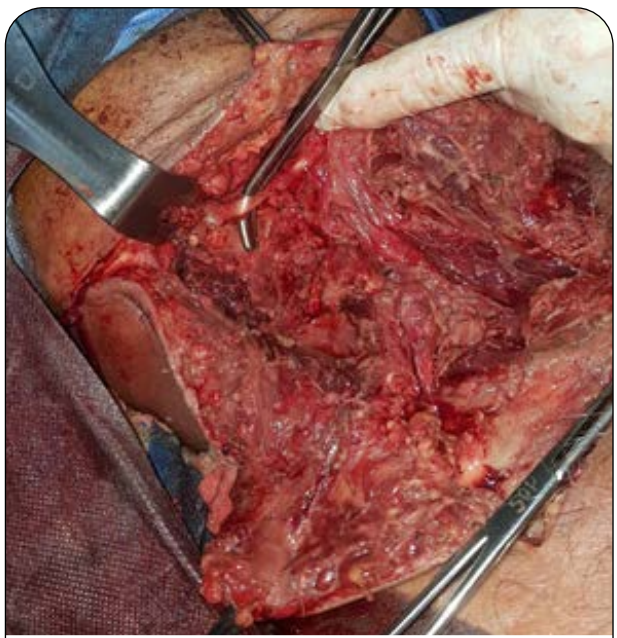

Figure 5. Ample lateral cervicotomy. Abundant gangrene and fetidness of the tissues. Handling was broad, enabling access to all neck spaces. Dissection pattern of the tissues from the neck to the anterior mediastinum was noted.

presses. The skin is handled by separated stitches for a second look of the neck ***(see figure 6)***.

After 24 hours we proceeded to reinspect the neck and placed Jackson-Pratt-type aspirational drainages. We recorded purulent material egression from the mediastinum, by which along with thorax surgery we opted for an anterolateral thoracotomy.

In this operatory act, 48 hours after the patient's admission, we performed a thoracotomy through anterolateral handling. We proceeded to the toilette and pleuro-pulmonary decortication due to a thickened pleura, and dense pleural and purulent liquid. We placed a pleural drainage tube and retro sternal tube for cleansing.

After 72 hours from admission, and on the third surgery. We noted right collarbone's compromission with purulent material egression, by which we proceeded with its dislocation and exclusion.

The patient did not require another surgical intervention. However, through daily follow-ups and exhaustive cleansing of the operatory wound, we achieved good results.

The retrosternal tube for cleansing was utilized for 5 days, for cleaning with a physiological saline solution, it was then withdrawn. The pleural drainage tube was withdrawn on the 35th day after its placement.

Secretion farming: Streptococcus constellatus, Staphylococcus Aureus. Received antibiotic therapy of ample specter with tigecycline for 14 days, and Meropenem and Vancomycin for 20 days directed to positive Cocos and Bacilos Gram.

During in-patient time in the intensive care unit, the patient acquired a pneumonia associated to the respirator by Acinetobacter baumannii and colonization by Klebsiella pneumoniae, carbapenemases producer. The patient also developed a neck-leveled auto-limited pleuro-atmospheric fistula which did not require surgical treatment ***(see figure 7)***.

Major neck asymmetry with akinesia was recorded. Trapezius and deltoid muscle atrophy with non-incapacitating postural deviation.

Patient was discharged after 63 days of in-patient stay, and presents a 9-month follow-up up to this date ***(see figure 8)***.

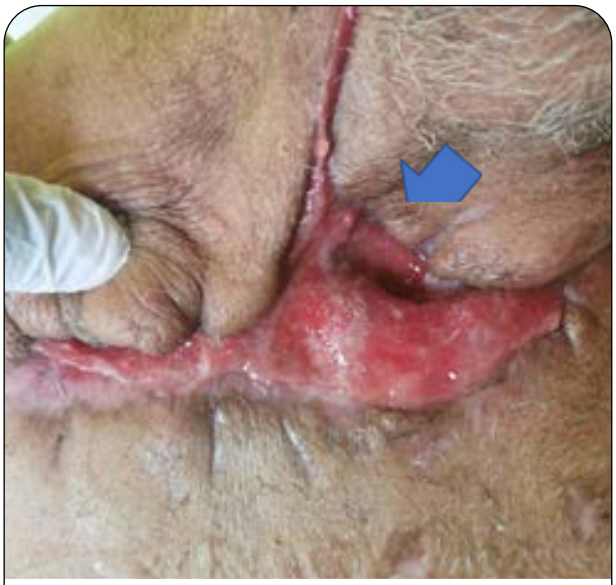

Figure 7. 30th day after the surgery. Proper lesion characteristics which heals on second occasion. Pleurocutaneous fistula's orifices is shown, it was treated with a pleural drainage tube in aspiration until its total remission after the 5th day.

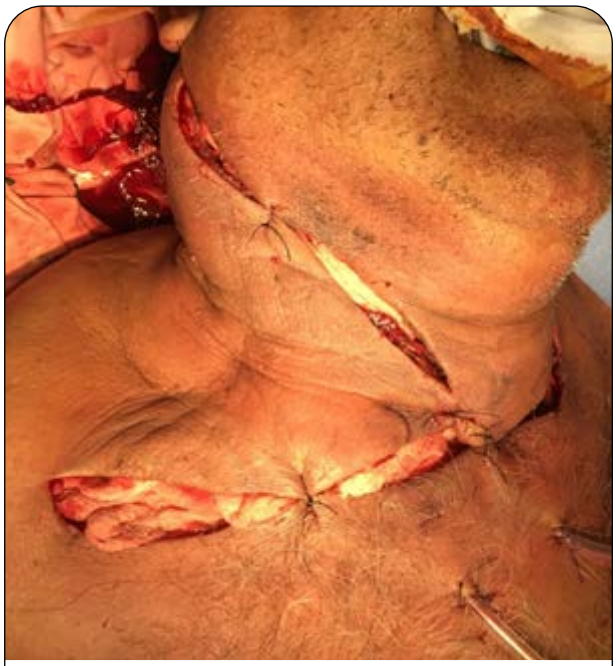

Figure 6. Post-operative look with impregnated compresses of oxygenated water and chlorhexidine. Drainages on continuous aspiration were placed.

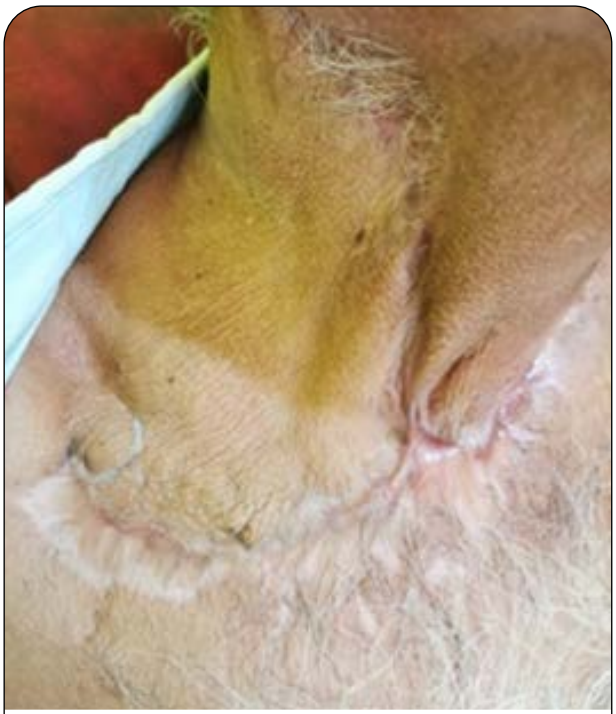

Figure 8. Look after 9 months. Total epithelialization of areas which healed on second occasion.

DISCUSSION

Ludwig's angina had a mortality close to 100 % in the pre-antibiotic era, with the arrival of antibiotics its mortality decreased to a 50 %. However, its associated mortality to mediastinitis still reaches 70 %.[5]

According to Flynn, the ideal surgical technique must assure access and communication between the involved subaponeurotic spaces assuring the change in atmosphere between the anaerobic and aerobic flora and be able to maintain enough time for the proper placement of drainages.[6]

We have seen patients in our practices who have been insufficiently drained, compromising their favorable evolution.

We debated with the authors of this work the "true" origin of this case. The surgical finding of a necrotic submaxillary gland with pus in its interior and the absence of other possible origins plus the isolated farming of a Viridans (Constellatus) species leads us to conclude the acute submaxilitis as the causing entity.

Post-operational follow-up of this type of patients is fundamental, with critical medicine, infectiology, and surgical equipment, farming must be varied to better adjust to the best possible antibiotic. The keys to the treatment center around 3 fundamental pillars: 1- general support measures. 2- antibiotic treatment of ample specter and 3- proper surgical treatment.[7]

Conflict of interest

Authors declare no conflict of interests.

Ethical considerations

Informed consent: informed consent was obtained from the patient for the presentation of this case.

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Author's contribution

Dr. Marcelo Samudio and Dr. Viviano Jara conceived the idea and worked on the development of the study. Dr. Feltes in bibliographic research and Dr. Schaerer in the final revision of the manuscript.

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