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CASE REPORTS

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SPEECH-LANGUAGE PATHOLOGY

Case reports rose to popularity around the same time as the creation of periodical journals, and are the greatest source of evidence to support the majority of clinical practices in speech-language pathology. There has been an overwhelming increase in publications of case reports in the last decade at the international level. Even when their contributions to evidence-based practices are called into question, case reports continue to be an essential part of science and contribute in a significant way to the formulation of hypotheses that can lead to more complex studies.

Keywords: *Case reports, speech-language pathology.*

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INTRODUCTION

The rise in case reports in speech-language pathology coincided with the creation of periodical journals. At the end of the 1930s, one of the most widely known associations in the world, the American Speech-Language-Hearing Association (ASHA[1]), had already published its first case report in the journal that was subsequently rena-

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1 ASHA published its first journal in 1936.

med the Journal of Speech Disorders, known today as the Journal of Speech, Language and Hearing Research. This report dealt with the case of a subject who had benefitted from a treatment focused on improving a pronunciation problem caused by the use of a dental prosthesis (1).

From that moment the publication of case reports gradually increased, though the last decade has seen the most dramatic increase of all. The ASHA's four journals[2] published approximately 288 case reports in the last century (1937-1999), and have continued to publish 569 more in the first 14 years of the 21st century alone (2). Certainly, this increase in case reports (and more generally, of publications) is to be expected from developed countries where speech-language pathology research advances at a much greater pace. This phenomenon does not tend to occur in countries like Colombia for a variety of reasons—among others, the lack of a tradition in publishing the results of interventions by professionals as well as the limited existence of specialized journals in the field.

However, the low publication rate in Colombia is not directly proportional to the actual practice of case studies. Within the schools of speech-language pathology, case studies are a common and routine practice that are built into programs of study. Professionals in training are exposed to case studies in a variety of contexts, both in courses dedicated exclusively to those case studies, as well as in professional practice situations where they select their own patients. These practices are designed to structure the evaluative-diagnostic processes as well as the speech-language pathology treatment process itself. All of this encourages assertive decision-making and fosters scientific thought as part of a systematic review of scientific literature.

THE CONTRIBUTIONS OF CASE REPORTS TO SPEECH-LANGUAGE PATHOLOGY

Discussing the contributions of case reports to speech-language pathology and the weight they hold in professional decision-making leads us directly to the topic of evidence-based practices (EBP). From the beginning, these EBP have tried to evaluate clinical interventions to prove their efficacy and demonstrate their effectiveness in clinical contexts (3). These are all part of research publications, with a clear preference for studies with randomized clinical trials and meta-analysis, which offer the possibility of testing the hypotheses behind treatment results (4). As such, case reports have been considered as a form of low-level research which often do not go beyond anecdotal reporting (5). This means that there is a lack of experimental control, and one is then unable to infer changes produced in subjects as a result of therapeutical interventions (6).

However, the results of clinical trials and meta-analysis should not be overestimated (7). EBPs are useful in different ways and use scientific evidence differently depending on the profession and the level of development they hold. In the case of speech-language pathology, which still lacks definitive scientific evidence and is relatively new in Colombia[3], EBPs are on a basic level in the construction of knowledge, that is, in an initial phase of documenting the results of interventions (3).

2 Journal of Speech, Language, and Hearing Research; Language, Speech, and Hearing Services in Schools; American Journal of Speech-Language Pathology and American Journal of Audiology.

3 In 2016, it will be 50 years since Speech-language therapy profession was established in Colombia.

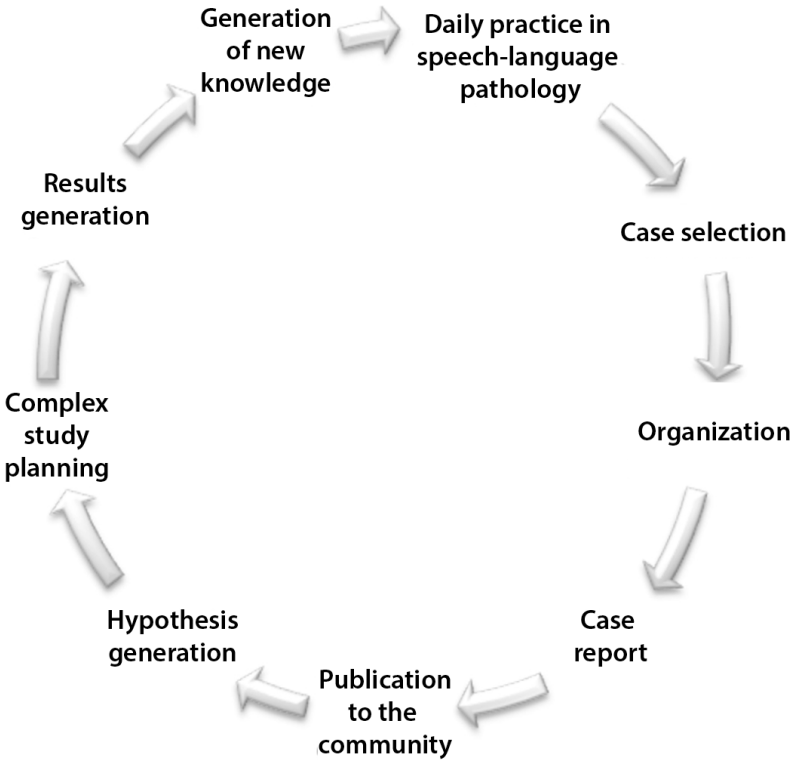

Fig 1. Transition of case reports to the generation of scientific knowledge

Furthermore, many of the studies with a random group design are not appropriate nor sufficient in evaluating interventions as the people who use the services of speech-language pathology are all different and have complex difficulties that affect more than one aspect of communication (3), which is why it would be more sensible and informative to base clinical practice on case reports (8).

In this way, the evidence that supports the majority of speech-language clinical practices is based on studies that are in phases I and II. They are based on clinical or case reports that show the potential effects of treatments and those that have no secondary effect (9). Therefore, these case reports are becoming increasingly more relevant in the field of speech-language pathology, contrary to what has occurred in other health professions (10).

TRANSITION FROM CASE STUDIES TO THE GENERATION OF NEW KNOWLEDGE

Case reports are necessary to support EBPs; however, it is important to remember that they have a mainly informative value. They are an important source in formulating hypotheses about specific problems regarding human communication, so they should become the base to justify more complex study (11). In this way, case reports continue to be a fundamental piece in generating knowledge and helping show to the community of professionals and patients how these interventions work. Figure 1 shows the place case reports occupy in the EBP cycle and how the clinical practice of speech-language pathology feeds into research and how research feeds into clinical practice.

Evidently, to generate and apply the knowledge gained by EBP, speech-language pathologists need to develop certain basic skills, such

as an efficient approach to literature search and critical reasoning when analyzing published studies; the latter has the purpose of determining if the reported interventions are legitimate, or rather if they are, in the words of Lof (12), mere folk medicine.

The systematization of case studies and their reporting in scientific literature will help advance the development of complex studies that allow for the validation of hypotheses and a generalization of results, with the logical effect of providing peace of mind to the academic and professional comunity of the efficacy of treatments.

CONCLUSION

Case reports continue to be relevant in the generation of new knowledge today. In speech-language pathology, they are the greatest source of information that supports practices based on evidence; even if they are widely criticized, it is necessary to continue their publication for the benefits they bring to clinical practice. Furthermore, case studies are a routine practice in academia and in the training of speech-language pathologists, so it is essential to generate a larger number of publications in specialized journals. Nonetheless, it is also important to raise the level of case reports to begin to publish studies that are more complex to confirm the hypotheses of therapeutic interventions.

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THE CLINICAL CASE: A STRATEGY FOR EFFECTIVE LEARNING

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There are different teaching strategies for ensuring significant learning, including research seminars, workshops, directed independent reading, the clinical case, the problem-based learning methods, in-depth lines of study, etc.

In many departments of the clinical area, such as the Department of Pediatrics, a clinical case taken from real life is used as an effective learning method. It consists of providing the case to all participants (students, professors, pediatricians) for study and analysis. This study is discussed in small groups and the solution or possible responses are presented in a meeting scheduled for this purpose.

As its main objective, this type of strategy aims to stimulate participants to interpret the information provided, integrate and apply the knowledge that they possess in order to solve a problem. In addition, it encourages research and the exploration of different articles in indexed journals with the goal of strengthening their response —or of contrasting it with responses of the authors—, of comparing their solution with those of other groups, of acquiring new knowledge, and also of learning to work as part of a team.

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Clinical cases aim, among other things, to achieve the following:

- To make students aware of an unusual presentation of a frequent disease. For example, a child that seeks attention for abdominal pain as a consequence of basilar pneumonia, which could skew the diagnosis.
- To present an infrequent case for which the diagnosis is not easy, and review the topic, such as congenital tuberculosis.
- To analyze a clinical syndrome that has different etiologies, and to learn how to arrive at a correct diagnosis, as is the case of the whooping cough.
- To present a new diagnostic procedure.
- To be aware of psychosocial aspects that may be fundamental in disease prevention, as may be the case of child abuse.
- To use a case to critically review the literature that exists on the disease.

Because of their high academic value, these cases are chosen purposefully for their oral and written presentation in such a way that the student may practice and strengthen their ability to write scientific text for publication.

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COMPLICATED CONGENITAL MALARIA DUE TO PLASMODIUM VIVAX

ABSTRACT

Congenital malaria is a disease that appears in the neonatal period and that, if not treated in a timely manner, may have fatal consequences for the newborn. According to statistics published in The State of the World Children 2009 Report, 3.7 million children under the age of 28 days die annually around the world at present. 8% of cases correspond to children under 5 years of age with malaria (1). Similarly, studies in endemic areas have reported incidences of congenital malaria of between 0.83 and 5.93% (2). Here, we present a case of congenital malaria in a one-month-old nursing infant whose mother received treatment for malaria from Plasmodium vivax (P. vivax) during gestation but suffered a relapse with a consequent compromise of the infant in utero. There is a need to recognize the high prevalence of this disease in our context and to know how to monitor and treat the disease in special cases like those of gestating mothers and newborn infants with congenital infections.

Keywords: *Congenital malaria, Plasmodium vivax, relapse, complicated malaria, treatment.*

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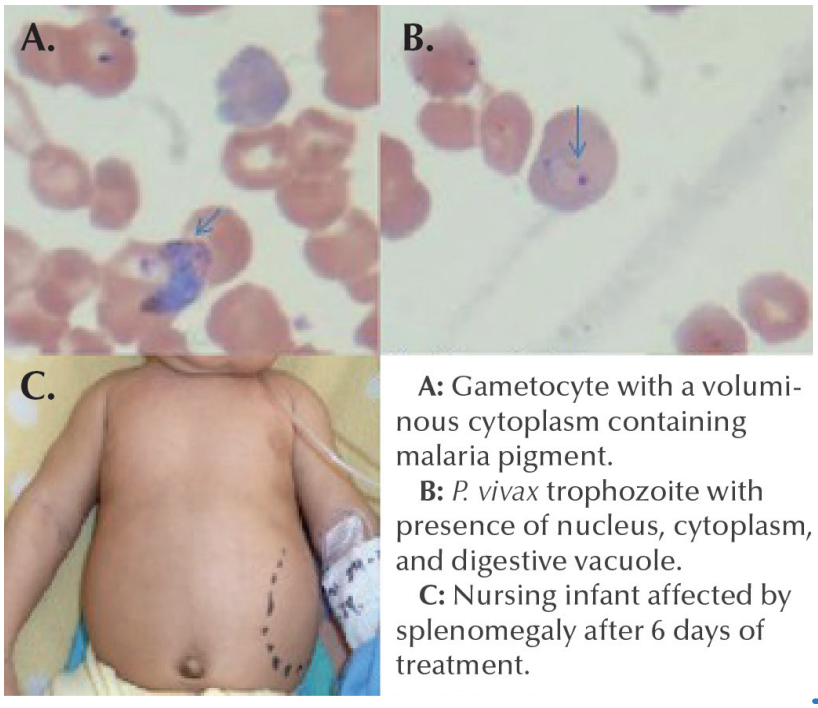

A: Gametocyte with a voluminous cytoplasm containing malaria pigment.
B: *P. vivax* trophozoite with presence of nucleus, cytoplasm, and digestive vacuole.
C: Nursing infant affected by splenomegaly after 6 days of treatment.

Fig 1. A: Gametocyte with a voluminous cytoplasm containing malaria pigment. B: P. vivax trophozoite with presence of nucleus, cytoplasm, and digestive vacuole. C: Nursing infant affected by splenomengaly after 6 days of treatment.

Colombia is recognized globally as the country with the second highest level of biodiversity after Brazil. This condition gives the country the characteristic of endemicity for many tropical diseases that affect a large portion of the population across the national territory and that can be fatal if they are not properly managed in a timely fashion.

Malaria has a high prevalence in our context. One quarter of Colombians inhabit zones where there is a risk of contracting the disease, with a endemicity of 75% in areas under 1500 meters below sea level (3), Nevertheless, the congenital presentation of the parasitosis due to Plasmodium vivax is very infrequent. Here, we present a case of congenital malaria complicated by *P. vivax* to warn clinicians about this entity.

CASE REPORT

A 54 day old patient at the time of the consultation, son of a gestating mother native to and coming from La Chorrera-Amazonas department. The mother had moved to Bogotá, the capital of Colombia, located at around 2600 meters above sea level in a non-malarial zone, since the vector of the parasite is not viable at this altitude. The gestating mother visited a doctor in the third trimester of gestation, and on the second day of her stay in the city, she started to experience general discomfort, fever, headache, and nausea, symptoms that led her to seek medical attention at another institution. Malaria due to *P. vivax* was diagnosed at 32 weeks of gestation and she was treated with oral chloroquine. A post-treatment check-up was performed at 35 weeks of gestation in which a negative thick drop test was found. Therefore, prenatal check-ups continued as normally.

The infant was born in that institution at 38 weeks of gestation by cesarean section due to an unsatisfactory fetal state during labor that made an emergency C-section necessary.

The newborn showed a spontaneous neonatal adaptation, an Apgar score of 8-9-9, and a weight of 3315 grams. The mother

suffered from postpartum endometritis, which required curettage. She showed satisfactory progression and was discharged with primaquine treatment for 14 days in the postpartum period. There were no further check-ups.

The infant was admitted to hospital with a presentation of fever measured at 39°C and irritability associated with diarrheal semi-solid stools, and the vomiting of food on 4 occasions. In the physical examination, the following was found: Heart rate at 168 beats per minute, respiratory rate at 42xmin, blood pressure at 82/46xmin, temperature at 39°C. Weight at 3.8Kg. The infant appeared pale with first degree jaundice and third degree dehydration, the liver palpable at 2 cm below the edge of the ribs. Fluid resuscitation was initiated with crystalloids at 20 mL/Kg. 2 boluses were required. The laboratory reported the following in the CBC: Leukocytes: 7930, lymphocytes: 48.9%, neutrophils: 24.4%, eosinophils: 13%, hemoglobin: 3.6g/dL, hematocrit: 11.6%, platelets: 65 000, C-reactive proteint 271.4, and the thick drop test was positive for *P. vivax*. The patient required a red blood cell transfusion and antimalarial treatment for complicated malaria was initiated with clindamycin and quinine dichlorohydrate over 7 days. The patient was monitored in the intermediate care unit and showed a satisfactory progression, with resolution of the fever after 24 hours of antimicrobial management, control of tachycardia, reduction of jaundice, and persistence of splenomegaly. Prior to discharge, the thick drop test was performed with a negative result.

DISCUSSION

The diagnosis of malaria in the mother during gestation was correct and timely since there was a high degree of suspicion due to her origin in an area endemic for the disease and her general symptoms that, although they are unspecific and can point to any febrile process, must be taken with the clinical context so that proper diagnosis and treatment can be made in order to reduce complications in the maternal-fetal unit. The treatment of gestating mothers with malaria caused by *P. vivax* should be limited to chloroquine. This is due to the fact that primaquine, which eradicates P. vivax tissue schizonts and hypnozoites, is counter-indicated during gestation and breast-feeding due to the risk of generating hemolytic anemia in the infant(4). As such, these parasitic forms can remain dormant, and, therefore, patients that are gestating or breast-feeding merit monthly monitoring with the thick drop test over the period in which *P. vivax* relapses are most frequent (2-6 months post-infection) (5). This is to evaluate relapses that require new rounds of treatment until the hypnozoites can be eradicated when breast-feeding has ended. Likewise, the newborn should be monitored with the thick drop test during the first month of life. If the parasitemia is detected, chloroquine should be administered again.

However, the child may develop complicated congenital malaria due to *P. vivax*. The incidence of congenital malaria in endemic zones is low, between 0.01 and 1.4% of live births. This is due to the passage of anti-plasmodium antibodies through the placental barrier and to the lower susceptibility of infection by this agent shown by fetal erythrocytes that have fetal hemoglobin (4).

Malaria is considered to be congenital when it develops within 7 days following birth in endemic zones, and within up to 2 months in non-endemic zones, as in this case (4). The most frequent agent is *P. falciparum*. Nevertheless, cases of infection by *P. vivax* have been described (6). The clinical manifestations are fever (100%), splenomegaly (93%), hepatomegaly (84%), anemia (85%), irritability (85%), vomiting (79%), jaundice (79%), and diarrhea (65%)(2,4), all of which were present in our patient. In this case, the alteration of the state of consciousness, the hemodynamic instability, and the severe anemia also define the diagnosis of complicated malaria, which is also more frequent in infections of *P. falciparum*. Mortality due to congenital malaria is 1%, but this increases to up to 20% if the patient has complicated malaria.

The treatment for non-complicated congenital malaria due to *P. vivax* is chloroquine. The treatment from complicated malaria in infants under 6 months of age is quinine with a loading dose of 20 mg/Kg IV in a DAD syringe at 5% or 10% to be infused over 4 hours, followed by a dose of 10mg/Kg every 8 hours to be infused over 2 hours. The medication should be administered orally once the patient can tolerate it over 7 days. In infants over 1 month, clindamycin at 20mg/Kg/day is added in 3 to 4 doses for 5 to 7 days (3,7) .

CONCLUSION

It is important to have a high degree of suspicion for tropical diseases given the context in which we live that has a high prevalence of malaria. In addition, physicians should be aware of the special treatment to be administered to infected gestating mothers and the monitoring to be conducted, both of her and the newborn, since there is a possibility of congenital infection, even when the mother has been adequately treated. Monitoring of the mother should be done monthly over the entire gestation period and will end once primaquine can be administered to eradicate dormant parasitic forms that reactivate the disease, and that can lead to consequences for both the mother and the newborn.

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SINGLE PORT LAPAROSCOPIC CHOLECYSTECTOMY IN SITUS INVERSUS TOTALIS

CASE REPORT

SUMMARY

Symptomatic cholelithiasis is a highly prevalent disease in Colombia, though its presentation in patients with *situs inversus totalis* has rarely been described. This report describes the case of a patient with symptomatic cholelithiasis and *situs inversus totalis* who underwent a single-port laparoscopic surgery after concomitant gallstones were ruled out. This procedure is a minimally invasive technique of SILS (Single Incision Laparoscopic Surgery).

Situs Inversus is a rare genetic disorder which is characterized by the transposition of the abdominal and chest organs through the sagittal plane, having a so-called "mirror image" effect. While the etiology is still unclear, it is often attributed to a genetic predisposition caused by an autosomal recessive gene of incomplete penetrance. The presence of symptomatic vesicular lithiasis in a patient with SIT not only creates diagnostic doubt but also presents a real challenge for treatment as any surgical procedure (especially laparoscopic cholecystectomy) will be more difficult due to the anatomical abnormalities of the patient.

Keywords: *Single-port, cholelithiasis, Situs Inversus*

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CLINICAL CASE

A 24-year-old female caucasian patient, homemaker, with a clinical history of several months of biliary colic associated with intermittent episodes of subjective acholia and jaundice, was admitted to hospital after her condition had worsened considerably in the previous eight hours. She was suffering from pain in the epigastric region and left hypochondrium, associated with multiple emetic episodes. Her vital signs were normal and she had no signs of peritoneal irritation or palpable abdominal masses. The physical exam showed no abnormalities. Blood test results revealed counts of leucocytes: 12100, neutrophils: 79.9%, hemoglobin: 13.8, hematocrit: 40.5, platelet: 641000, ALT: 47, AST: 33.8, amylase: 83, alkaline phosphatase: 606, BT: 1.9, BD: 1.6 and BI: 0.3. A full abdominal ultrasound showed cholelithiasis with cholecystitis, and *situs inversus totalis*. A magnetic resonance cholangiopancreatography (MRCP) was conducted, which revealed a normal bile duct, cholelithiasis, and *situs inversus totalis.*

HISTORY

- Pathological: *situs inversus totalis*, spastic colon.
- Pharmacological: none stated
- Surgical procedures: none stated
- Allergies: none stated

The patient was explained the benefits of the single-port procedure as a minimally invasive alternative as well as the associated risks (morbidity and mortality), focusing on the possibility of bile duct injuries and postoperative hernias. The patient then gave her informed consent to San José Hospital in the city of Bogotá.

SURGICAL TECHNIQUE

A single-port laparascopic cholecystectomy was performed by making a single longitudinal umbilical incision of approximately 2cm. The umbilicus was detached, in which a single port was inserted (SILS®, Covidien), using one trocar of 10 mm for the camera and 2 trocars of 5 mm for the clamps. The surgeon was positioned between the legs of the patient and the assistant at the right of the patient as the dissection of the cystohepatic triangle was initiated. The LIGAMAX® instrument was used for the cystic duct and artery. Later, a hemostatic check was performed and the vesicle was extracted through the port (the umbilicus). Then, the aponeurosis was closed with absorbable suture and the skin was closed with non-absorbable suture.

Surgical time was approximately one hour, without complications. The procedure can be viewed here:
http://youtu.be/GumF_1WLFNM.

The next day, the patient showed tolerance of oral intake and was subsequently released from hospital without complications. Post-operative tests for the first month were successful as well.

DISCUSSION

Situs Inversus Totalis (SIT) is the one of the rarest congenital conditions. Associated with compromise related to surgery, the condition was reported for the first time in a human be-

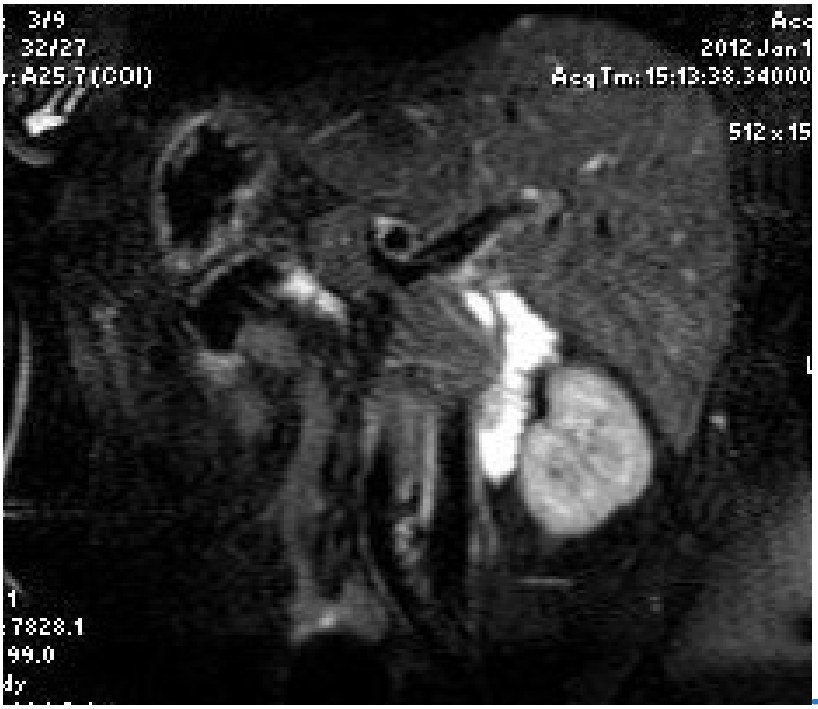

Fig 1. MRCP (Magnetic Resonance Cholangiopancreatography)

ing by Fabricius in 1600, though its etiology remains unclear. In SIT, patients present with dextrocardia and the complete transposition of abdominal viscera, which is often referred to as "mirror-image". This condition is present in the general population between 0.002 to 0.01%, with a slightly higher prevalence in men. While life expectancy is not affected nor is it associated with compromise related to surgical conditions, it is associated with other cardiovascular conditions such as the Tetralogy of Falot, or the transposition of the great vessels.

There is no current evidence of a predisposition to vesicular lithiasis in SIT patients, but this condition often delays the diagnosis, and can even lead to an erroneous diagnosis or changes in the treatment of patients with this condition and their complications.

Situs inversus presents a big challenge in particular to surgical procedures, and in our case, to the laparoscopic approach. There are great technical difficulties as a surgeon works in two-dimensional surgical fields and with mirror images.

In the various laparoscopic procedures in patients who undergo four-port surgery, the right-handed surgeon often has the most difficulty as it involves changing position to be on the left side of the patient, contrary to classical open techniques. In our case, the single-port technique with a midline approach and the surgeon positioned between the legs of the patient made for a more comfortable dissection of the vasculo-biliary structures. However, the one-port technique is complicated by the difficulty in triangulating the instruments, a basic component of minimally invasive surgery.

Therefore, the proper positioning of the surgeon in relation to the patient is vital. In our case, thanks to the surgeon's previous knowledge of *situs inversus totalis*, the necessary position between the legs of the patient was assumed and surgery through a single port was performed, dissecting towards the opposite side of the patient with the special anatomical challenges of the patient in mind.

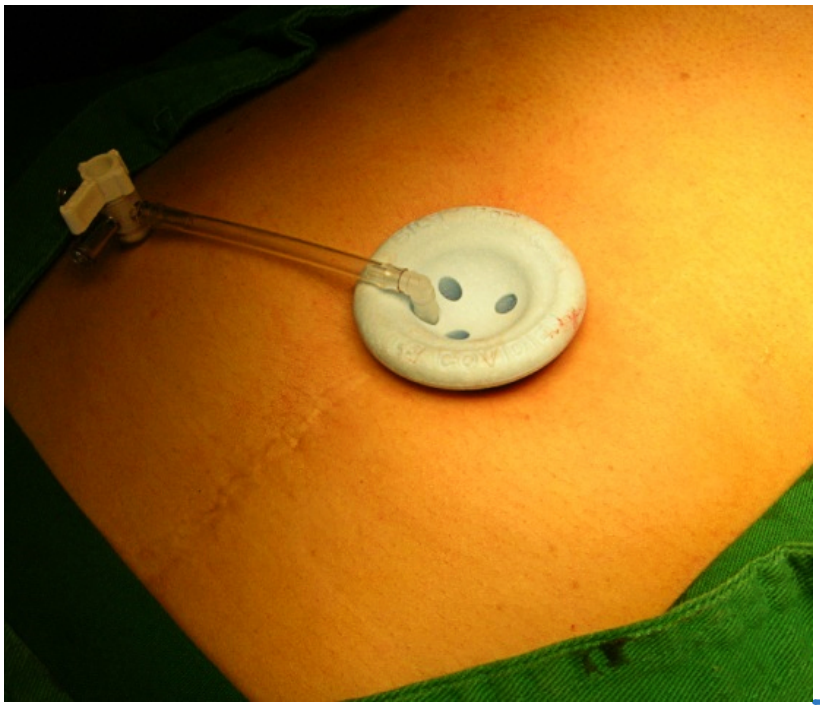

Fig 2. Single-port (SILS ® Covidien). (Reference photo of port)

CONCLUSIONS

Acute cholelithiasis and cholecystitis have a high incidence in the general population; in this case, there was also an anatomical disorder with genetic characteristics, which complicates the diagnosis and any potential surgical procedures. This results in a greater technical difficulty and the need for greater dexterity on the part of the surgeon due to the type of incision and complex surgical technique required.

In the case described in this article, the patient presented a satisfactory evolution and post-operative recovery. To this date, there are no published reports of a similar case in Colombia.

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THORACOSCOPIC MANAGEMENT OF AN ESOPHAGEAL LUNG REPORT OF A CASE

Esophageal lung is a rare bronchopulmonary foregut malformation in which the main stem bronchus arises from the esophagus. Since the description by Keely et al. in 1960, fewer than 25 cases have been reported. We present a case of a 4-month-old female, who was referred to our institution after 2 months of management for recurrent respiratory infections. Contrast studies were performed during the evaluation and a right bronchography was identified in the esophagogram. Bronchoscopy was performed confirming the atresic right bronchus. Complementary imaging and a cardiology evaluation confirmed the absence of major vascular anomalies—in particular, a pulmonary artery sling that has been described in relation to this entity. Due to the hypoplastic lung in the absence of major vascular anomalies, thoracoscopic pneumonectomy was deemed possible. The procedure was performed with four ports and 3 mm equipment was used. Special attention was paid to first identifying and dissecting the vascular structures, and then the arising esophageal bronchus was dissected. The hypoplastic lung was extracted through a small incision inferior to the axilla. We consider that because of the hypoplastic lung and vessels, the thoracoscopic approach is safe and feasible for the management of the esophageal lung and even for the esophageal bronchus in the absence of major vascular anomalies.

Keywords: *Esophageal lung, esophageal bronchus, thoracoscopic pneumonectomy.*

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INTRODUCTION

Esophageal lung is a rare bronchopulmonary foregut malformation in which the main stem bronchus arises from the esophagus (1). Symptoms of this entity are described as nonspecific, with chronic cough, recurrent pulmonary infections and respiratory distress as the usual presenting features (2). Usually, the treatment of choice has been the division of the communicating tissue and the resection of the anomalous lung tissue (1). We present a case of a 4-month-old female with a right esophageal lung managed with a right pneumonectomy using the thoracoscopic approach.

CASE REPORT

A 4-month-old female was referred to our institution after 2 months of management for recurrent respiratory infections. She was born at 34 weeks of gestation and weighed 1600 grams, requiring 17 days in the neonatal care unit for apneas and a hypoplastic right lung.

Several studies were performed during these 2 months. Chest X-rays showed right diffuse infiltrates and a markedly hyperinsufflated left lung (fig 1). Due to the persistent respiratory involvement, upper gastrointestinal series were made, in which a right bronchography was identified in the esophagogram (fig 2). An echocardiogram and CT scan with vascular reconstruction were also performed and no major vascular anomalies were identified; the pulmonary artery sling, an entity that has been specifically reported in association with the esophageal lung was ruled out in particular. A citomegalovirus infection was confirmed and treated.

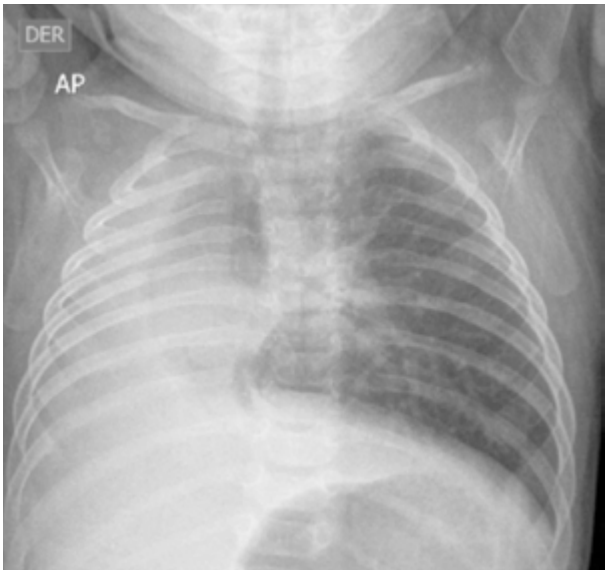

Fig 1. Chest X-ray. Right diffuse infiltrates and markedly hyper insufflated left lung.

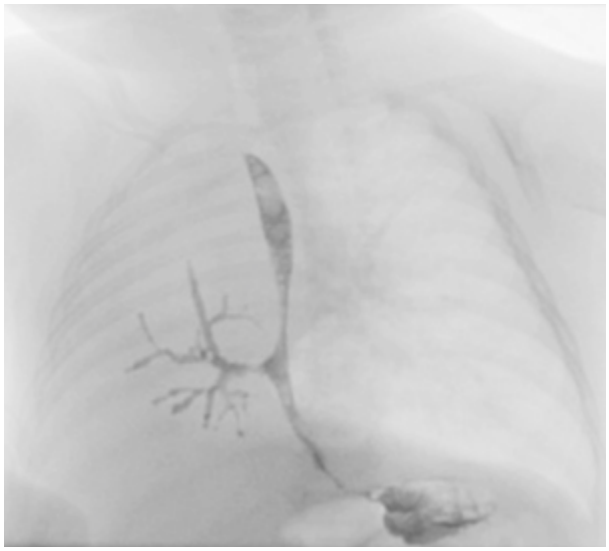

Fig 2. Esophagogram, with right bronchography.

Upon arrival to our institution, a multidisciplinary group evaluated the patient. She presented as nutritionally impaired with respiratory distress, requiring supplementary oxygen and nutritional support. A cardiology evaluation confirmed again the absence of major vascular anomalies with a hypoplastic right lung artery.

Due to the hypoplastic lung in the absence of major vascular anomalies, a thoracoscopic pneumonectomy was deemed possible and she was scheduled for surgery.

Immediately before surgery, a rigid bronchoscopy and esophagoscopy were performed. An atresic right bronchus was confir-

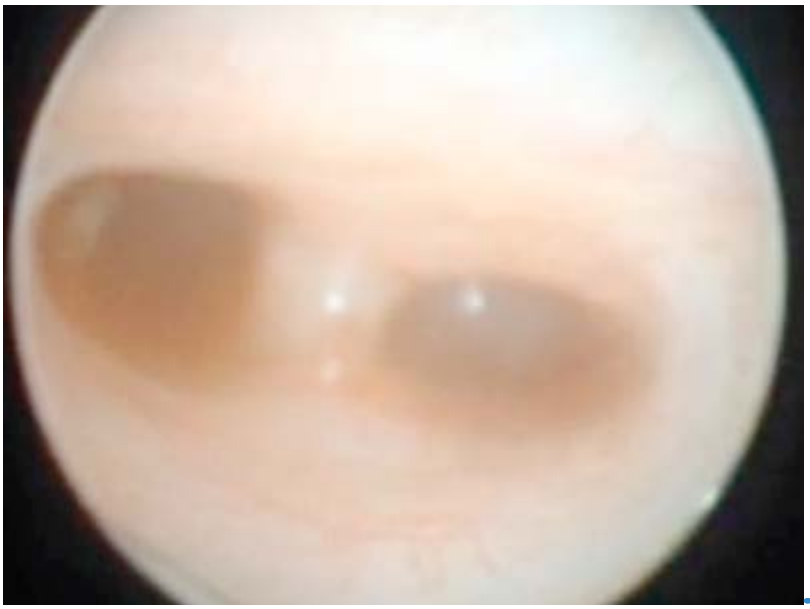

Fig 3. Bronchoscopy. Atresic right bronchus.

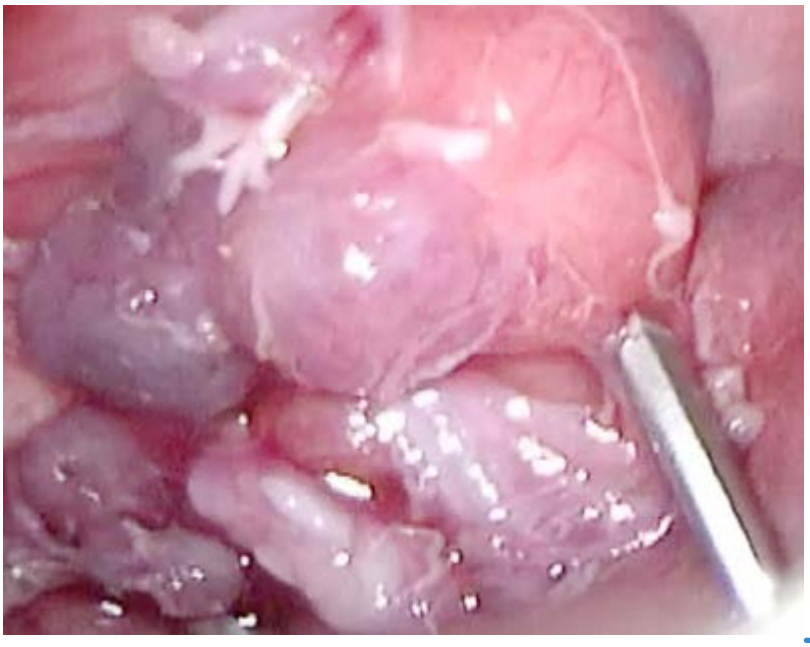

Fig 4. Enlarged lymph nodes covering the hilum

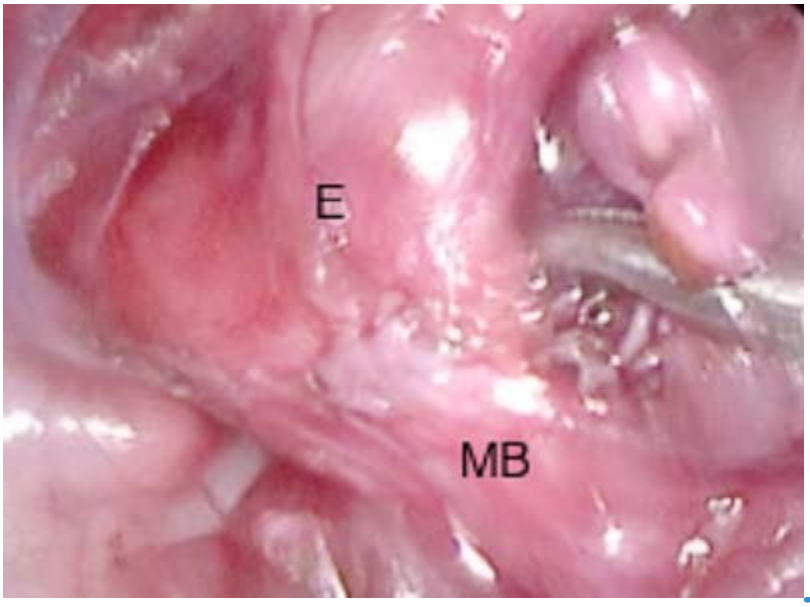

Fig 5. Main Bronchus (MB) arising from the esophagus (E).

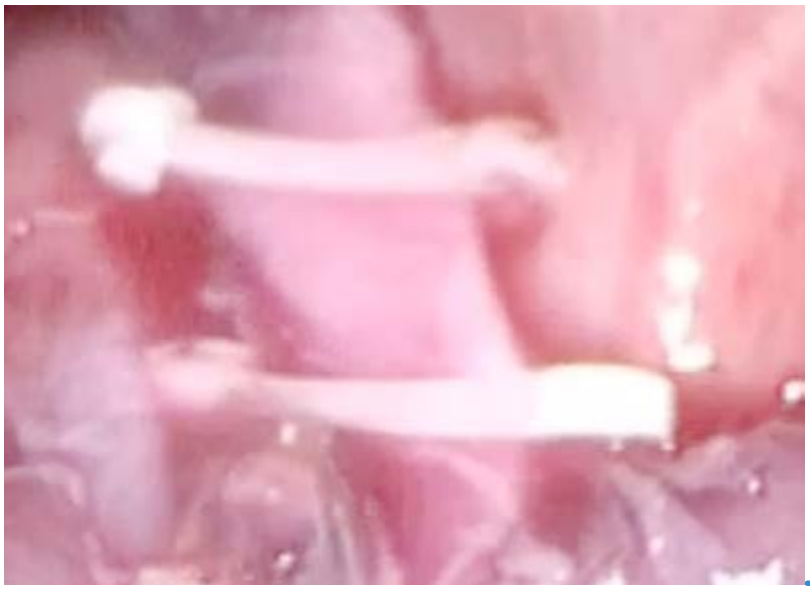

Fig 6. Hypoplastic right pulmonary artery.

med (fig 3) and the right bronchus arising from the esophagus was identified.

The patient was placed in the left lateral decubitus position, and four ports were placed with one port of 5 mm and three ports of 3 mm. 3 mm equipment was used. A hypoplastic right lung was confirmed and the absence of lobes was noted. Special attention was paid in the identification of the vascular structures at the pulmonary hilum that was covered by enlarged lymph nodes (fig 4). After these structures were identified, the esophagus was visualized and the right bronchus arising from the esophagus was dissected circumferentially (fig 5). 5 mm hem-o-loks were used to ligate proximally and distally the anomalous arising bronchus, which was subsequently transected with scissors. Then, the pulmonary artery (fig 6) was ligated proximally and distally with 5 mm hem-o-loks and transected with scissors. Finally, the pulmonary veins were ligated with 5 mm hem-o-loks proximally and transected. After the vascular structures and esophageal bronchus were divided, the hypoplastic lung was extracted through a small incision inferior to the axilla.

The patient was kept in hospital for one month after surgery due to a was initiated early in the postoperatory period through a nasogastric tube and oral intake was initiated after extubation was performed. The patient was discharged with low flow supplementary oxygen. In tests performed at the second, third, and fourth month, the patient was doing well with adequate scarring process but still with a low flow supplementary oxygen requirement. (fig 7). Nevertheless, due to a markedly hyperinsufflated left lung and right mediastinal shift (fig 8), the possibility of right pneumonectomy syndrome was taken into account. Unfortunately, the patient died after 7 months due to a viral community -acquired pneumonia.

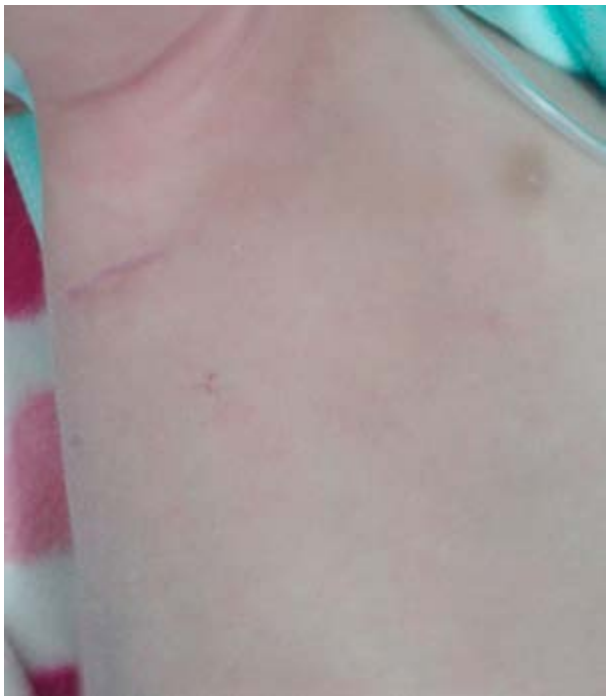

Fig 7. Outpatient control.

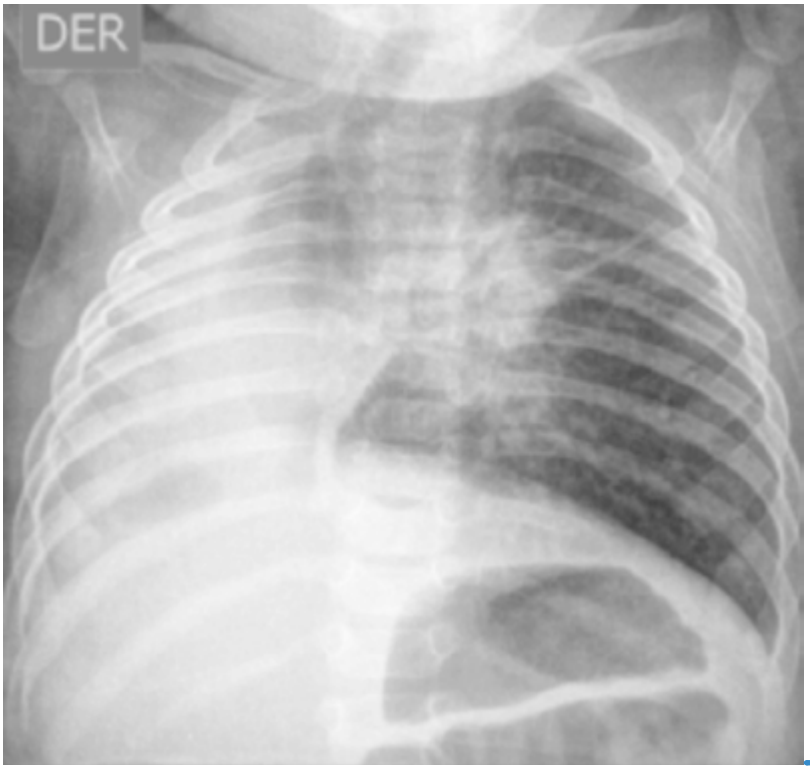

Fig 8. Control chest X-ray. Persisted hyperinsufflated left lung with right mediastinal shift.

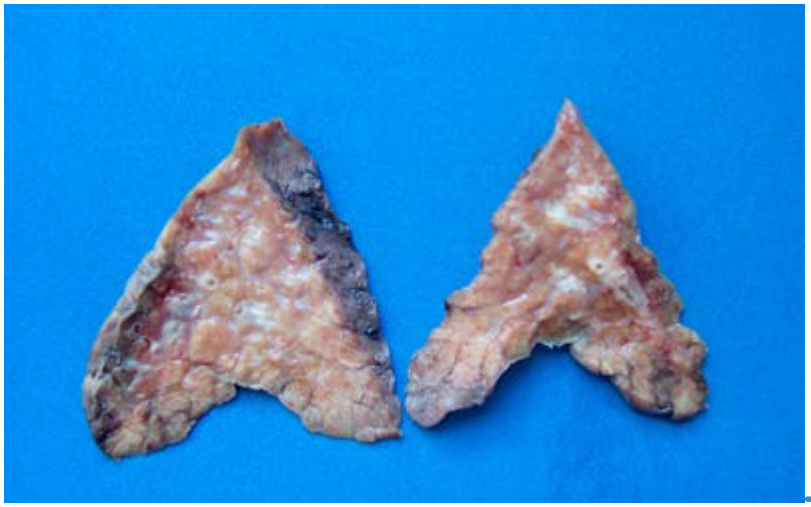

Fig 9. Right pneumonectomy: small, rubbery airless lung without lobulation.

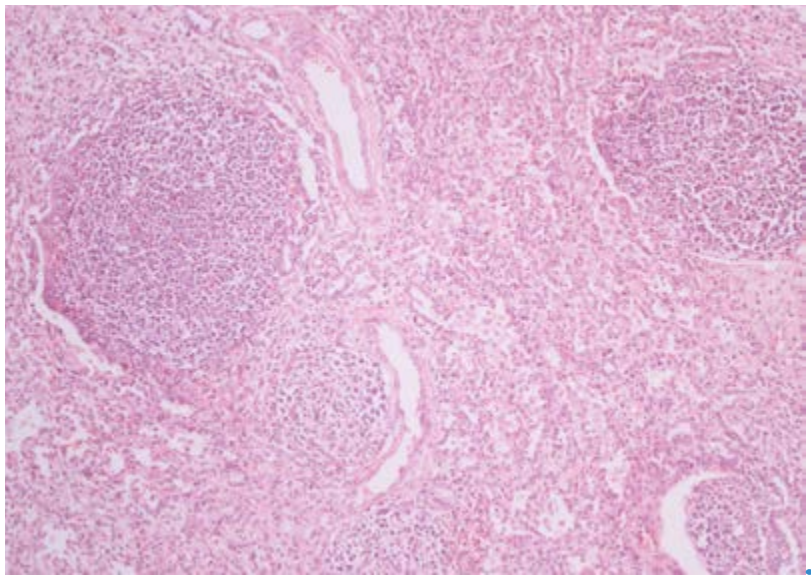

Fig 10. Lung parenchyma with marked lymphocytic infiltrate, presence of lymphoid follicles and well-formed germinal centers (H&E 100X)

Pathology evaluation of the surgical specimen showed a small, rubbery lung without lobulation, which weighed half of that expected for her age (15.5/33 g) (fig 9). The alveoli were small, delineated by cuboidal epithelium and there was marked lymphocytic infiltrate with presence of lymphoid follicles and well-formed germinal centers in bronchiolar walls as a sign of chronic infection (Fig 10).

DISCUSSION

The esophageal lung and esophageal bronchus are rare bronchopulmonary foregut malformations (1)(4). Since the description by Keely et al. in 1960, fewer than 25 cases of esophageal lung have been reported (1). The etiology is still unknown, but the evidence of squamous epithelium and respiratory epithelium in the communicating tissue between the lung and the esophagus supports the ventral foregut budding theory (3).

Most cases have been reported in the right side as in our case (1). Clinical presentation as described above is nonspecific, with chronic cough, respiratory distress and recurrent pulmonary infections (2).

This kind of pathology strongly supports the necessity of upper gastrointestinal tract images with contrast in patients with chronic respiratory symptoms, as it was used to diagnose this patient and repeated by many authors like Robin Cotton. Other malformations have been described in relation to this condition, such as esophageal atresia, duodenal atresia or stenosis, congenital heart disease and pulmonary artery sling (1)(4). These must be studied before surgery.

Traditionally, the treatment of choice has been the division of the communicating tissue and the resection of the anomalous lung tissue (1,6). Nevertheless, Lallemand et al., reported two successful reimplantations of esophageal bronchus to the trachea in the neonatal period (5,6), which may be only achievable early in life and has not been reported for complete lung communication. Even though reports of right postneumonectomy syndrome have not been found in the reviewed literature, this option should be considered. Due to the hyperinsufflated left lung and the absence of the right lung after surgery, there will be a continuous shifting of the mediastinum to the right, raising the possibility of developing this syndrome.

As in the opinion of the authors of this report, despite the unrelated death of the patient, we consider that due to the hypoplastic lung and vessels, the thoracoscopic approach is safe and feasible for the management of the esophageal lung and even for the esophageal bronchus in the absence of major vascular anomalies.

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TRAUMATIC ABDOMINAL WALL HERNIA FROM HANDLEBAR INJURY, LAPAROSCOPIC REPAIR REPORT OF TWO CASES

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ABSTRACT

Although rare, traumatic abdominal wall hernia associated with handlebar injury is a well-described entity in the pediatric population with about 40 cases and only one laparoscopic repair reported in children. We present two cases of male patients, 9 and 13 years old, who were assessed in our emergency room for blunt abdominal trauma associated with handlebar injury. The patients showed signs of handlebar trauma in the abdominal wall: one presented with a painful mass, and the other with intermittent pain in the area of trauma with no palpable mass. Neither of the patients were hemodynamically unstable or showed any peritoneal signs. Ultrasound and CT scans were performed in both patients to identify abdominal wall hernias containing bowel content in the absence of other injuries. Laparoscopic repair was performed uneventfully in both patients with interrupted non-absorbable multifilament suture with 2 and 3 ports respectively. Oral intake was initiated one day after surgery and both patients were discharged the following day. In the follow-up visit, the patients were asymptomatic and no signs of abdominal wall hernias were found.

Laparoscopic repair of blunt traumatic abdominal wall hernias is safe and technically possible in children and should be considered as the standard initial approach in the stable patient.

Keywords: *Handlebar injury, blunt abdominal wall hernia, laparoscopic repair of abdominal hernia.*

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INTRODUCTION

Although handlebar injuries are well known in children, abdominal wall hernias secondary to this mechanism are rare. There are few reported cases of this kind, with about only 40 reported cases identified in the reviewed literature (2). Traditionally, management has consisted in repairing the defect through an incision above the hernia or by laparotomy, though other authors propose a conservative approach in the asymptomatic patient (3,4), and there is only one report of laparoscopic repair (1). We present two cases of abdominal wall hernias repaired successfully by laparotomy.

CASE 1

A 13-year-old boy was referred to our institution three days after suffering a handlebar injury in the right iliac fossa. He presented with history of abdominal pain, nausea and vomiting. On examination he was hemodynamically stable, though a handlebar sign was present in the right iliac fossa (fig 1) with tenderness. No palpable mass was found.

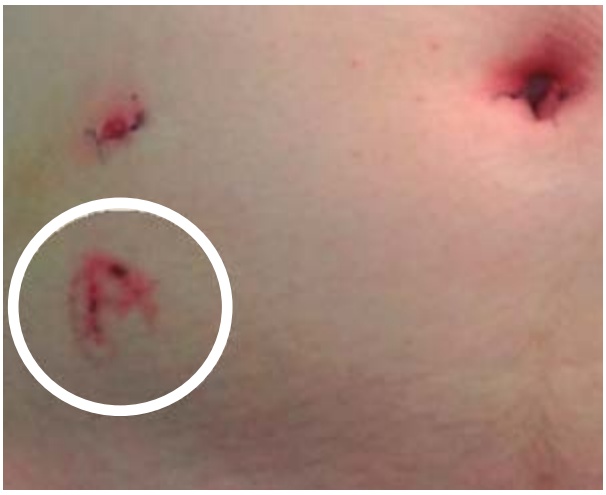

Fig 1. Handlebar sign.

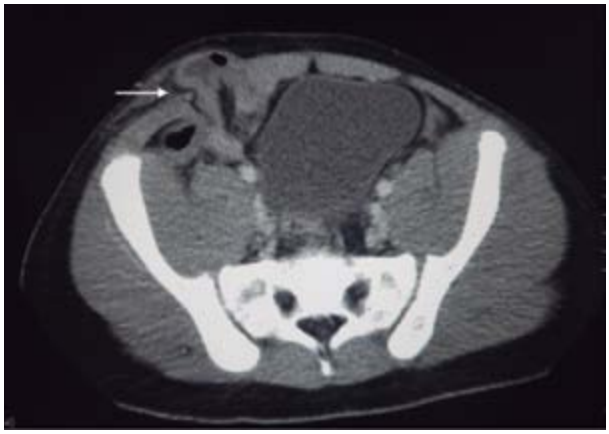

Fig 2. CT scan, with an abdominal wall defect and small bowel trhough it.

An abdominal ultrasound was performed in the referring institution with an image that suggested a traumatic abdominal wall hernia. A CT scan was performed, without evidence of lesion in the solid viscera and the traumatic hernia was confirmed with bowel protruding trough the defect (fig 2).

The patient was taken to the operating room. The laparoscopic approach was performed with one 10 mm port at the umbilicus and two 5 mm ports in the right flank and left iliac fossa, respectively. A 2 cm defect above the internal right inguinal ring was found (fig 3), compromising the peritoneum and transverse and oblique muscles.

The defect contained a segment of ileum, which was reduced, and no signs of ischemia or lesion in its mesentery were found. The defect was closed (fig 4) with interrupted non-absorbable multifilament sutures using the extracorporeal knot tying technique. Oral intake was initiated the next day, and the patient was discharged the following day. In the first follow-up visit, the patient was doing well, without palpable masses or abdominal pain. After two years, the patient remains asymptomatic regarding the abdominal wall hernia with no recurrence.

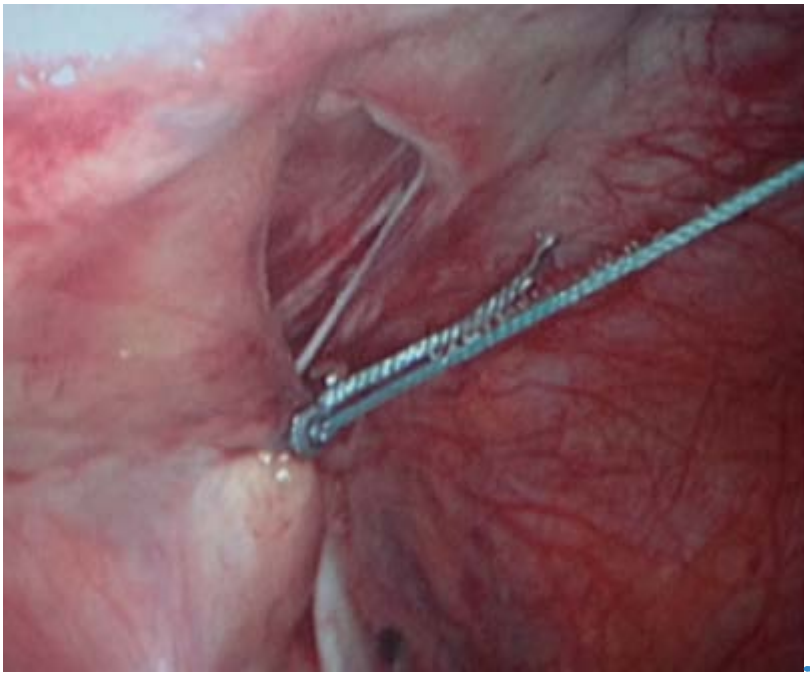

Fig 3. Traumatic abdominal wall defect above the internal inguinal ring.

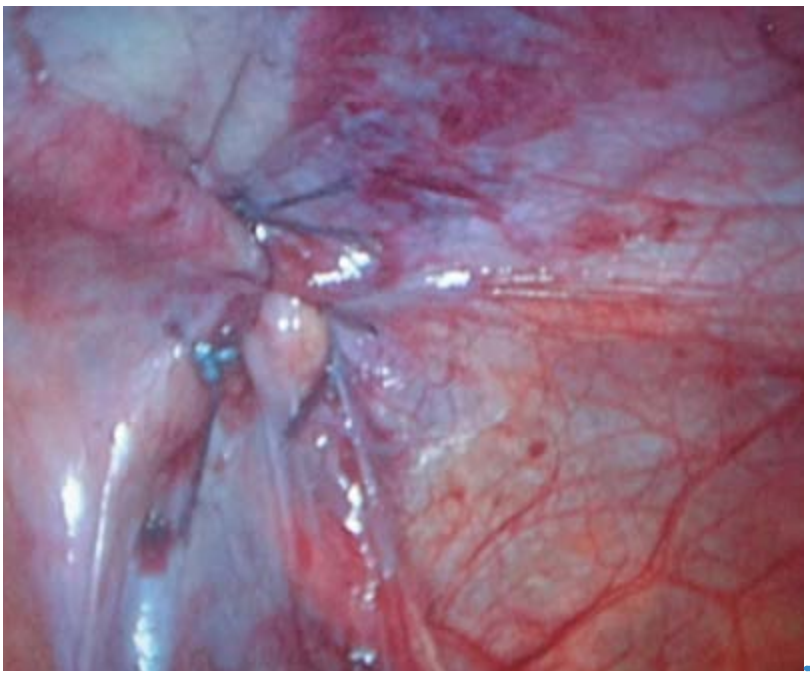

Fig 4. Closed defect.

CASE 2

A 9-year-old boy presented to our institution 12 hours after suffering a handlebar injury in the upper-left abdomen. After the injury, he noticed a painful abdominal mass at this level, and subsequently, vomiting. On examination, the patient was hemodynamically stable, with the handlebar sign (Fig 5) at the level of the upper-left abdomen, with a tender palpable mass.

An ultrasound was performed, and an abdominal wall hernia was identified and confirmed by a CT scan evaluation (Fig 6) in which no other lesions were found. The abdominal wall hernia was filled by a small bowel segment.

Based on these findings, the patient was taken to the operating room for laparoscopic repair. Two 5 mm ports were used, one through the umbilicus and one in the right iliac fos-

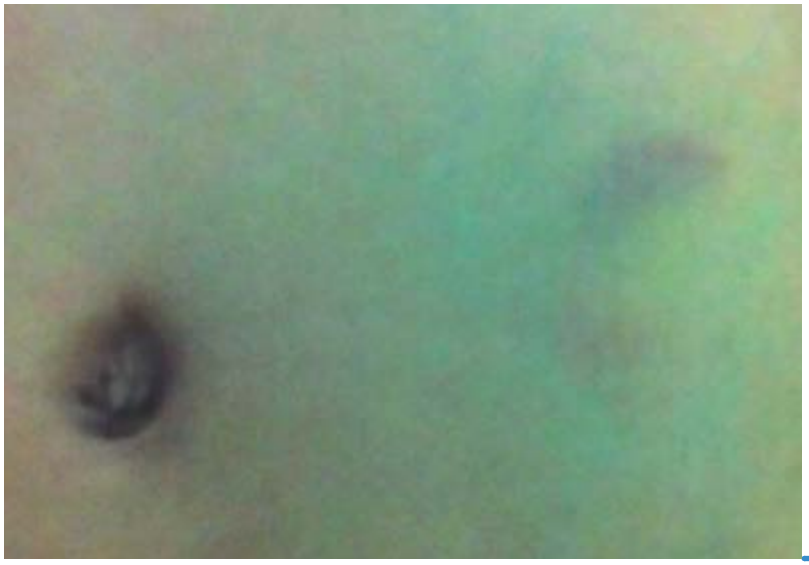

Fig 5. Handlebar sign.

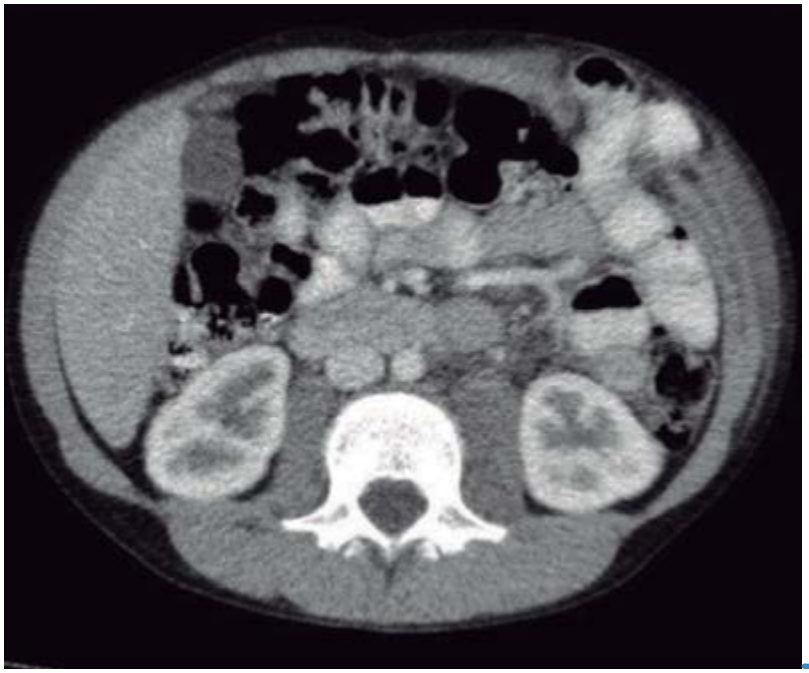

Fig 6. CT scan demostrates the presence of an abdominal wall hernia with bowel content.

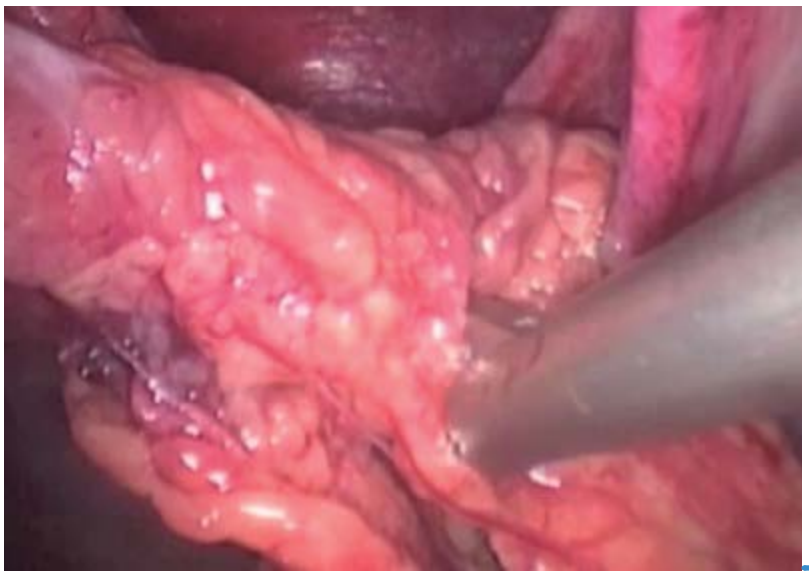

Fig 7. Abdominal wall hernia with omentum.

sa, after the pneumoperitoneum was insufflated. Only the omentum (Fig 7) was identified inside the hernia, and was reduced.

The defect was about 4 to 5 cm long compromising the peritoneum and transverse and oblique muscles. It was corrected with interrupted non-absorbable sutures. The needle was passed through 2 mm skin incisions above the hernia and recovered with an endoscopic needle holder. Then, the edges of the hernia were taken and the needle was recovered again trough the skin incision. The suture was tied until the defect was corrected completely. Oral intake was initiated and tolerated the next day and the patient was discharged the following day. In the follow-up visit, the patient was doing well, without signs of hernia recurrence.

DISCUSSION

Abdominal wall hernias represent about 1% of lesions related to blunt trauma (1). Although rare, traumatic abdominal wall hernia associated with handlebar injury is a well-described entity (1) in the pediatric population with about 40 cases reported in the reviewed literature (2). Most cases of pediatric handlebar injuries occur between the ages of 9 and 14 (8). Abdominal wall hernias can be detected soon after handlebar injury or a few days after as it is not always evident in the physical examination, such as it was in our first case and other reported cases (5). Additionally, a lack of knowledge of this pathology might result in missed and underreported cases in the asymptomatic patient (4).

As described above, a high index of suspicion is needed to perform the diagnosis (7), and complementary imaging using abdominal ultrasound and CT scan might also be necessary. In one of our children, clinical findings were insufficient to perform the diagnosis and complementary images were needed. CT scans will also help in the evaluation of additional injuries (5).

Usually in children, these kinds of hernias are secondary to a low energy trauma, as in bicycle handlebar injuries (5), and it is secondary to a sudden application of blunt force that is insufficient to penetrate the skin but strong enough to disrupt the muscle and fascia (6). In the reviewed literature no major related injuries were reported and were correlated with the findings in our cases (6).

Open layer repair has been used traditionally either through an incision above the hernia or by laparotomy (2,5). Laparoscopy has been used for diagnostic purposes (2,6) and there is one report of laparoscopic repair using an Endo Close TM system (Covidien, Mansfield, MA) (1). We present two different techniques that can be used in the management of this disease that are reproducible with adequate results. Other authors have postulated the possibility of a conservative approach for the asymptomatic patients, as they identify spontaneous resolution with time (3,4). Nevertheless, we don't have enough experience with this approach to be able to recommend it. Early primary repair is also reported to limit ongoing morbidity related to pain, limitation of function, and hospital stay (7).

Laparoscopic repair of blunt traumatic abdominal wall hernias is safe and technically possible in children, and should be considered as the standard initial approach in the stable patient.

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HIV DRUG RESISTANCE IN A 13 YEAR OLD BOY, A MULTICASUAL PROBLEM

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ABSTRACT

Background: In 2011, at least 34 million people live with HIV. HIV vertical infected children require close follow-up from all the way through diagnosis to treatment and management of complications.

Case summary: This is the case of a 13-year-old male patient with HIV (vertical transmission) diagnosed at 4 months old. His HIV diagnosis was made in the context of opportunistic manifestations of immunodeficiency because his mother did not access prenatal check-ups. He developed AIDS sequelae such as spastic paraparesia derived from HIV myelopathy and CMV retinitis due to immunodeficiency; these diseases presented in first two years of life. After three years from HAART initiation, the patient was exposed to inadequate HAART (ritonavir without another protease inhibitor), and experienced a first change of therapy due to virological failure. Subsequent treatment regimens —a sum of 7— presented failures in their formulation and

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this, along with delays due to administrative issues, led to the patient developing multiresistance to most of antiretrovirals given. The patient died mainly from multiorganic failure due to HIV and wasting syndrome.

Conclusion: Congenital HIV is a fundamental issue in public health. It is a preventable disease, and perinatal management, including diagnosis and treatment, is a must. Treatment has demonstrated effectiveness when it is given with proper schemes and adherence. Administrative barriers led to failures in treatment and this affects the prognosis of any patient with HIV. This case is an example that highlights the relationship between virological and clinical failures with health system barriers, in HIV infected children. Managing gaps in diagnosis, antiretroviral administration, and follow up of HIV infected children translates into the prognosis of future adolescents and adults.

Keywords: *HIV, children, HIV drug resistance.*

CLINICAL CASE: DESCRIPTION

The following data was extracted from the complete patient's clinical history, interviews with relatives and the Foundation coordinator.

The patient was the product of third pregnancy of a 27-year-old mother. The patient's mother did not access HIV testing during pregnancy or antenatal care, and we have no information about why she did so. The patient was born at term in a public hospital by vaginal birth. An HIV test was not performed at birth. The patient's parents worked as street vendors and were diagnosed with HIV infection at same time as their son. There is no information about lactation. Mother died by the time the patient was 1 year old due to AIDS related complications (tuberculosis and wasting syndrome). The father continued to live but had sequelae of neurotoxoplasmosis. The patient's two siblings are not infected. By age of 2, the patient entered a foundation for HIV infected children in Bogotá, where they assumed responsibility for his comprehensive care until death. The patient was affiliated to the contributive health regime, his health care was provided in multiple institutions, but mainly in a third level hospital where he was attended by a pediatric infectologist.

The patient was diagnosed with HIV infection stage C3 by the age of 1 after multiple hospitalizations. His exams showed severe immunodeficiency and multiple opportunistic diseases (pulmonary tuberculosis, myelopathy from HIV with spastic paraparesia and CMV retinitis, malnutrition, Pneumocystis jirovecii pneumonia). All this clinical information was validated by specialists through the clinical history. The confirmation test for HIV (western blot) authorization was made with six months of delay. Table 2 shows viral loads and CD4 cell counts over the span of his life as related to ARV regimens.

The first ARV regimen lacked potency (Zidovudine, Didanosine and ritonavir without another protease inhibitor because of a lack of oral solution presentations), leading to incomplete suppression of viral replication, which was interpreted as virological failure. Multiple empirical ARV regimens were given until a necessary viral genotypification was ordered at the age of seven —with a two years delay between when the exam was ordered and when it was actually carried out—. Genotypification showed resistance to all known NRTI's and to PI. Further genotypifications showed new mutations and polymorphisms that confer resistance to almost all ARVs. Table 1 shows the history of antiretroviral resistance mutations and polymorphisms.

Date	Age	Viral Load	Cd4	Art
01/04/2001	17 months	>500.000 copies/mL (Log > 5.70)	185	Zidovudine + didanosine + ritonavir
22/02/2002	2 years, 4 months	800.000 copies/mL	219	Lopinavir/ritonavir + nevirapine + zidovudine
16/06/2004	4 years, 7 months	No data	272	
11/08/2004	4 years, 9 months	210785 copies/mL (Log 5.32)	444	Stavudine + Lopinavir/ritonavir + lamivudine
30/03/2005	5 years, 5 months	>500.000 copies/mL (Log > 5.70)	408	
02/09/2005	5 years, 10 months	423 copies/mL (Log 2.62)	320	
01/02/2006	6 years, 3 months	28934 copies/mL (Log 4.46)	261	
30/11/2006	7 years, 1 month	358.215 copies/mL (Log 5.55)	338	
09/10/2007	7 years, 11 months	>500.000 copies/mL (Log > 5.8)	108	Lopinavir/ritonavir + lamivudine + efavirenz
03/08/2009	9 years, 9 months	405.169 copies/mL (Log 5.61)	15	
02/09/2010	10 years, 10 months	201.177 copies/mL	313	
08/11/2010	11 years	864.917 copies/mL	113	
06/01/2011	11 years, 2 months	646.186 copies/mL (Log 5.81)	165	Raltegravir + Etravirine+Enfuvirtide + Maraviroc
29/06/2011	11 years, 8 months	110.864 copies/mL (Log 5.04)	7	
01/07/2012	11 years, 9 months	185.006 copies/mL (Log 5.27)	4	Raltegravir + Tenofovir/emtricitabine + darunavir
10/01/2013		44.002 copies/mL (Log 4.64)	14	

Table 1. Immunovirologic tests and ARV.

Antiretroviral Group	Mutations And Polymorphisms
Reverse transcriptase inhibitors	M41L, D67N, K70R, Y181C, T215Y, K219E, M184V, T69E, M184, G190A, Y188L, T215F/V
Protease inhibitors	A71V, I54L, I62V, I84V, L10F, L33F, L63P, L90M, M36I, M46L, V82A, K20R, F53L/F, Q58E, K43T, I13V, L89V, V11I, E35D, H69R, G73S

Table 2. Antiretroviral resistance mutations and polymorphisms.

6 months after the genotype results, CD4 had decreased to 66 cell/uL with consequent clinical deterioration that required further hospitalizations. Rescue therapy was initiated, but viral load (VL) continued to rise. A tropism test for the CCR5 co-receptor recommended the use of Maraviroc, a novel therapy that represented an option; however there was a 4 month suspension of ARV because of delay in supply.

The patient received a 5-month treatment with Maraviroc and Enfuvirtide. The VL decreased to 1/8 of the prior count. Nevertheless, the CD4 count continued to decrease from 165 to 7 cells. A new genotypification showed prior resistance plus probable or emergent resistance to NRTI and Non-nucleoside retrotranscriptase inhibitors (NNRTIs). New VL and CD4 cell counts confirmed persistence of virological failure. After one last AR change, the case was presented at a bioethical committee of the last institution where he was attended. They recommended suspension of all ARV drugs because of its lack of effectiveness and administration of analgesia for pain relief in the context of palliative care. The patient died after a complicated respiratory infection 4 months after ARV discontinuation.

DISCUSSION

This case represents the consequences of the mismanagement of an HIV infected patient at several levels. The first one was the missed opportunity for HIV prenatal diagnosis in his mother while she was pregnant. Poverty due to informal employment affects the access to HIV tests because the economic resources are limited and often they are invested in food and living instead. It is necessary to ensure that all pregnant women access HIV and syphilis tests during antenatal controls, hopefully in first trimester, one way to do this is by rapid point of care tests that do not require administrative authorization or other administrative barriers. Also, HIV tests must be administered during the third trimester and labor.

The second level of failure was directly related to the empiric administration of multiple ARTs to this patient before genotypic evaluation for HIV ART resistance. The elevated viral load and the positive selection induced by multiple empiric antiretrovirals resulted in multidrug ART resistance, as has been demonstrated in several studies (9 - 11). As in adults, the genotype is an accurate measure for determining the best ART for each patient when virological failure is detected. As such, it must be made as soon as the failure is diagnosed (30).

Drug resistance cases in children may become a public health problem since they behave as regular adolescents and adults, with a high risk of spreading the infection, leading to the consequences that that implies (31, 32).

The delay in health services and the excess of administrative procedures that are present in Colombia's health system affects the clinical course of pathologies such as HIV and AIDS related complications. This leads to cases undertreated sequelae and the loss of life expectancy, especially in children.

Due to the great capacity of the HIV virus to acquire resistance to drugs, it is necessary to provide the best of care to HIV patients, and especially prenatally infected pediatric patients, in terms of early diagnosis, HIV resistance evaluations, and ART treatment (33). This includes the availability of resistance testing to select the best ARV combination for each patient and a multidisciplinary approach to determine the best course of management in cases of reactivation and re-emergence of latent virus when the conditions are favorable.

CONCLUSION

This case highlights the challenge of middle and low income countries in the diagnosis, evaluation, and treatment of HIV infected children. There is a peremptory need to continue efforts to guarantee antenatal controls and HIV tests for all pregnant women. Women in poverty must be a priority for these programs. Physicians that treat HIV positive children must be trained in this specific issue, Clinical Practice Guidelines must be taken account, especially now that guidelines were published in 2015 with evidence-based recommendations. The adherence to guidelines will lead to fewer undertreated children and a better quality of life for this population.

The access to health system services must be assured in order to apply the guideline's recommendations, especially when diagnostic procedures and antiretrovirals are not included in the health plan. There must be an adequate approach to management including not only the treatment of sequelae and the complications of pathologies but also prevention.

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