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Endocrinology and Infertility. Baltimore, USA: Willliams & Wilkins, 1994:1-967.

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INDEX

SPECIAL BRAZILIAN CONGRESS OF ULTRASOUND 30 YEARS

From October 18th to 21st, at the Frei Caneca Convention Center, in São Paulo, we have an appointment with scientific knowledge, professional appreciation and friendship at the 27th Brazilian Congress of Ultrasonography and 19th International Congress of Ultrasonography FISUSAL, edition special celebration of SBUS's 30th anniversary. With more than 190 national and international speakers, book launches and several other attractions, this will certainly be a unique and remarkable scientific event, commensurate with this important date for Brazilian ultrasound.

For this edition of the largest and most traditional specialty event in Brazil to be a great success, we hope to have your presence. We met in October, at the Frei Caneca Convention Center, in São Paulo.

ANTONIO GADELHA DA COSTA
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ORIGINAL ARTICLE

"CURRENT TRENDS AND CHALLENGES IN THE TRAINING OF ULTRASONOGRAPHERS IN BRAZIL: A COMPREHENSIVE ANALYSIS"

FERNANDO MAUAD [1], AUGUSTO BENEDETI [1], YUJI MATSUI [1], RUI FERREIRA [2], HEVERTON PETTERSEN[2], FRANCISCO MAUAD FILHO [1]

ABSTRACT

INTRODUCTION: The training of ultrasound physicians is a specialized field of medicine that involves the use of ultrasound for diagnosing and monitoring diseases in various parts of the body. Preparation in this area requires specialized training after graduation from medical school, typically in fields such as radiology, gynecology, obstetrics, or related areas. This allows physicians to acquire the necessary knowledge to perform and interpret ultrasound examinations in their specific fields. Ultrasonography in Brazil began in the 1970s and has evolved significantly, extending beyond anatomical identification to include the study of cardiovascular physiology through Doppler velocimetry.

OBJECTIVE: This study aimed to understand the role of ultrasound physicians in Brazil and propose a training model for general and specialized ultrasound physicians in medical fields.

METHODS: In 2017, EURP/FATESA conducted a survey to estimate the number of ultrasound physicians in the Brazilian market, which was approximately 79,000 at the time. Responses from 864 ultrasound physicians were validated through a questionnaire covering topics such as the work region, reasons for choosing ultrasound, years of experience, possession of specialization titles, and perceptions regarding the need for additional ultrasound training.

RESULTS: The survey results showed that the Southeast region had the majority of ultrasound physicians, followed by the South, Northeast, Midwest, and North regions. The choice of the profession was related to both aptitude and preference, as well as job opportunities and lifestyle. However, most physicians did not possess specialization titles in ultrasound. The majority of respondents had less than six years of experience in ultrasound, highlighting the relatively new nature of this field in medicine. Moreover, more than half of the physicians believed that additional specialization was necessary to work in ultrasound subareas, with most considering that training should take at least two years.

DISCUSSION: The results revealed that regions with higher medical density and economic and technological resources had more ultrasound physicians. The lack of specialization titles and proper training was a concerning finding, given the importance of ultrasound in medical practice. The survey emphasized the need for standardization in the training of physicians in ultrasound.

CONCLUSION: The authors propose that the training of a general ultrasound physician should involve two years of training with an established program, allowing the physician to work in outpatient and hospital settings and take an evaluation exam. Additionally, ultrasound physicians with more than four years of experience could be allowed to take the general ultrasound certification exam if they were recommended by qualified members. For specialization in specific areas, the professional should obtain a recognized title in the specialty, pursue postgraduate studies in the field, and be recommended by a qualified ultrasound physician. This study highlights the importance of proper training for ultrasound physicians to ensure the quality of healthcare.

KEYWORDS: ULTRASOUND, CERTIFICATION, TRAINING, SPECIALIZATION, EXTENSION, PRACTICE, PERFOMANCE

INTRODUCTION

The training of medical ultrasonographers is a specialized area of medicine that involves the use of ultrasound for diagnosis and monitoring of related diseases in all parts of the body.

Training in this field requires a specialized training program after completing medical school. Typically, doctors interested in becoming ultrasonographers pursue specialization in radiology, gynecology and obstetrics, or related fields such as fetal medicine, internal medicine, or cardiology, or they seek out specialized courses. This additional training enables them to acquire the necessary knowledge to perform and interpret ultrasound examinations in their respective areas of practice [1,2].

The practice of ultrasonography in Brazil began in the 1970s, and over this period, we have witnessed significant technological advancements. These advancements have allowed ultrasonography to go beyond simple identification of

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organ anatomy. With the incorporation of Doppler velocimetry, it has become possible to study cardiovascular physiology and understand its pathological mechanisms.

During the ultrasound examination, the patient is present, and it is crucial for the ultrasonographer to ask relevant clinical questions to understand the purpose of the exam. This clearly demonstrates that ultrasonography is based on three essential pillars: anatomy, physiology, and clinical aspects. These elements play a fundamental role in the practice of medicine in Public Health.

After the medical history-taking, the physician performs a physical examination of the patient, using the images generated by ultrasound of the relevant structures. This process starts with the patient's chief complaint to understand the current illness and reconstruct their clinical history. The result is a comprehensive ultrasonographic report.

The training of medical ultrasonographers is ongoing and involves the continuous updating of knowledge and skills due to technological advancements and the evolution of medical practices. Participation in conferences, workshops, and continuing medical education courses is crucial for ultrasonographers to stay current and enhance their skills [3-7].

Medical ultrasonographers play an important role in modern medicine because ultrasonography is a valuable tool for early disease diagnosis, guiding invasive procedures, monitoring pregnancies, and tracking various medical conditions. Their specialized training and technical skills enable them to provide quality patient care, contributing to the improvement of healthcare [4-10].

METHODS

A research study conducted by EURP/FATESA in 2017 estimated that there were approximately 79,000 ultrasonographers in the Brazilian market at that time.

In light of this data, a questionnaire was developed and validated by 864 medical ultrasonographers. The questionnaire consisted of the following questions:

Work region? Why did you choose to be an ultrasonographer? How long have you been performing ultrasounds? Do you have specialist titles? What is your training in ultrasound? Do you believe that to work in a sub-area of ultrasound, additional specialization is necessary? And how long do you believe it takes to train a medical professional in ultrasound?

The objective of collecting responses to this questionnaire was to gain an understanding of how medical professionals are practicing in the field of ultrasonography and to facilitate the development of a proposal for training general ultrasonographers in specialized medical areas.

RESULTS

The results of the research conducted by EURP on the profession of ultrasonographers reveal valuable information about their work regions, reasons for choosing this career, possession of specialist titles, experience in the field, and training in ultrasonography.

Table 1 shows the distribution of the working regions of the interviewed ultrasonographers.

Brazilian region	%
South	45%
Southeast	20%
North East	16%
Midwest	10%
North	9%

Table 1: Work Regions of Ultrasonographers.

Table 2 presents the reasons why the respondents chose the profession of ultrasonographer.

Reasons	%
Aptitude/preference	45%
Good job opportunities	21%
Lifestyle	16%
Others (professional enhancement)	15%
No response	3%

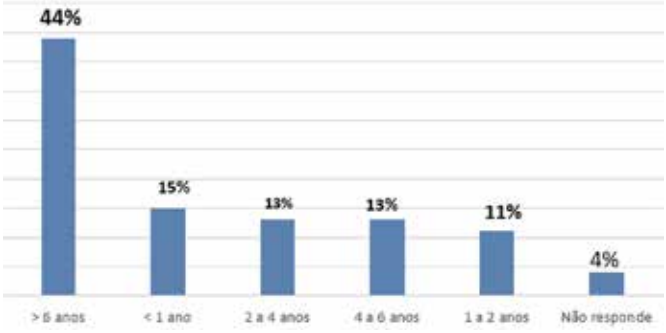
Table 2: Reasons for Choosing the profession of ultrasonographer

Table 3 illustrates an analysis of the possession of specialist titles in ultrasonography.

Specialist title	%
Does not have	69%
FEBRASGO	14%
CBR	12%
No response	5%

Table 3: Do you have any specialist titles in your field of practice?

Graph 1 shows how long the ultrasonographer has been performing ultrasound examinations.



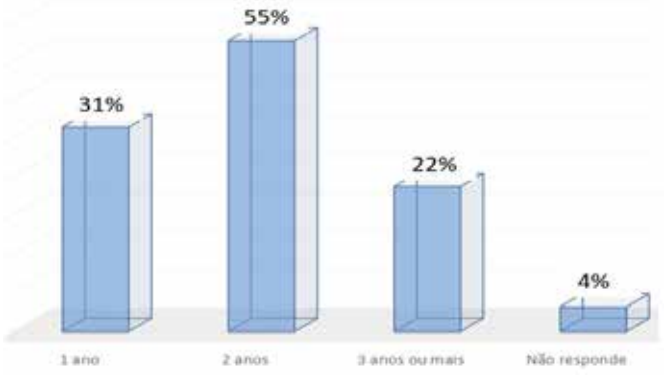
Graph 1: How long have you been performing ultrasounds?

Table 4 displays the responses regarding the need for additional specializations to work in subareas of ultrasonography.

Need for specialization	%
Yes	51%
No	45%
Did not answer	4%

Table 4: Does Working in a Subarea of Ultrasonography Require Additional Specialization?

Graph 2 illustrates the time considered necessary for the training of a professional in ultrasonography.



Graph 2: Time Believed to Be Necessary for the Training of an Ultrasonographer.

Table 5 provides information about the training of professionals in ultrasonography.

Professional Training	%
Improvement Course (180 to 360 hours)	39%
Extension Course (<180 hours)	21%
Training Course (Medical Residency, 2800 hours/year for two years)	16%
LATU SENSU Graduate Course (>360 hours)	15%
No Response	9%

Table 5: What is your background in US?

DISCUSSION

The distribution of medical ultrasonographers in Brazil.

When evaluating the distribution of medical ultrasonographers by region in Brazil, it can be observed that 45% of them are located in the Southeast region, which is home to the largest portion of the Brazilian population, estimated to be around 52-53% currently. Following that, we have the South region, with 17% of the doctors, equivalent to its population proportion. The Northeast region, representing 18% of the Brazilian population, comes in third place. Finally, the Central-West and North regions, which account for approximately 10% of the population, have a lower presence of medical ultrasonographers.

It is evident that regions with higher medical density, economic power, and technological advancements have a significantly larger number of ultrasonographers. Additionally, the Southeast region, which concentrates the largest centers for education, research, job markets, and technological investments, has the highest proportion of specialist physicians in ultrasonography per 100,000 inhabitants. States such as São Paulo, Rio de Janeiro, Rio Grande do Sul, and Santa Catarina exhibit a ratio between 6.6 and 13.9 doctors per 100,000 inhabitants, while the North and Northeast regions have a ratio between 1.3 and 3.7. This reinforces the correlation between the concentration of ultrasonographers and medical density, highlighting that areas with more doctors per capita also have a higher number of specialists in ultrasonography (Table 1).

Motivation for choosing the profession of ultrasonographer.

When asking the respondents why they chose ultrasonography as their profession, 45% of them indicated it was due to their aptitude and personal preference, while 37% mentioned being attracted to the job opportunities and lifestyle associated with this career. This preference for ultrasonography based on aptitude can be attributed to the increasing demand for rapid diagnostic exams, which significantly contribute to improving patient care and monitoring.

On the other hand, the emphasis on good job opportunities underscores the importance of promoting the training of more ultrasonographers, recognizing the growing demand for these professionals in the healthcare field (Table 2).

Qualification in the area of ultrasonography.

An analysis of the possession of specialist titles in ultrasonography revealed a significant concern, as approximate-

ly 70% of professionals do not possess any title certifying them as medical ultrasonographers. This is relevant because doctors can work in various areas of medicine, emphasizing the importance of enhancing the qualification of these professionals. Improving qualification is essential for obtaining more accurate results in exams, which, in turn, directly impacts the quality of medical care. This situation highlights the need to establish guidelines and standards for the training of doctors in ultrasonography (Table 3).

Experience in the area of ultrasonography.

Analyzing Graph 1, we can observe that the significant majority of the physicians who participated in the survey, equivalent to 66%, have less than six years of experience in the field of ultrasonography. This data reflects the relatively recent incorporation of ultrasonography into the landscape of medical semiology.

The increasing participation of professionals in ultrasonography in recent years suggests a growing interest in this imaging diagnostic technique. This can be attributed to its essential role in contemporary medicine, providing rapid and non-invasive diagnoses, as well as detailed patient monitoring.

This trend also underscores the importance of continuous improvement and education for physicians in the field of ultrasonography, ensuring the provision of quality services and contributing to the advancement of diagnostic medicine (Graph 1).

Need for specialization.

Table 4 presents revealing data regarding the need for additional specializations to work in subareas of ultrasonography. According to the responses of the research participants, 51% stated that yes, it is necessary to pursue additional specialization, while 45% claimed it is not necessary. A small group, corresponding to 4%, did not provide a response.

These results indicate that half of the interviewed professionals are involved in specific areas of practice in addition to ultrasonography. This underscores the relevance of ultrasonography in clinical and diagnostic practice across various medical specialties. As ultrasonography technology advances, understanding specializations within specific areas of medicine becomes essential.

The role of ultrasonography in contemporary medicine is undeniably essential, enabling precise and non-invasive diagnoses in various specialties. Therefore, the pursuit of additional specializations in this field demonstrates the commitment of physicians to provide quality care and stay abreast of technological advancements to better serve patients (Table 4).

Time required for professional training in ultrasonography.

The analysis of the research reveals valuable information about the time considered necessary for the training of a professional in ultrasonography. According to the responses of the participants, 31% believe that one year of training is sufficient, while the significant majority, representing 55%, considers that at least two years of training are necessary. Additionally, 22% of the respondents mentioned that ideal training requires three or more years. A smaller group, equivalent to 4%, chose not to answer this question.

These data highlight the complexity and scope of the field of ultrasonography, as well as the importance of the time dedicated to the training of professionals. The majority agrees that a period of at least two years is essential to acquire the knowledge and skills necessary to competently work in this field.

This perception reinforces the idea that ultrasonography is a medical discipline that requires a broad range of knowledge and specific practices. The pursuit of continuous improvement and a more robust training in this field contributes to ensuring the quality of services provided and the advancement of diagnostic medicine (Graph 2).

Table 5 presents crucial information about the training of professionals in ultrasonography. The responses of the participants reveal that:

• 39% completed advanced training courses, lasting from 180 to 360 hours.

• 21% took extension courses, lasting less than 180 hours.

16% underwent training courses, which include medical residency with a workload of 2,800 hours per year over two years.

• 15% obtained training in LATU SENSU undergraduate courses, lasting 360 hours or more.

• 9% chose not to answer the question.

These data raise significant concerns, as 60% of professionals reported having less than 360 hours of training in ultrasonography, while the previous Graph 2 indicates that the ideal training requires a minimum of two years. Therefore, the majority of respondents appear to lack the appropriate training time to become proficient in this field.

This situation raises important questions about the qualification of professionals in ultrasonography and underscores the need to establish stricter training and certification standards to ensure the quality of services provided to patients. The pursuit of a more solid and comprehensive training is essential to enhance the practice of ultrasonography and promote advancements in diagnostic medicine (Table 5).

The pursuit of specialization courses (postgraduate lato sensu) is a trend observed among ultrasonography professionals, not only as a means of enhancing their knowledge in this field but also as a way to establish a strong professional identity. This quest for specialization plays a crucial role in advocating for the profession of medical ultrasonographer.

Over more than three decades, we have played an active role in the training of professionals in ultrasonography. During this period, over a third of the practicing ultrasonographers in Brazil, estimated at 90 thousand physicians, have undergone our training. Since 2003, we have been offering specialization programs in ultrasonography (postgraduate lato sensu), which means that approximately 18 thousand physicians have completed these specializations in our institution.

Furthermore, since 2010, we have been promoting the

Advanced Course, which is recognized by the Brazilian College of Radiology (CBR). Currently, we offer 15 slots per year, over two years, totaling a workload of 4,200 hours. This course is considered a comprehensive training for medical ultrasonographers, as all participants, at the end of the two years, take the General Ultrasonography title examination offered by the CBR and pass it.

In 2022, we established a partnership with the Brazilian College of Radiology, under the presidency of Prof. Dr. Valdair Muglia, and with the current president of the Brazilian Society of Ultrasonography (SBUS), Prof. Dr. Rui Gilberto Ferreira. The aim of this partnership was to bring ultrasonographers together and strengthen the field of Ultrasonography.

In 2023, in Brazil, which currently has 546,000 physicians, averaging about 2.56 doctors per inhabitant, it is estimated that there are approximately 90,000 ultrasonographers and around 51,350 ultrasound machines, representing an average of 1.75 ultrasonographers for each device. Ideally, considering two doctors per device, we would expect to have 102,700 ultrasonographers in Brazil. It's important to note that ultrasonography has been recognized as a specialized field, being the most recent specialty established by the Federal Council of Medicine (CFM).

Regarding the role of ultrasound in medical specialties, we believe that a physician's training should include obtaining a specialist title in the specific area of the specialty, a postgraduate specialization in that field, and proof of at least one year of experience in ultrasound within that specialty. These criteria are essential to ensure the quality and competence of professionals working in this important field of diagnostic medicine.

CONCLUSION

The profession of ultrasonographer in Brazil has undergone various transformations and challenges over the years. The rapid advancement of technology in the field of ultrasound has led to a significant increase in demand for qualified professionals capable of conducting precise exams and contributing to the diagnosis and treatment of a wide range of medical conditions.

It has been observed that currently, the majority of physicians working in ultrasonography have less than six years of experience in this field, reflecting the relatively recent emergence of this specialty within medical semiology. However, the pursuit of specialized courses, such as Postgraduate Lato Sensu, has become a trend among ultrasonographers, not only as a way to enhance their skills but also to strengthen their professional identity and the advocacy for the profession.

The training of these professionals is crucial to ensure the quality of ultrasonography exams and proper patient care. Although there are different types of courses and training programs, it is evident that a significant portion of ultrasonographers have less than 360 hours of training in the field. It is important to highlight that more comprehensive courses, such as postgraduate Lato Sensu specialization and the course of improvement recognized by the CBR, play a fundamental role in the training of competent ultrasonographers.

The partnership between the Brazilian College of Radiology (CBR) and the Brazilian Society of Ultrasonography (SBUS) demonstrates a joint effort to strengthen the field of ultrasonography in Brazil. Ultrasonography is recognized as a medical area of expertise, and the pursuit of specialist titles in the specific specialty area, postgraduate Lato Sensu specializations, and proven experience are essential criteria for physicians who wish to excel in this area.

The training of a general ultrasonographer should follow a two-year training program with well-defined guidelines. This program should prepare the ultrasound physician to work not only in outpatient settings but also in hospitals, providing comprehensive practical training that allows them to take an assessment exam in which they can demonstrate their solid knowledge and clinical skills.

Furthermore, it's important to consider that ultrasound physicians with more than four years of experience in the field may be allowed to take the general ultrasound certification exam. To do so, they should be recommended by other qualified ultrasound professionals who can attest to their competence and experience.

When it comes to Ultrasound serving as a medical subspecialty within a specific field, professionals must meet additional requirements. They need to obtain the specialist title in the specific specialty area in question, complete a postgraduate course in that specific area, and be recommended by experienced ultrasound physicians who can validate that the professional has been working in the field for more than one year.

These measures are aimed at ensuring that ultrasonographers have the proper training and experience to provide high-quality services, whether they are working as general ultrasonographers or in specialized areas. These guidelines promote excellence in the practice of ultrasonography.

Despite the challenges and the need for improving the training and qualification of ultrasonographers, the profession plays a crucial role in diagnostic medicine, contributing to the early detection of diseases and effective patient monitoring. With a growing number of physicians seeking to enhance their skills in ultrasonography, it is expected that the quality of exams and patient care will continue to improve, benefiting the overall health of the population.

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ORIGINAL ARTICLE

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THYROID NODULES WITH AN INDICATION FOR FINE NEEDLE ASPIRATION CYTOLOGY, WHICH SHOWED BENIGNITY - PICTORIAL ESSAY

THATIANY PASLAR LEAL, HUSSEIN ALI TAHA, LEONARDO DE SOUZA PIBER

ABSTRACT

Introduction: The thyroid is an endocrine gland that regulates thyroid hormones. Thyroid nodules are common, detected by ultrasound (US). Malignancy is rare, and the diagnosis depends on fine needle aspiration cytology (FNAC). ACR TI-RADS stratifies the risk of malignancy based on ultrasound findings.

Objective: Presenting images of thyroid nodules classified as TI-RADS 3, 4, and 5, highlighting their confirmed benignity through FNAC.

Methods: Images of TI-RADS 3, 4, and 5 nodules were collected from a diagnostic center, with FNAC indicating benignity.

Results and Discussion: The images show TI-RADS 4 and 5 nodules with highly suspicious findings, but with benign cytology. This emphasizes the need for a comprehensive evaluation, considering the nodule's size. Echogenic foci increase suspicion but are not definitive. Similarly, isolated shape doesn't guarantee malignancy. The decision for FNAC should take into account various characteristics.

Conclusion: The study highlights the importance of an individualized evaluation of nodules, even in TI-RADS 4 and 5 with suspicious findings. There is no definitive characteristic of malignancy, and size influences the decision for FNAC. ACR TI-RADS is useful but should be used in conjunction with clinical assessment to avoid unnecessary procedures. With advanced ultrasound techniques, the management of thyroid nodules has improved.

KEYWORDS: ACR TI-RADS; THYROID; ULTRASONOGRAPHY; THYROID ULTRASONOGRAPHY; ONCOCYTIC CYTOLOGY.

INTRODUCTION

The thyroid is an endocrine gland responsible for iodine uptake, and its microscopic architecture provides it with the ability to secrete and store thyroid hormones. Among the diseases that affect the thyroid, thyroid nodules are common and highly prevalent [1-3].

They are found in approximately 8% of the adult population through palpation, 40% through ultrasound (US), and in 50% of the population through pathological examination and autopsy. Their detection has increased 2 to 4 times in the last three decades, primarily due to the advent of imaging techniques, particularly the increased use and advancement of ultrasound [3-7].

The malignancy of these nodules is relatively rare, being diagnosed in less than 10% of the nodules found. The diagnosis of malignancy depends primarily on fine needle aspiration cytology (FNAC). The appropriate indication for which nodules should undergo FNAC and which should be clinically followed can be determined using a system of stratification and progressive categorization that predicts malignancy based on ultrasound findings, the Thyroid Imaging, Reporting and Data System published by the American College of Radiology (ACR TI-RADS) [3, 8-15].

The characterization of a thyroid nodule based on the system defined by ACR TI-RADS ranges from 1 to 5 and is primarily centered around evaluating five morphological characteristics: composition, echogenicity, shape, margin, and echogenic foci. Each of these characteristics receives a score, which is then summed up to determine the classification that is related to the suspicion of nodule malignancy. The recommendation is directly linked to a sixth characteristic, which is the size of the nodule. The highest potential risk of malignancy falls under the category TI-RADS 5. In other words, the system follows a pattern of increasing severity, with nodules considered non-suspicious up to the classification of TI-RADS 2. TI-RADS 4 nodules are characterized by a wide range of morphological possibilities and are considered moderately suspicious [8,9-12,14,16-17].

Among the parameters evaluated on ultrasound, the following stand out: solid nodular composition greater than 50%; irregular shape; nodule visualization taller than wide; echogenic foci; solid eccentric portion; changes in echogenicity, such as marked hypoechogenicity; and solid nodules.

The shape of the nodule and the presence of echogenic foci, when considered individually, both receive a score of 3, classifying the nodule as TI-RADS 3, probably benign or mildly suspicious, with a minimum estimated risk of malignancy of 14.1%. The degree of suspicion for malignancy pro-

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gressively increases based on morphological findings, reaching 45% for TI-RADS 4 and 89.6% for TI-RADS 5, which is highly suspicious. Depending on the size, fine needle aspiration cytology (FNAC) or clinical follow-up is recommended [15-19]. The indication for FNAC for ACR TI-RADS 3, 4, and 5 occurs when they have a larger diameter of 2.5, 1.5, and 1 cm, respectively.

The TI-RADS aims to classify the risk of a nodule being malignant in order to assist the physician in making the most appropriate decision, which may include observation of the nodule, follow-up ultrasound monitoring, or recommending fine needle aspiration cytology (FNAC) for cytological analysis of the nodule. This system seeks to streamline the interpretation of images obtained by radiologists by establishing a practical and common risk stratification, thereby maintaining a consistent language that can reduce interobserver and interdevice variations as well as the confusion in report interpretation and findings [3,14-16]. This approach helps avoid unnecessary invasive procedures, such as FNAC, in a significant number of patients [11,15-19].

Thyroid ultrasound is the initial evaluation of the gland. FNAC, on the other hand, is a useful method, cost-effective, and less invasive for thyroid cancer detection. Most thyroid nodules are benign, and when they are malignant, they are mostly well-differentiated carcinomas.

OBJECTIVE

The objective is to display ultrasound images of thyroid nodules classified by ACR TI-RADS as mildly suspicious, moderately suspicious, and highly suspicious, corresponding to TI-RADS 3, 4, and 5, based on shape and/or the presence of echogenic foci, with cytology confirmed as benign through US-guided FNAC.

METHODS

This is a pictorial essay, meaning it consists of a collection of original images from the database of an imaging diagnostic center in São Paulo.

The eligibility criteria included nodules classified according to ACR TI-RADS as 3, 4, or 5, based on shape and/or the presence of punctate echogenic foci, with an indication for fine needle aspiration cytology. When these nodules were punctured for cytological evaluation, the results showed cellular benignity.

RESULTS AND DISCUSSION

The morphological characteristics that contributed to the scoring leading to the TI-RADS classification for each nodule will be highlighted.

In the shape category of ACR TI-RADS, the evaluation includes the relationship between the anteroposterior diameter (vertical in relation to the skin) and the horizontal diameter, measured in the transverse plane of the lobe. Nodules that are taller than they are wide, with a ratio greater than 1 for anteroposterior to horizontal diameter when measured in the transverse plane, have a higher suspicion of malignancy. On the other hand, benign nodules often have their major axis parallel to the skin, making them wider than they are tall.

Figure 1 corresponds to a 61-year-old woman with a nodule classified as ACR TI-RADS 4. The nodule is isoechoic, predominantly solid, well-circumscribed, and taller than it is wide, measuring 4.3 cm. Cytology revealed a benign follicular nodule associated with chronic lymphocytic thyroiditis, classified as Bethesda category II.

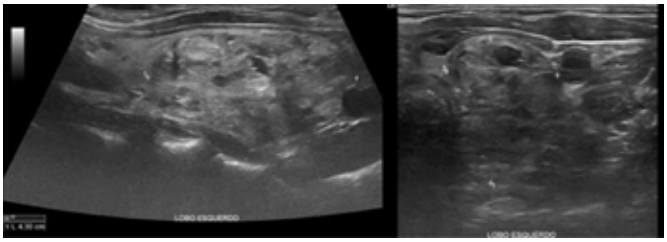

Figure 1 – Isoechoic nodule, predominantly solid, well-circumscribed, taller than it is wide. ACR TI-RADS 4.

Figure 2 refers to a nodule from a 40-year-old man. The nodule is isoechogenic, predominantly solid, circumscribed, taller than wide, just like figure 1, it is a TI-RADS 4. The indication for FNAC did not follow the ACR TI-RADS recommendation, as the nodule does not present more than 1.5cm, and a nodule suggestive of atypia of undetermined significance was revealed, classified as Bethesda III.

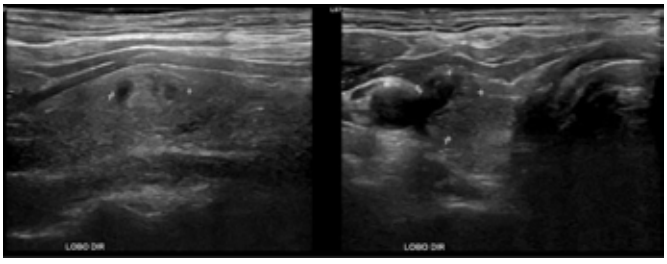

Figure 2 – Isoechoic nodule, predominantly solid, well-circumscribed, taller than it is wide. ACR TI-RADS 4.

Figure 3 is from a 50-year-old male patient, classified as ACR TI-RADS 5 and Bethesda category II, with findings suggestive of colloid goiter. The nodule is isoechoic, predominantly solid, well-circumscribed, taller than it is wide, and has punctate echogenic foci smaller than 1 mm.

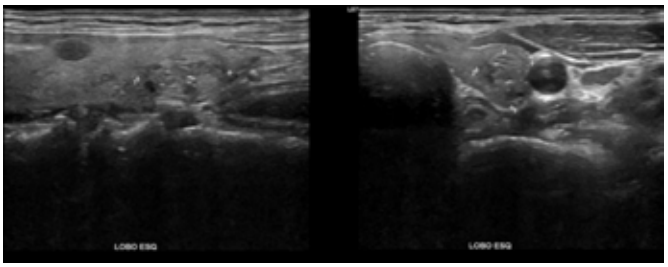

Figure 3 – Isoechoic nodule, predominantly solid, well-circumscribed, taller than it is wide, and with echogenic foci. ACR TI-RADS 5.

Echogenic foci are pinpoint findings of significantly increased echogenicity within the nodule and have been associated with both benign and malignant lesions. Although several studies demonstrate high specificity for the presence of echogenic foci in malignant nodules, this finding is not pathognomonic of malignancy. These are hyperechogenicities compared to the surrounding tissue and can vary in shape and size, as well as occur individually or in association with posterior acoustic shadowing artifacts.

Punctate echogenic foci or microcalcifications do not typically exhibit posterior acoustic shadowing. Macrocalcifications are large calcifications capable of generating posterior acoustic shadowing and may have irregular shapes. Peripheral calcifications are calcifications that occupy the periphery of the nodule, not necessarily continuous, and they usually produce acoustic shadows that obscure the central content of the nodule.

Figure 4 shows an isoechoic nodule, predominantly solid, wider than it is tall, and with echogenic foci, classified as ACR TI-RADS 4. When fine needle aspiration was performed, the cytological findings were suggestive of a follicular nodule, categorizing it as Bethesda category II.

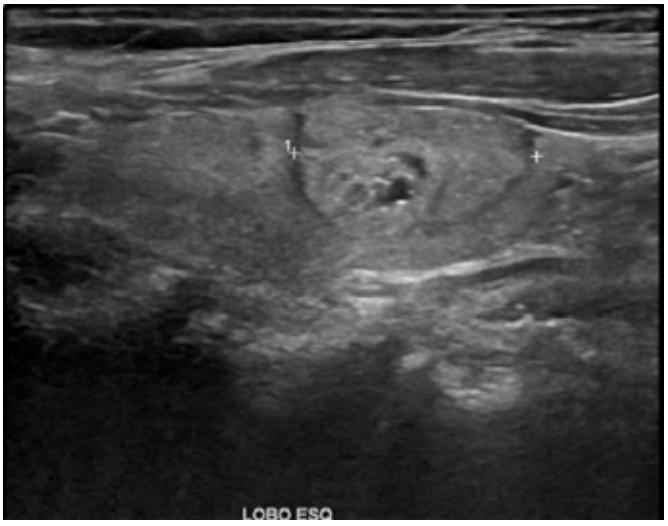

Figure 4 – Isoechoic nodule, predominantly solid, well-circumscribed, wider than it is tall, with echogenic foci. ACR TI-RADS 4.

Figure 5 shows a well-circumscribed, isoechoic/mixed nodule, wider than it is tall, with the presence of echogenic foci, classifying it as TI-RADS 4. This nodule is observed in a 23-year-old female patient.

When FNAC was performed, the findings were suggestive of atypia of undetermined significance, categorizing it as Bethesda III. This diagnosis occurs in approximately 30% of patients.

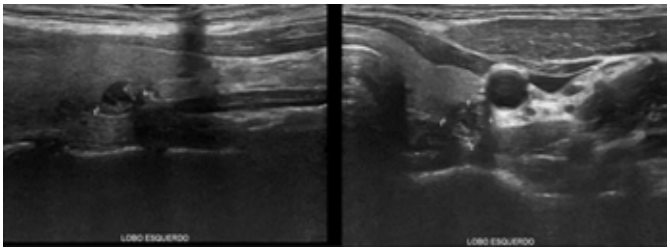

Figure 5 – Isoechoic/mixed nodule, well-circumscribed, wider than it is tall, with echogenic foci. ACR TI-RADS 4.

The nodules presented in this study classified as ACR TI-RADS 4 exhibit significant variability, with a predominant feature being isoechoic or predominantly solid composition, as evidenced in Figures 1, 2, 4, and 5. In Figure 3, the nodule had echogenic foci, was taller than it was wide, resulting in a 6-point increase in TI-RADS, classifying it as TI-RADS 5. This is different from Figure 4, where the nodule is wider than it is tall, and thus remains classified as TI-RADS 4.

Figure 6 displays a hypoechoic nodule (+2 points) in ACR TI-RADS. In addition to being solid, wider than it is tall (+3 points), and having echogenic foci (+3 points), the nodule is also irregular (+2 points), classifying it as TI-RADS 5. This nodule is from a 72-year-old female patient, and the cytological findings were suggestive of a benign follicular nodule, categorized as Bethesda category II.

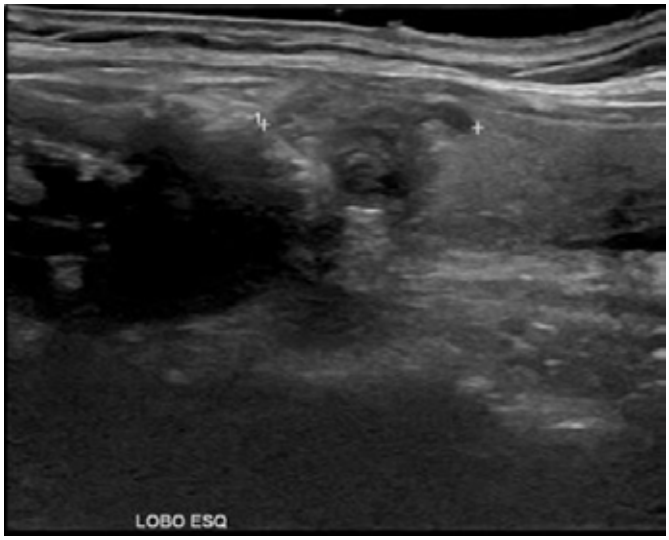

Figure 6 – Hypoechoic nodule, solid, well-circumscribed, wider than it is tall, irregular, with echogenic foci. ACR TI-RADS 5.

Figure 7 also shows a hypoechoic nodule, predominantly solid, taller than it is wide, without echogenic foci. Therefore, it scores 6, classifying it as TI-RADS 4. Since it was larger than 1.5 cm, FNAC was indicated, and the findings were suggestive of chronic lymphocytic thyroiditis, categorized as Bethesda category II.

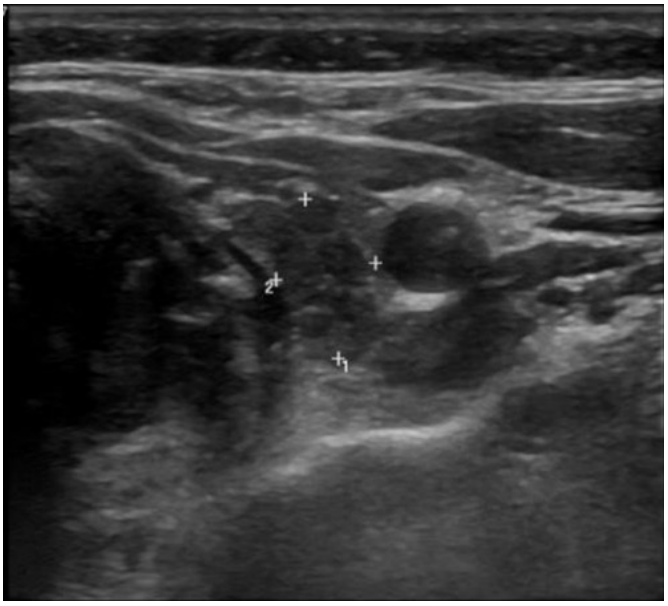

Figure 7 – Hypoechoic nodule, predominantly solid, well-circumscribed, taller than it is wide. ACR TI-RADS 4.

In Figure 8, the nodule is hypoechoic, solid, irregular, wider than it is tall, with the presence of discrete echogenic foci, classifying it as ACR TI-RADS 5. However, when FNAC was performed, the cytological findings were suggestive of atypia of follicular cells with rare papillary arrangements and nuclear grooves, categorizing it as Bethesda category III.

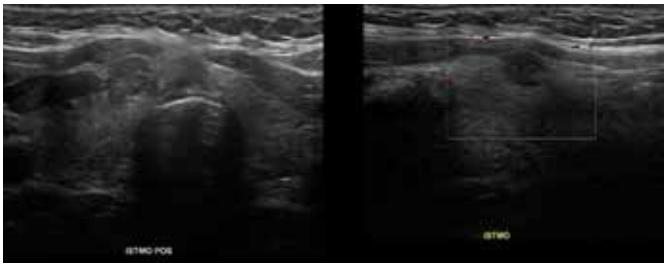

Figure 8 – Hypoechoic nodule, solid, irregular, wider than it is tall, with echogenic foci. ACR TI-RADS 5.

The nodule in Figure 9 is isoechoic to the thyroid gland parenchyma and predominantly solid, which earns it 2 points in ACR TI-RADS. The nodule is taller than it is wide and has echogenic foci, classifying it as TI-RADS 5. Despite the high suspicion of malignancy, when FNAC was performed, the findings were suggestive of colloid goiter, categorized as Bethesda category II. This is a benign nodule with low malignancy risk, and therefore, the standard management is clinical and ultrasound follow-up at the determined intervals.

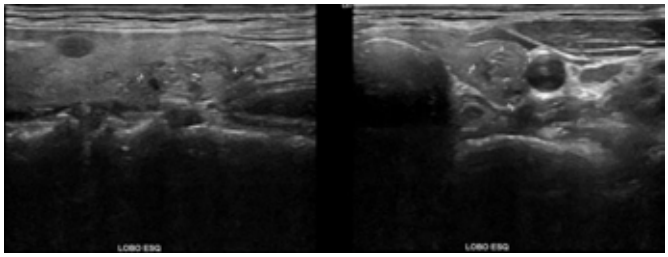

Figure 9 – Isoechoic nodule, solid, taller than it is wide, with echogenic foci. ACR TI-RADS 5.

Figure 10 is from a 33-year-old female patient with a hypoechoic nodule, meaning it appears less echogenic than the surrounding thyroid parenchyma. It is nearly completely solid, well-circumscribed, taller than it is wide, and has echogenic foci. It is classified as ACR TI-RADS 5, and the cytological findings were suggestive of a benign follicular nodule associated with oncocytic metaplasia, categorized as Bethesda category II.

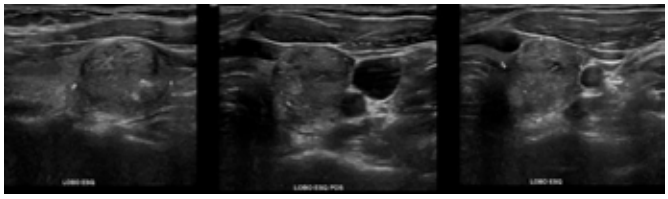

Figure 10 – Hypoechoic nodule, solid, taller than it is wide, with echogenic foci. ACR TI-RADS 5.

Evaluating the figures in which echogenic foci are present, it is possible to demonstrate that these findings elevate the ACR TI-RADS, often associated with categorization 4 or 5, and do not necessarily correspond to microcalcifications.

In Figure 11, there is a hypoechoic nodule that is predominantly solid, well-circumscribed, taller than it is wide, with discrete macrocalcifications present. Therefore, it is classified as TI-RADS 5, and the cytology result is Bethesda category II.

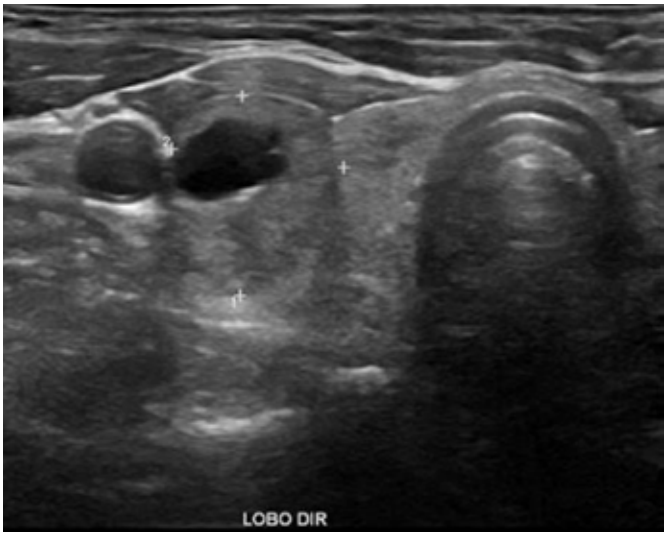

Figure 11 – Hypoechoic nodule, predominantly solid, well-circumscribed, taller than it is wide, with macrocalcifications. ACR TI-RADS 5.

In Figure 12, we observe a TI-RADS 5 nodule that is hypoechoic, solid, irregular, and taller than it is wide in a 40-year-old female patient. The Bethesda classification for this nodule was category III, which represents an indeterminate cytology result.

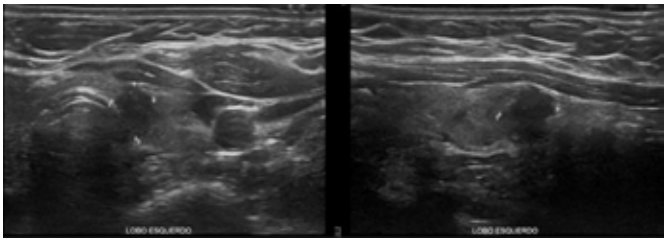

Figura 12– Nódulo hipoecogênico, sólido, irregular e mais alto que largo. ACR TIRADS 5.

CONCLUSION

With the advent of and improvements in ultrasound techniques, thyroid nodules are more easily evaluated and diagnosed. There is no single morphological characteristic that is pathognomonic of malignancy. Therefore, it is the responsibility of the imaging specialist to recognize the morphological diversity of nodules, regardless of their ACR TI-RADS classification. Specifically, when it comes to grades 3, 4, and 5, the range of variations should be observed.

Regarding the findings of echogenic foci and shape, although they score more points than other criteria, when present, they do not guarantee malignancy but rather increase suspicion. Based on this, taking into account the nodule's size, it may be indicative of PAAF. As demonstrated in the presented cases, when aspiration is performed, it can reveal benignity.

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CASE REPORT

PRIMARY CUTANEOUS FOLLICLE CENTER LYMPHOMA AND THE HIGH-FREQUENCY ULTRASOUND AS A DIAGNOSTIC TOOL

ATHOS MARTINI, ARIEL ROSA, TELMA SAKUNO, GABRIELLA FUNCHAL, ESTEVÃO VANZO, MATHEUS PACHECO

ABSTRACT

This case report describes the use of high-frequency ultrasound (HFUS) as a diagnostic tool for cutaneous lymphomas. Cutaneous lymphomas are classified into T-cell and B-cell lymphomas, with B-cell lymphomas characterised by few lesions with rapid growth.

The patient in this case report presented with an intensely vascularized reddish-brown nodule on the left shoulder. HFUS revealed a heterogeneous tumour lesion located in the epidermis and subcutaneous, infiltrating the adjacent muscles with increased vascularization. Computed tomography (CT) confirmed the presence of an expansive lesion. Anatomopathological examination revealed a primary cutaneous follicle center lymphoma. A finding of interest was the presence of the Grenz zone, which was seen on both ultrasound and histopathology.

While HFUS has been used for various dermatological conditions, there is limited data available on its use for skin lymphomas.

This case report highlights the potential use of HFUS as a non-invasive, repeatable, and objective monitoring tool for cutaneous lymphomas.

KEYWORDS: HIGH-FREQUENCY ULTRASOUND; CUTANEOUS LYMPHOMAS; DERMATOLOGICAL ULTRASOUND; SKIN ULTRASOUND

INTRODUCTION

Cutaneous lymphomas are classified according to their cellular origin into T-cell lymphoma and B-cell lymphoma. The annual incidence rate is 0.3 per 100,000 inhabitants. 65% of cases are T-cell, 25% are B-cell, and 10% are true histiocytic lymphomas or other rare types of lymphomas. From a dermatological perspective, B-cell lymphomas are characterized by few lesions, typically nodules or infiltrates, and they tend to exhibit relatively rapid growth. [1,2].

There are few reports in the literature regarding the diagnosis of cutaneous lymphomas using ultrasound. Our aim with this report is to highlight high-frequency ultrasound as a potential diagnostic and monitoring tool for cutaneous lymphomas.

CASE REPORT

Male patient, 43 years old, construction worker. He denied any comorbidities, continuous medication use, or allergies. No history of smoking, alcohol consumption, or prior skin cancer. He reported having the lesion on his left shoulder for five months. The condition began a year before with the appearance of similar lesions on the right shoulder, anterior chest, and back, all of which spontaneously disappeared within a few months. He experienced a burning sensation and occasional itching in the affected area, with no change in the mobility of the affected shoulder. The patient mentioned recurrent cervical lymphadenopathy during this period. No weight loss reported, and serologies were negative.

On physical examination, he presented with an intensely vascularized erythematous-brownish nodule, along with some papules on the surface. The lesion measured 22 x 15 cm and was located on the left shoulder. It felt warm, soft, and elastic to the touch (Figure 1). The only notable laboratory findings were elevated inflammatory markers (CRP and ESR), with no other noteworthy abnormalities.

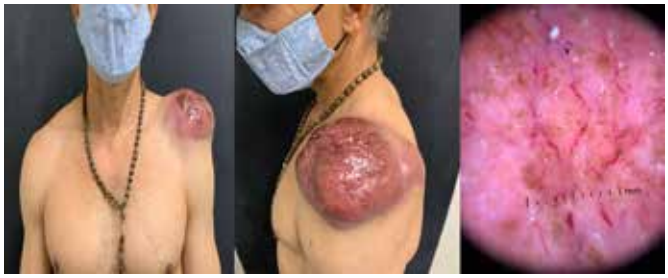

Figure 1: Clinical lesion on the right shoulder with dermoscopy (right).

High-frequency ultrasonography (HFUS) of soft tissues was performed with an 18MHz transducer and showed a heterogeneous tumor lesion located in the epidermis and subcutaneous cellular tissue, infiltrating the adjacent muscles and with increased vascularization at SMI (Superb Micro-vascular Imaging) (Figure 2).

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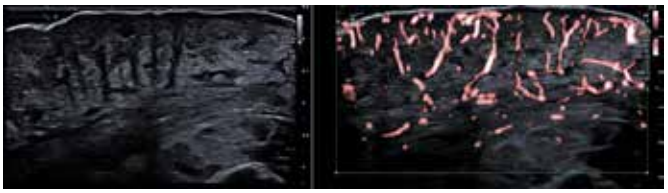

Figure 2: Heterogeneous lesion involving the dermis, subcutaneous tissue, and infiltrating, with increased vascularity on SMI.

The computed tomography (CT) scan revealed an expansive lesion in the right shoulder with soft tissue density and an infiltrative appearance. It was inseparable from the rotator cuff musculature and the deltoid muscle, measuring approximately 13.7 x 12.5 cm. It also showed some hypoattenuating areas suggestive of cystic/necrotic degeneration, along with diffuse lymphadenopathy.

The histopathological examination of the lesion revealed diffuse proliferation of atypical lymphoid cells, without affecting/respecting the epidermis, in addition to details of the inflammatory infiltrate of monomorphic cells, of medium to large size. Immunohistochemistry showed positive CD79 for all cells, B lymphocyte marker and CD10 and Bcl2: markers of systemic centrofollicular lymphoma. (Figure 3).

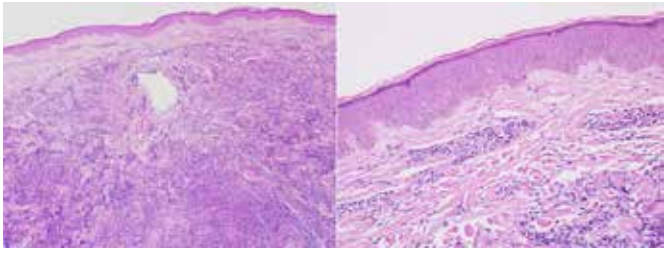

Figure 3: Histological sections of the skin reveal a highly cellular lesion involving the entire dermis and hypodermis, composed of atypical lymphocytes, including centrocytes and centroblasts, with a diffuse pattern of infiltration and rare residual follicular centers. The epidermis and a small portion of the papillary dermis (Grenz zone) are spared (HE, 10x / 40x).

One noteworthy finding in the case was the presence of the Grenz Zone (a spared dermal zone between the epidermis and the tumor) and its ultrasound translation as a hypoechoic band between the epidermis and the affected dermis (Figure 4).

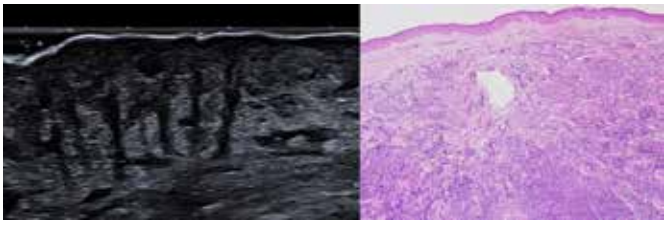

Figure 4: Correlation between ultrasound and histopathology. In both, it is possible to observe the intact epidermis and a zone of uninvolved dermis between the epidermis and the tumor (Grenz Zone).

Diagnosis of large B-cell lymphoma was confirmed, with a centrofollicular phenotype rich in T lymphocytes, with cutaneous infiltration. The patient maintained follow-up with hematology, who began the R-CHOP chemotherapy protocol.

DISCUSSION

The clinical history, physical examination and imaging tests such as HFUS help in the diagnosis of cutaneous lymphomas. Confirmation, however, is essentially obtained through histological and immunohistochemical examinations. Follicular center cells generally express CD20+, CD79a+, BCL-6+ and BCL-2- (possibly with a weak expression of BCL-2 in a minority of B cells), with variable expression of CD43 and CD106 [2,3]. Left untreated, the lesions will enlarge and may become locally aggressive [4].

Although HFUS has been available since 1979, it is increasingly being used in a growing number of applications, including clinical and experimental dermatology. [5]

HFUS (high-frequency ultrasound) is fast, non-invasive, and reproducible, making it an objective monitoring tool. Furthermore, since ultrasound images allow for quantitative assessment of treatment response, it can be used in numerous skin conditions, including inflammatory conditions(atopic dermatitis, psoriasis); it can also be used to guide therapeutic interventions in various dermatoses. To date, the oncological use of USG-AF has focused primarily on melanoma and non-melanoma skin cancers, with little data available on its use in cutaneous lymphomas [6].

To our knowledge, the ultrasonographic characteristics of cutaneous lymphomas and their similarity with histology have been little explored, although many articles on dermoscopic characteristics of cutaneous lymphomas have been published in recent years. Recently, ultrasonography has been proposed to monitor response to therapy in mycosis fungoides (a subtype of T lymphomas) [7].

As ultrasound characteristics, in cutaneous lymphomas of the dermal-epidermal layer, irregularly shaped hypoechoic areas were observed. These gaps presented intralesional vascularization [8]. In the case presented, what drew attention was the presence of preserved epidermis and a strip of normal dermis between the epidermis and the tumor lesion, which is translated into histopathology as the Grenz zone. This finding, in the context of the clinical hypothesis of lymphoma, leads us to think about B-cell lymphomas and not T-cell lymphomas (since the latter group presents as a characteristic epidermotropism, that is, the involvement of the epidermis by lymphocytes T, the discovery of the Grenz zone is not expected).

CONCLUSION

We have a limited arsenal of imaging tests for the diagnostic assistance of cutaneous lymphomas, and HFUS has proven to be an important and useful tool in this role. In this report, we emphasize the potential of high-frequency ultrasound as a diagnostic and monitoring instrument for cutaneous lymphomas. Additionally, we highlight the importance

of the correlation between ultrasound and histopathology to better understand the examination findings, such as the ultrasonographic translation of the Grenz Zone in the presented case.

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CASE REPORT

SEGMENTAL AGENESIS OF THE VAGINA AND FALLOPIAN TUBES: A CASE REPORT

WALDEMAR NAVES DO AMARAL[1], LUANA KRONIT BASTOS VIGGIANO[2], JOEDNA VIRGÍNIA BORGES GARCIA[3]

ABSTRACT

Müllerian anomalies are defined as the absence or underdevelopment of structures derived from the Müllerian duct, including the fallopian tubes, uterus, and the upper two-thirds of the vagina. It is considered a variant of Mayer-Rokitansky-Küster-Hauser Syndrome (MRKH) and the most common form of Müllerian dysgenesis. MRKH involves the agenesis or dysgenesis of the Müllerian portion of the vagina and uterus during embryogenesis, clinically presenting as primary amenorrhea, normal secondary sexual characteristics, a 46,XX karyotype, and a short vagina (depth of 1 to 2cm).

Diagnosis can be made through physical examination, hormonal profiling, and imaging studies. Studies suggest that the preferred first-line treatment should involve the use of vaginal prostheses for pressure dilation, with surgery reserved for cases where clinical treatment is unsuccessful. The objective of this study is to describe the case of a young patient who experienced severe pain in the hypogastric region, primary amenorrhea, and normal secondary sexual characteristics. During the diagnostic investigation, the occurrence of MRKH was confirmed.

KEYWORDS: MAYER-ROKITANSKY-KÜSTER-HAUSE SYNDROME, MÜLLERIAN AGENESIS, NORMAL SEXUAL CHARACTERISTICS, PRIMARY AMENORRHEA, HYPOGASTRIC PAIN

INTRODUCTION

Müllerian anomalies are defined as the absence or hypoplasia of structures derived from the Müllerian duct, including the fallopian tubes, uterus, and upper two-thirds of the vagina, and are considered a variant of Mayer-Rokitansky-Küster-Hauser Syndrome (MRKHS), which is the most common Müllerian dysgenesis. It is the second most common cause of primary amenorrhea, with an estimated incidence of approximately 1 in 4,500 female births[1,2].

MRKHS can be characterized by uterine muscular buds and normal fallopian tubes, either in its complete form or in its partial form, where uterine and fallopian remnants may be asymmetric. It was initially reported by Columbus in 1562. Later, Mayer in 1829 and Rokitansky in 1838 described the changes found during autopsies of what was then called "bipartite uterus." Kuster in 1910 suggested surgical therapy, and in 1962, Hauser described the syndrome, which consists of normal external genitalia, absent vagina, absent or rudimentary uterus, normal fallopian tubes and ovaries which may be associated with renal and skeletal abnormalities[4].

It is a rare condition in which patients have a 46,XX karyotype and normal secondary sexual characteristics, as the ovaries are present and functional, but menstruation does not occur. Its etiology is unknown, although there are hypotheses suggesting a genetic cause[2,11].

The syndrome is classified into three forms based on the involvement of structures beyond the reproductive system. The typical type, Type I, is characterized by changes restricted to the reproductive system. The second, Type II, is an atypical syndrome in which there is asymmetry in the uterus and anomalies of the uterine tubes. This form may be associated with ovarian disease, renal, bone, and congenital otological abnormalities. The third type, called MURCS, involves uterovaginal hypoplasia or aplasia, renal, bone, cardiac, and digital malformations. In the kidneys, there may be unilateral agenesis, horseshoe kidney, renal hypoplasia, ectopic kidneys, and hydronephrosis. In the bones, vertebral abnormalities are common, including vertebral fusion, particularly in the cervical region, Klippel-Feil syndrome, and scoliosis. Cardiac and digital abnormalities can also occur, such as syndactyly and polydactyly[2].

Surgical and non-surgical procedures allow the creation of a neovagina in patients, providing them with the opportunity to have a normal sexual life. Additionally, through assisted human reproduction techniques and uterine transplantation, women can have biological children[2,13].

In this present study, we present the case report of an 18-year-old adolescent with severe suprapubic pain, primary amenorrhea and normal secondary sexual characteristics, who underwent clinical and radiological investigation, leading to a diagnosis of MRKH syndrome, where surgical treatment was suggested and accepted in mutual agreement with the patient.

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CASE REPORT

N.E.O, 18 years old, female, white. She sought the gynecology service reporting intense pain in the hypogastric region and primary amenorrhea. She reported seeking healthcare services since the age of 14 due to amenorrhea complaints, without imaging investigation. At the age of 15, she began experiencing dysmenorrhea (pain scale: 10/10), associated with nausea and vomiting, pain in the lower limbs and lower back, with cyclic recurrence lasting one week per month. The patient sought the gynecology service again at the age of 18 with a significant worsening of pelvic pain, with no significant improvement with the use of analgesics, including a combination of non-steroidal anti-inflammatory drugs and strong opioids. The patient reported having her first sexual intercourse at the age of 18, denied dyspareunia or sinus bleeding.

Personal history with ongoing monitoring for keratoconus. Denies smoking and alcohol consumption, sedentary lifestyle. Regarding medications, she is taking continuous combined ethinylestradiol and cyproterone, as well as analgesics, including a strong opioid.

In the general physical examination, no alterations were observed, with no signs of chromosomal abnormalities. Pubertal development was classified as Tanner stage M5 P5. During the gynecological evaluation, vulvar inspection revealed the presence of small and large labia without a patent vaginal orifice (figure 1).

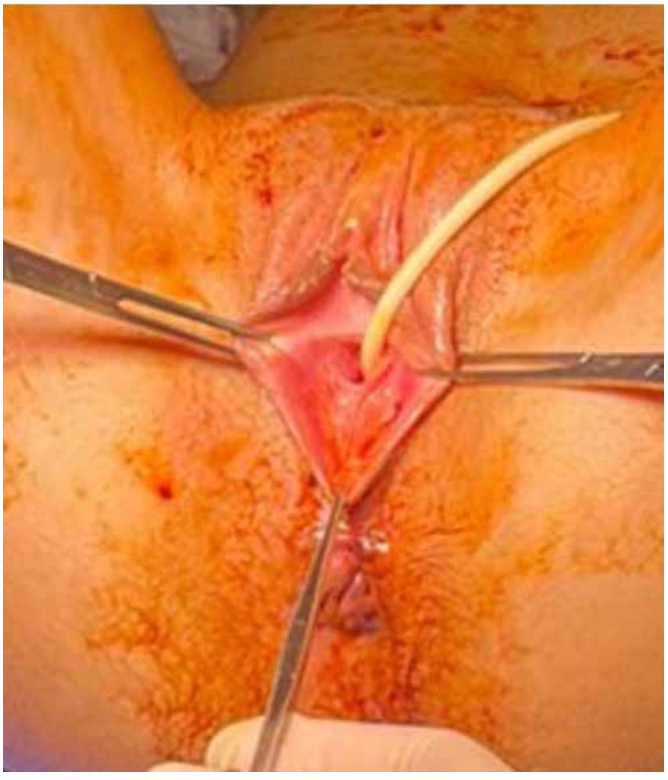

Figure 1: Image of external genitalia with the presence of labia minora and majora without changes. Absence of vaginal canal.

Complementary exams carried out

Endovaginal pelvic ultrasound, showing regular contours of the uterus and precise limits, measuring 11.00 x 5.70 x 6.90cm and a volume of 224.97cm³. Myometrium with heterogeneous texture. Endocervical canal closed. Bilateral ovaries without changes. Normal bilateral kidneys. Bladder visualized without abnormalities. (Figure 2)

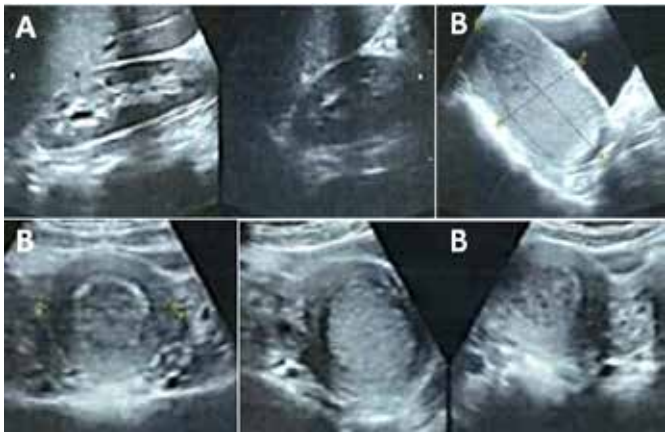

Figure 2: Images verified on abdominal ultrasound A: Normal kidneys. B: Large hematometra.

Pelvic magnetic resonance imaging: collapsed vagina, with the proximal and middle parts showing linear material (suspected fibrosis) with a distance of 5cm between the end of the vagina and the uterus, with a large amount of hematic material distending the endocervical canal, suggestive of hematocolpos.

After discussion with the patient, and aiming to improve incapacitating cyclic pain, the case was managed by performing a laparotomy to remove the rudimentary uterus. Follow-up appointments were scheduled for the creation of a neovagina.

Surgical procedure with a Pfannenstiel incision, no visualization of the fallopian tubes, confirmed the presence of bilateral ovaries with normal macroscopy. A single uterus distended by hematic content was observed, along with lesions compatible with endometriosis in pelvic organs, without communication with the vagina and without the presence of a uterine cervix. Clamping, sectioning, and ligature of the round ligaments, uterine vessels, cardinal ligaments, and uterosacral ligaments were performed. The complete release of the rudimentary uterus was achieved after releasing fibrosis linked to the proximal region of the vagina and the distal part of the uterus, with no access to the vagina. The opening of the surgical specimen, uterus, showed a volume of hematic content. (figures 3 and 4)

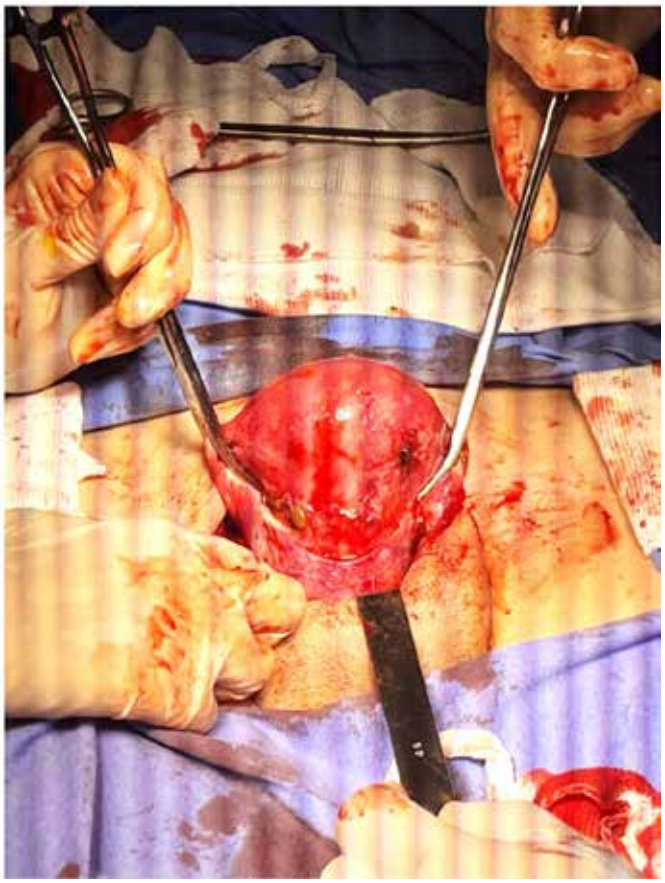

Figure 3: Intraoperative image of a rudimentary uterus with hematometra. No visualization of uterine tubes. Presence of hyperchromatic images suggestive of endometriosis lesions.

Figure 4: Images of a rudimentary uterus. In the left image, there is no presence of the uterine cervix. In the right image, there is a presence of abundant hematologic content.

DISCUSSION

MRKHS is a rare condition, with an incidence of 1 in 4,500 female births, making it the second most common cause of primary amenorrhea, after gonadal dysgenesis. It is defined as the absence or hypoplasia of structures derived from the Müllerian duct, including the fallopian tubes, uterus, and the upper two-thirds of the vagina, with MRKHS being the most common form of Müllerian dysgenesis[2,5].

The Müllerian and Wolffian ducts are the embryological precursors of the female and male internal reproductive systems, coexisting undifferentiated in the embryo until genetic determinants lead to their differentiation into ovaries or testes. In females, the Müllerian ducts differentiate into the Fallopian tubes, uterus, cervix, and the upper portion of the vagina[1,2], while the Wolffian ducts degenerate. When the formation and differentiation pathways of the Müllerian ducts are compromised during embryonic development, various Müllerian anomalies can occur, ranging from minor anatomical variations to complete aplasia of the structures that make up the female reproductive system. Within the spectrum of Müllerian anomalies, the most prevalent is vaginal agenesis, present in 90% of cases of malformations. It results from an inhibitory defect in the correct embryonic development of the paramesonephric ducts and can be associated with uterine anomalies, including agenesis, hypoplasia, duplication, or even a normal uterus, variability that characterizes MRKHS[7,12]. MRKHS occurs when both Müllerian ducts fail to develop, resulting in a rudimentary solid uterus and a non-patent vagina in patients with a 46 XX karyotype, normal Fallopian tubes, and ovaries[2,3].

Müllerian aplasia and incomplete Müllerian fusion are associated with the familial occurrence of the most common Müllerian differentiation disorders in girls. Its cause is not yet well defined but is related to some genes. It is evident that the HOX genes, a family of regulatory genes that encode transcription factors, are essential for the correct development of the Müllerian duct during embryonic development, and WNT4 may participate in uterine development, as a mutation of WNT4 has been reported in cases of MRKHS with hyperandrogenism[3,13].

While most cases are sporadic, the increasing number of familial cases, the pattern of congenital malformations involved in the syndrome, and the association with chromosomal rearrangements indicate that genetic factors may trigger the development of the syndrome. The most frequently suggested mode of transmission of SMRKH is autosomal dominant with incomplete penetrance and variable expressivity due to a single gene mutation. The association of Müllerian dysgenesis with various extragenital anomalies suggests that key genes involved in fetal development and sexual differentiation, such as HOX, WNT, and those encoding anti-Müllerian hormone and its receptor, may be involved in the development of the syndrome. For first-degree relatives, the risk of recurrence is 1-5%[2,3].

Generally asymptomatic during childhood, Müllerian malformations are mostly detected in adolescence, with the average age of diagnosis being between 15 and 18 years, reflecting the typical late diagnosis of these congenital anomalies[7].

The typical presentation is characterized by primary amenorrhea, with or without cyclic pain, such as complaints of hypogastric, lumbar, or pelvic pain, in patients who normally go through puberty, i.e., breast development and pubic hair

growth with normal stature, being, therefore, a pure female type but without menstruation, and without signs of virilization. In these patients, almost always, there is an outline of the vagina in the distal segment, which guides our treatment indication. The gynecological examination may detect the absence of the vaginal canal or vaginal shortening[2,5,7].

In cases where vaginal agenesis occurs with normal uterine development (6-10%), the diagnosis can be made early. The presence of a functioning uterus with obstruction of the menstrual outflow tract results in hematometra, characterized by intermittent pelvic and lower back pain occurring approximately every 3-4 weeks. This atypical and rare presentation of MRKHS is associated with normal pubertal development, endocrine status, and external genitalia. It begins with intense, intermittent, and monthly hypogastric and lower back pain, consistent with hematometra, at the age of [13]. Therefore, the age of diagnosis can be early[7,11].

In the presence of clinical suspicion, a meticulous physical examination is the first and essential step in establishing the diagnosis of the aforementioned syndrome. This examination reveals the development of secondary sexual characteristics at a normal stage for the patient's sex and age, a vulva with normal formation, and complete or partial absence of the vagina. Complementary tests show the 46XX karyotype, and pelvic imaging tests confirm the presence of normal ovaries and a rudimentary uterus. Ultrasound, even through the suprapubic abdominal route, can be sufficient in many cases. If there is doubt, magnetic resonance imaging (MRI) should be performed to define the diagnosis, as it has higher sensitivity and specificity in evaluating the syndrome. These tests will typically reveal the presence of symmetric or asymmetric uterine agenesis and complete absence or marked hypoplasia of the upper and middle portions of the vagina. Since it results from a different embryonic precursor, the lower third of the vagina is present, and this vaginal remnant may have varying depths (2-7 cm). Laparoscopy is indicated only when assessment by the two previous methods is unsatisfactory, and when it is possible to formulate a therapeutic plan through this procedure[2,5,7].

When diagnosing MRKHS, it is important to consider and exclude differential diagnoses in situations where the patient has primary amenorrhea and developed secondary sexual characteristics. This includes conditions like congenital absence of the uterus and vagina, isolated vaginal atresia with androgen insensitivity syndrome, and transverse vaginal septum with imperforate hymen[5,10].

In the investigation of the described case, in line with the literature, continued after the physical and gynecological examinations which raised suspicion of a possible genital tract malformation, the investigation proceeded with a transvaginal pelvic ultrasound, which suggested the diagnosis of segmental agenesis of the vagina and uterine tube (a Rokitansky variant) with a significant hematometra. In other words, there was the presence of vaginal agenesis with hematometra and intact adnexal regions (normal ovaries).

The diagnosis of these cases is not complete without investigating the possible presence of associated systemic malformations, which are evident in cases of the atypical form of the syndrome. In the investigation of this case, a renal and bladder ultrasound was performed, which, in the absence of findings, ruled out the association with ureterovesical malformations. With the absence of other malformations, the patient likely had a variant of MRKHS[6,7].

This is a syndrome with a significant psychological impact on young affected women, not only due to its clinical manifestations but also because of its interference with sexual life and the ability to conceive. It's important to highlight the psychological changes caused by the anatomical alterations that characterize it, leading to distress, anxiety, psychological consequences, and a decreased quality of life for patients after the diagnosis is confirmed. Infertility is the most challenging aspect to accept, which is why a multidisciplinary approach is necessary. The therapeutic approach for these cases will involve not only the repair of congenital anatomical defects but also a multidisciplinary assessment that includes psychological support as an integral part of treatment[5,7].

The recommended anatomical treatment is the creation of a neovagina, either through surgical or non-surgical means, which can allow these patients to have a normal sexual life. When the surgical approach is chosen, uterine remnants may be removed to prevent future endometriosis. For patients who wish to have children, adoption should be encouraged, and they should be presented with the possibility of having biological children through assisted reproductive techniques[5,8].

The Frank method and surgical neovaginoplasty (Vecchietti method) are the most commonly mentioned options in the literature for the treatment of Rokitansky syndrome. In the Division of Gynecological Clinic at the Department of Obstetrics and Gynecology of the Hospital das Clínicas at the University of São Paulo Medical School, the first choice for treating this syndrome is the Frank method, which involves progressive dilation of the vaginal canal with a rigid acrylic mold. When the patient adheres to the method and does it correctly, a functional vagina for sexual intercourse can be achieved in approximately six months, on average[2,15].

The choice of method still depends on the surgeon's preferences. Among the possible techniques is the Vecchietti procedure or traction neovaginoplasty, which does not require an external tissue graft and can be performed laparoscopically. However, this procedure carries potential complications related to the traction threads placed in the vesicorectal space and possible posterior vaginal prolapse. The Davydov three-stage technique involves abdominal mobilization of peritoneum, fixation of the peritoneum to the vaginal introitus, and closure that sutures the top of the new vagina. While the Davydov procedure is advantageous in terms of granulation and healing in the neovagina, the neovaginal tissue lacks lubrication, and the procedure carries the risk of intestinal and bladder injury. Intestinal neovaginoplasty typically uses the sigmoid colon and provides lubricated tissue with an excellent blood supply; however,

the procedure requires intestinal anastomosis and is associated with complications, including significant vaginal discharge, postoperative ileus, intestinal obstruction, intestinal ulceration, risk of malignancy, and colitis. The McIndoe procedure allows a vaginal approach to create the neovagina. Various types of graft materials have been used for the McIndoe technique, including autologous skin grafts, typically from the buttocks or thigh, amnion, peritoneum (Davydov procedure), autologous in vitro vaginal tissue, and labial or gracilis myocutaneous flaps. In this procedure, we modified the donor area to be the abdomen, allowing primary closure and a discreet scar. To prevent stenosis and achieve proper graft fixation in the recipient area and prevent shearing, we used a polyurethane foam. Various materials have been used to make these vaginal molds: a condom mold filled with cotton, a polyethylene bag filled with fiberglass wool, an inflatable vaginal stent, a vacuum-expandable condom mold, Surgi-Stuf, ORFIT "S" material, a polystyrene mold. With this surgical technique, we achieve an anatomically and functionally adequate neovagina[8,14].

There is a range of interventions available, including surgical and non-surgical options, that allow for the creation of a neovagina in patients. Regardless of the chosen modality, treatment should provide an anatomically and physiologically normal vagina with an appropriate length to ensure proper sexual function. However, treatment should only be initiated when the patient desires to start her sexual life. In this case, the most relevant need was to address the hematometra. Therefore, in the present case, therapeutic options were made based on three distinct points: treatment of the hematometra via laparotomy hysterectomy, a new intervention to provide functional sexual life, and addressing the inability to conceive[5,7,9].

CONCLUSION

The present case presented is a variant of MRKHS in which the uterus is preserved, the secondary sexual characteristics are within the normal range, and there are no associated systemic malformations. This condition has a significant impact on the patient's life.

The inherent difficulty in diagnosing the spectrum of Müllerian malformations is evident. Among which, it describes a condition that requires early diagnosis mainly at the clinic, where ultrasound may be sufficient in many cases to confirm the diagnosis. And if there is any doubt, MRI should be performed together. In this investigation, we can identify which type of Müllerian malformation the patient has, so that appropriate surgical planning or non-surgical interventions can be proposed, including multidisciplinary assessment, due to the great psychological impact of these patients.

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CASE REPORT

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SURPRISING IDENTIFICATION OF MECKEL'S DIVERTICULUM WITHIN A FETAL OMPHALOCELE DURING PRENATAL CARE

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ABSTRACT

Omphalocele is a rare congenital abdominal wall defect that results in herniation of bowels, stomach, and even liver for inside the umbilical cord. Herein we present a case of a Meckel's diverticulum (MD) which was unexpectedly identified inside an omphalocele of a fetus. The mother was a 25-year-old pregnant woman referred due to a fetal omphalocele, which was first visualized at 16 weeks of gestation. On ultrasonography at 21 weeks and 6 days, there was an apparently abnormal circular-shaped image inside the umbilical cord, suggestive of an omphalocele. Fetal MRI confirmed this diagnosis and revealed that some small intestine segments, and portions of mesentery and transverse colon were in an extra-abdominal situation. Moreover, a hyperintense image in T1 and T2 sequences, measuring about 3.9 x 3.8 x 3.6cm, was observed inside the omphalocele, which was compatible with a MD. The child was born by cesarean section, at 40 weeks gestation, weighing 4,030g. The omphalocele was surgically closed, with resection of the MD followed by a terminoterminal anastomosis of the ileum, on the second day of life. The pathology was also compatible with the diagnosis of MD. This report highlights the importance of the proper determination of the omphalocele content still during the prenatal period, since this may present unexpected abnormalities, such as a MD, and thus modify the gestational management as well as birth planning and postnatal care.

KEYWORDS: MECKEL'S DIVERTICULUM; PRENATAL DIAGNOSIS; OMPHALOCELE; ULTRASOUND; MAGNETIC RESONANCE IMAGING

INTRODUCTION

Omphalocele is a rare congenital defect of the abdominal wall that results in the herniation of intestines, stomach, and sometimes even the liver, into the umbilical cord. It has been reported in 3.38 out of 10,000 pregnancies[1]. Although omphaloceles can occur as isolated anomalies, up to 70% of them are associated with other malformations or syndromes[2]. Their diagnosis can often be made prenatally, typically through ultrasound[1]. The presence of intestinal loops or protrusion of the liver and/or stomach into the umbilical cord after 11 weeks is considered non-physiological. After the diagnostic confirmation, the herniated contents should be evaluated, and magnetic resonance imaging (MRI) can be used for better visualization of anatomical details[2].

Here we present a case of Meckel's diverticulum (MD) unexpectedly identified within a fetal omphalocele.

CASE REPORT

A 25-year-old woman in her first pregnancy was referred for evaluation due to a fetal omphalocele visualized at [21] weeks. She reported smoking (about 5 cigarettes/day) and alcohol consumption in the first month of pregnancy. Additionally, she experienced frequent vaginal bleeding from the 2nd to the 4th month, some of them in large amounts, along with contractions in the third month, requiring bed rest. The husband was a 33-year-old, healthy, non-consanguineous man. There was no family history of congenital defects or genetic diseases.

The image of the omphalocele was first visualized at [16] weeks of gestation. Previous ultrasound examinations at 8 and 11 weeks had not described this finding. At [21] weeks and 6 days of gestation, an apparently abnormal circular-shaped image was observed within the umbilical cord (Figures 1A and 1B). However, at 30 and 34 weeks, only intestinal loops were seen inside the omphalocele. In the last examination at 37 weeks, the omphalocele measured 5.4 x 5.3 x 4.6 cm. Subsequent fetal MRI revealed the defect in the closure of the anterior abdominal wall, located in the umbilical region, apparently covered by a membrane, measuring about 1.7 cm. These findings were consistent with an omphalocele. In addition, there were some segments of the small intestine, along with a small portion of the mesentery and a segment of the transverse colon, in an extrabdominal position. An asymptomatic and hyperintense image was observed on T1 and T2 sequences, measuring about 3.9 x 3.8 x 3.6 cm, indicative of a Meckel's diverticulum (MD) located within the

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omphalocele (Figures 1C and 1D). The fetal karyotype (46, XY) and Doppler echocardiography were normal.

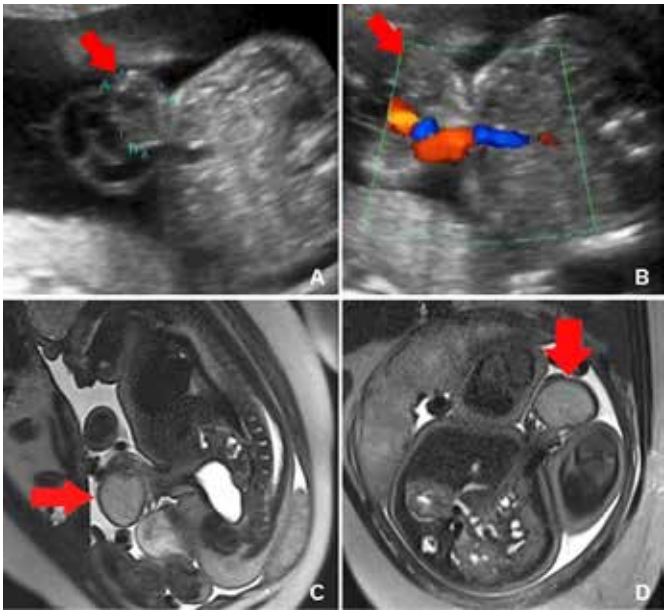

Figure 1. Fetal ultrasound at 21 weeks and 6 days showing the omphalocele with an apparently abnormal circular-shaped image within the umbilical cord (red arrows) (Fig. 1A and 1B). Fetal MRI revealing the omphalocele. There was a hyperintense image inside it, on T1 and T2 sequences, consistent with a Meckel's diverticulum (red arrows) (Fig. 1C and 1D).

The child was born via cesarean section at 40 weeks of gestation, weighing 4,030 g, and had Apgar scores of 10 at the first and fifth minutes. The child underwent omphalocele closure surgery with resection of the Meckel's diverticulum on the second day of life (Figures 2A and 2B). An ileal anastomosis was performed 18 cm from the ileocecal valve (Figure 2C). The pathological examination of the lesion confirmed the diagnosis of Meckel's diverticulum.

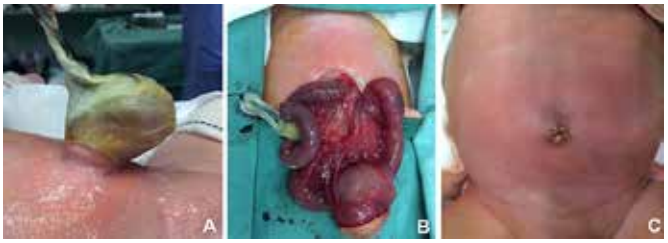

Figure 2. Postnatal appearance of the omphalocele, observed immediately after birth and before surgery (Fig. 2A). In Figure 2B, the contents of the omphalocele, with a focus on the Meckel's diverticulum (MD), can be seen. Figure 2C shows the final appearance of the patient's abdomen after surgery.

DISCUSSION

Meckel's diverticulum (MD) is a congenital anomaly characterized by a true diverticulum that involves all layers of the intestine and is part of the spectrum of anomalies resulting from incomplete closure of the omphalomesenteric duct, which develops during the 6th week of embryogenesis. Its prevalence in the general population varies between 0.3% and 2.9%, with a predominance in male patients in a ratio of approximately 1.5 to 4 times more cases in males than in females.

Meckel's diverticulum (MD) can be located between 7-200 cm proximal to the ileocecal valve (with an average of 52.4 cm) and may have a length ranging from 0.4-11 cm, and a diameter of 0.3-7 cm. Ectopic gastric and pancreatic tissues have been described in patients with MD, and these findings are related to symptomatic cases, primarily due to the occurrence of bleeding[3]. Additionally, MD has been reported in association with omphalocele[4], as observed in our patient.

Despite the frequency of Meckel's diverticulum (MD) in the general population and it being the most common congenital anomaly of the gastrointestinal tract (2-3% of the population), its prenatal diagnosis, as reported in the present case, is considered rare[4,5]. This may occur because ultrasound, although widely used for screening during pregnancy, may not be able to adequately distinguish the structures present inside the umbilical cord. Additionally, MD can be associated with other umbilical cord anomalies, including omphalocele4, as observed in our case.

In fetal ultrasound evaluation, MD appears as an ovoid, anechoic structure containing fluid inside, with the appearance of a wall with multiple layers. Doppler imaging does not show flow inside it. The echogenicity of the mass may increase due to debris, becoming hyperechoic in mid-pregnancy and isoechogenic at term due to the presence of meconium inside the intestinal loops. This can make its visualization challenging and, thus, prevent the diagnosis of MD[4-6].

Regarding the differential diagnosis of MD, one should consider cysts and pseudocysts inside the umbilical cord, as well as intestinal duplication, mesenteric cyst, choledochal cyst, and urachal abnormalities, especially in the early second trimester of pregnancy[6,7]. Other relevant differential diagnoses include intestinal obstruction and ovarian cyst, particularly after the twentieth week of gestation[6].

Fetal MRI can be an important complementary exam to ultrasound in cases of omphalocele, as it allows for better visualization of its content[8]. In our literature review, we found a significant shortage of descriptions of MD evaluated through fetal MRI5. The normal fetal gastrointestinal tract typically exhibits the following findings: after 24 weeks of gestation, the colon and rectum appear hyperintense on T1-weighted images and hypointense or with intermediate signal intensity on T2-weighted images (due to the presence of meconium); the jejunum is hyperintense on T2 and hypointense on T1, mainly due to its liquid content, and is usually visualized through T2-weighted images in coronal planes. Normally, duplicated cysts show a hypersignal, similar to fluid, on T2-weighted images and a hyposignal on T1 when visualized through fetal MRI. Meconium cyst, segmental ileal dilatation, and colonic pouch exhibit a hypersignal (meconium-like) on T1-weighted images associated with a

hyposignal on T25. In our case, we observed an unusual cystic image within the omphalocele, which appeared hyperintense on both T1 and T2, indicative of MD.

Despite the description that fetuses with prenatally diagnosed MD are more likely to be born prematurely and have low birth weight[9], our patient did not exhibit such findings. However, it is important to be vigilant for these potential manifestations in order to plan the delivery more effectively.

Most children born with MD are asymptomatic[10], as observed in our patient. However, some newborns may experience lower gastrointestinal bleeding, intestinal obstruction, and local inflammation shortly after birth, which can lead to perforation and, consequently, an increased risk of morbidity and mortality[3].

Hemorrhage in patients with MD can be associated with the presence of ectopic gastric tissue, which is reported in 24.2-71% of symptomatic individuals, who are typically young. The main complication reported, not only in pediatric patients but also in symptomatic adults, is intestinal obstruction (in 35.6-46.7% of cases); hemorrhage and inflammation are also common complications in patients of both age groups[3].

This report highlights the importance of accurately determining the content of the omphalocele, as it may be able to identify anomalies such as MD and modify gestational management. For example, in cases of MD, patients may benefit from birth planning, not only due to the risk of prematurity and low birth weight but also due to the risk of hemorrhage. Additionally, prenatal diagnosis aids in the development of the surgical plan to be carried out after birth. In cases of MD, it is also important that umbilical cord clamping after birth be done away from the base to avoid possible iatrogenic ileal atresia[5]. Supplementary examinations, such as fetal MRI, can also complement the ultrasound assessment and, consequently, help define the correct diagnosis, which implies proper management, follow-up, and treatment[2].

CONCLUSION

In summary, cases of omphalocele diagnosed in the prenatal period should be carefully investigated, as they often do not consist of isolated anomalies. The awareness of additional malformations, such as MD, identified through ultrasound, with or without supplementary examinations like MRI, is of great importance because it directly affects gestational management, birth planning, surgical approach, and postnatal care.

The subsequent steps after diagnosis are crucial to reduce infant morbidity and mortality rates, thereby modifying the prognosis.

ACKNOWLEDGEMENTS

We would like to thank the patient and his family, as well as other colleagues from the Fetal Medicine Service at Hospital Materno Infantil Presidente Vargas (HMIPV), for their contribution to the development of the study.

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CASE REPORT

BRAZILIAN JOURNAL OF ULTRASONOGRAPHY 2023; 35: 29-31 - DOI - 10.29327/2320983.31.35-6

EXTREMELY LONG UMBILICAL CORD: A CASE STUDY ON ITS SIGNIFICANCE IN DIAGNOSIS AND PRENATAL CARE

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ABSTRACT

Access to the placenta is represented by the umbilical cord. Therefore, complications linked to the umbilical cord are directly conditioned to the nutritional part of the fetus. Problems relating to a long umbilical cord include risk of preterm labor, placental abruption, and vascular occlusion from thrombosis. Closer to delivery, it also predisposes to cord circulation in the cervical region, prolapse and true knot. Umbilical cord abnormalities can be detected by ultrasound, with important diagnostic and prognostic implications in terms of perinatal morbidity and mortality.

In this study we present the case report of a pregnant woman with previous ultrasound showing delayed intrauterine regrowth. At delivery, newborn with extremely long umbilical cord, six circular cords in the cervical region.

This abnormality in the umbilical cord can have numerous clinical repercussions for fetal well-being, showing the importance of knowing the anatomy and normal development of the umbilical cord for accurate prenatal diagnosis and evaluation.

KEYWORDS: LONG UMBILICAL CORD, ULTRASOUND, UMBILICAL CORD ABNORMALITIES

INTRODUCTION

Complications related to the umbilical cord are directly linked to the fetus's nutrition because the umbilical cord serves as the fetus's access channel to the placenta. These complications can also be associated with fetal malformations, chromosomal problems, and complications of the pregnancy itself. The length of the umbilical cord can be observed through ultrasound starting from the 7th week of gestation. Until the 30th week, its growth mirrors that of the fetus, but after that, it grows more rapidly, reaching a length of 30 to 70 centimeters. The umbilical cord contains two arteries and one vein, which can be identified via ultrasound starting from the 10th week, using a technique called color Doppler ultrasound.

The development of the umbilical cord is related to the formation of the fetus's anterior abdominal wall, and its growth is influenced by the fetus's movements and the amount of amniotic fluid present. Long umbilical cords may be associated with conditions such as maternal diabetes and Rh isoimmunization, increasing the risk of premature birth, placental abruption, and vascular obstruction due to thrombosis. Near the time of delivery, situations like nuchal cord can occur (when the cord completely encircles the fetus's neck, with an incidence of 25%), and in some cases, multiple wraps can occur, especially due to fetal movements.

Abnormalities in the umbilical cord can be identified through ultrasound, which has significant implications for the diagnosis and prognosis regarding perinatal health, meaning the period surrounding childbirth. Therefore, it is essential to understand the normal anatomy and development of the umbilical cord to make an accurate diagnosis during prenatal care. The aim of this study is to describe a case of pregnancy in which a long umbilical cord is causing fetal growth restriction.

CASE REPORT

S.L.N.S.M, 22 years old, GIII, PI, CI, AI. The LMP on 03/05/21, EDD on 12/10/21, maternal weight gain of 15 kg, using 25 mcg/day of levothyroxine. In the 1st trimester, experienced bleeding and placental detachment, prescribed progesterone 200 mg until 12 weeks with remission. In the 2nd trimester, without complications, normal serologies. Starting from November 3rd, ultrasound diagnosed Intrauterine Growth Restriction (IUGR), fetus weighing 2025g with no apparent causes and normal blood flow, placenta at maturity grade II. On November 16th, weight was 2,166g, and on November 23rd, weight was 2,434g, showing a slight increase compared to the previous weight. (see Figure 1).

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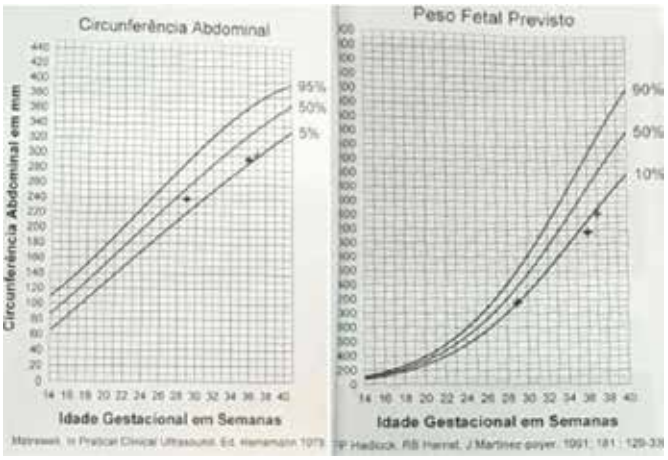

Figure 1: Image with the evolution curve of fetal biometric data.

Complementary exams carried out

Obstetric ultrasound (23/11/21) revealed a fetus in a longitudinal position, breech presentation. Fetal heart rate 131 bpm. Fetal weight 2,434 grams, fetal height 47.3cm. Posterior topical insertion placenta. Thickness 39mm, heterogeneous texture and grade II maturity, with two arteries and a normally inserted vein. Normal amniotic fluid, amniotic fluid 14cm. Topical, single pregnancy, 37 weeks and 4 days. Weight below the 10th percentile. Maternal-fetal Doppler within normal limits. (figure 2)

Figure 2: Images from the ultrasound performed on 11/23/2021.

Cesarean section was performed 11/28, breech presentation, female fetus, alive and with thick meconium amniotic fluid, APGAR 8/9, six cervical circulars of the umbilical cord measuring 155cm (figure 3), extraction of normal placenta weighing 635g, fetus measuring 45cm, weight 2,380g.

Figure 3. Image of long umbilical cord: measuring 155cm.

DISCUSSION

Access to the placenta is represented by the umbilical cord. Therefore, complications linked to the umbilical cord are directly dependent on the nutritional part of the fetus. Cord abnormalities may be associated with fetal malformations, chromosomal aberrations and complications related to pregnancy itself[1-5].

In embryogenesis, the umbilical cord presents its development related to the formation of the anterior abdominal wall. As the amnion expands, the embryo is covered with amniotic epithelium. From this connection, it develops blood vessels and fuses with the omphalomesenteric duct between 7-8 weeks of menstrual delay, giving rise to the umbilical cord. The blood vessels of the allantois give rise to the vessels of the umbilical cord; thus, the umbilical arteries are in continuity with the iliac arteries inside the pelvis [1,2,5,6].

The umbilical cord is first seen between 7-8 weeks of missed menstruation. At this gestational age, the length of the umbilical cord is approximately equal to the crown-rump length. It continues to be the same length as the fetus throughout pregnancy until the 30th week, after this age, the cord represents faster growth. Its diameter is normally less than 2cm. As the umbilical cord grows, it develops up to 40 coils, which are due to helical muscle layers within the umbilical arteries. This winding helps the cord resist compression from blood vessels. Its development in length and spiraling is related to fetal movement and the amount of amniotic fluid, through the tension force applied to this cord. However, there must be an adequate amount of fluid and

fetal activity to provide normal length and expiration [5-7].

Since the growth of the cord is related to fetal movement and the amount of amniotic fluid. Fetuses with movement restrictions due to anomalies of the central nervous system or skeletal dysplasias and oligoamnios, or fetuses that present an embryonic defect, causing defects in the limbs and body wall, causing a short, smaller than average umbilical cord. These conditions are related to prematurity, growth restriction and fetal distress, Down syndrome [1-3].

Long cords are associated with maternal diabetes mellitus, Rh isoimmunization, hydrops, diffuse hematoma, and the correlation with increased fetal movements and long cord is unclear. In addition, problems related to long cords include risk of premature labor, placental abruption and vascular occlusion due to thrombosis. Habek et al report an umbilical cord measuring 190cm and six cervical circular cords [7]. Closer to birth, it also predisposes to cervical circular cord, prolapse and true knot. The circular cord is a loop of cord, completely surrounding the neck with an incidence of 25%, increased risk of fetal death when there is more than one circular, especially in relation to movements [1,4,5].

The umbilical cord contains two arteries and one vein. The umbilical vein carries oxygenated blood from the placenta to the fetus, where it comes into contact with the left portal vein of the liver. The umbilical arteries are continuous with the internal iliac arteries, and carry deoxygenated blood from the fetus to the placenta. The umbilical arteries are confirmed by ultrasound, visualizing two vessels lateral to the fetal bladder. These vessels in the cord are surrounded by Wharton's jelly, a gelatinous connective tissue that protects the umbilical vessels against compression. The vascular abnormalities found are the most common single umbilical artery; and the presence of more than one umbilical vein and more than two umbilical arteries are rare abnormalities [6,7].

The cord can have different implantation locations in the placental disc, in the central portion of the placenta, in the eccentric insertion, in the marginal insertion and in the velamentous insertion. The first two do not show any difference in pregnancy. However, veiled insertion can cause problems during pregnancy, such as growth restriction, prematurity, fetal hemorrhages, fetal death, among others [3,4,6].

The ultrasound identifies the umbilical cord from the 8th week onwards, and as the pregnancy progresses its changes become more apparent, in which length, thickness, number of vessels, place of implantation in the placenta and the presence of cysts and neoplasms can be assessed[2,3].

The normal length varies between 30-70cm, in long cords > 70cm, true, circular knots are investigated in the cord in any part of the body, being more frequent in the cervical region. When its thickness is greater than 2cm, maternal pathologies are investigated, gestational diabetes, alloimmunization, among others.

When the umbilical cord is short, there is a loss of the coin stacking signal, formed by the agglomeration of the normal umbilical cord [3,7].

Given the abnormalities that arise from the umbilical cord, and the numerous clinical repercussions that can have on fetal well-being, the importance of knowing the anatomy and normal development of the umbilical cord for accurate diagnosis and prenatal evaluation is demonstrated[3].

CONCLUSION

The present case is a case of an extremely long umbilical cord, with six circular cords, which resulted in intrauterine growth restriction and low birth weight.

The need for early diagnosis and the importance of knowing the anatomy and normal development of the umbilical cord for more accurate diagnosis and prenatal evaluation are evident. This diagnostic integration promotes more detailed and therefore safer prenatal care for the maternal-fetal binomial, and for the clinician to develop an appropriate therapeutic strategy.

In view of umbilical cord anomalies and the clinical repercussions that can compromise fetal well-being, the importance of knowing the anatomy and normal development of the umbilical cord for accurate diagnosis and prenatal evaluation and appropriate therapy is demonstrated.

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FIBROMATOSIS COLLI: A BENIGN CERVICAL PSEUDOTUMOR AND ITS ASSOCIATION WITH ANOMALIES IN THE FIRST RIB – CASE REPORT

RAFAELA CARDOSO GIL PIMENTEL, AUGUSTO CESAR GARCIA SAAB BENEDETI, FERNANDO MARUM MAUAD, YUJI MATSUI, FRANCISCO MAUAD FILHO

ABSTRACT

Fibromatosis colli (FC) is a rare benign disease in which there is proliferation of the fibrous tissue of the sternocleidomastoid muscle (SCM), with complications during childbirth or genetic factors in neonates. The objective of the present study is to show a case of CF in an adult after a contact injury between a rudimentary rib and the SCM muscle. Ultrasound (US) as a complement to the clinic is the method of choice for diagnosis and to prevent any unnecessary invasive intervention, with conservative treatment with anti-inflammatories, physiotherapy, chiropractic and, as a last resort, surgery, if symptoms worsen during follow-up or occurrence of complications.

KEYWORDS: CERVICAL, FIBROMATOSIS, ACCESSORY RIB, ULTRASOUND, STERNOCLEIDOMASTOID

INTRODUCTION

Fibromatosis colli (FC), also known as congenital muscular torticollis (CMT), is a rare benign pseudotumor. Its tumor classification is somewhat misleading since it does not exhibit malignant characteristics but rather a proliferation of fibrous tissue in the SCM muscle, causing diffuse enlargement and other reactive changes. The causes of this growth are still under study, potentially resulting from repetitive muscle trauma, complicated childbirth, and genetic factors [1-2]. This case report aims to describe a case of fibromatosis colli in an adult caused by repetitive SCM injury due to a first rib deformity.

CASE REPORT

S.B, 25 years old, attended the service to undergo an ultrasound (US) of the cervical lymphatic chain due to suspected lymphadenopathy.. On physical examination the patient presented a palpable nodule in the right clavicular region, painful to touch and during movement, which began two weeks ago after prolonged air travel.

On ultrasound examination a thickening with a nodular pattern was noted in the distal insertion region of the right SCM, with flow capture on Doppler study, measuring 0.8cm – figures 1 and 2.

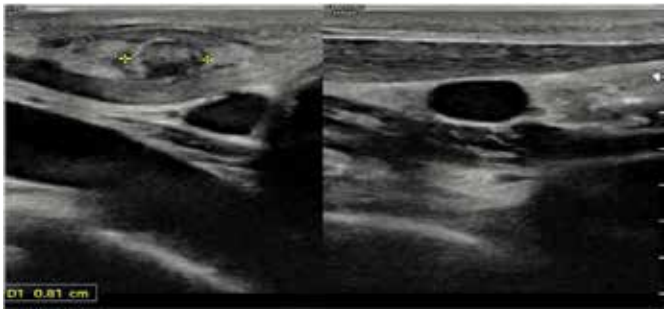

Figure 1: Right and left SCM muscle respectively.

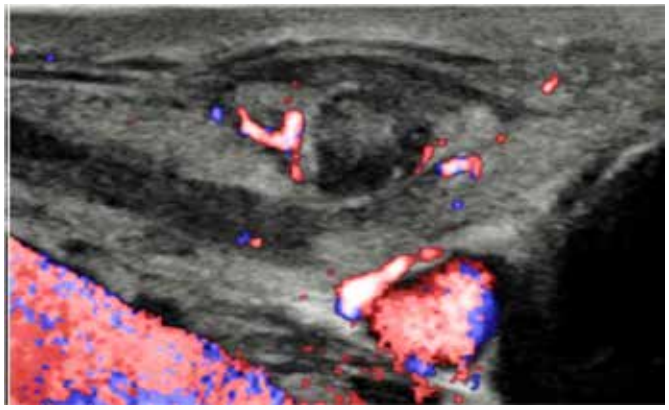

Figure 2: In the distal insertion of the right SCM, thickening in a nodular, heterogeneous pattern, with flow capture on Doppler study and increased echogenicity of the adjacent fat.

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Using the elastography technique of the lesion, a central solid consistency can be seen (figure 3)

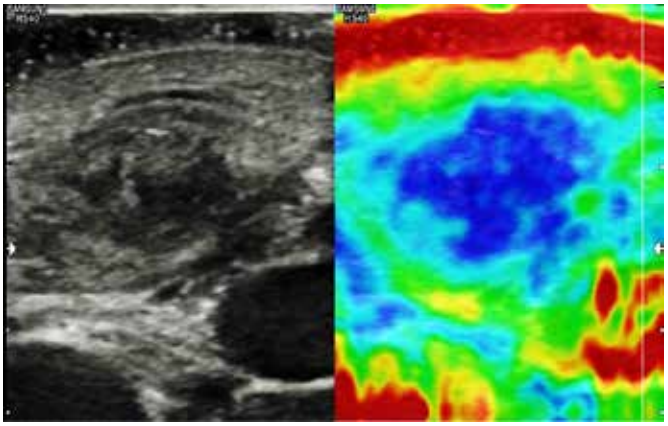

Figure 3: Elastography of the lesion, showing its solid consistency throughout its central area.

In addition to having cervical adenomegaly with a reactional appearance, bilaterally, but evident at levels IIa, measuring 3.4cm on the right side and 3.0cm on the left (figure 4). Remainder of exams within normal parameters.

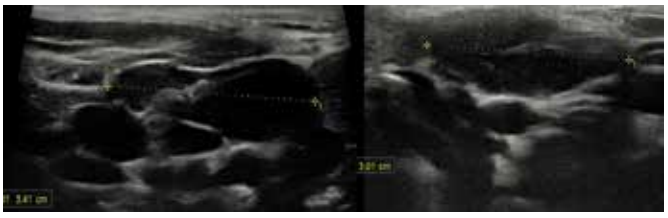

Figure 4: Enlarged lymph nodes at right and left level IIA respectively.

A chest X-ray was performed in both views after US, and a deformity in the first rib was observed (figure 5). Among the differential diagnoses, a rudimentary rib could be considered.

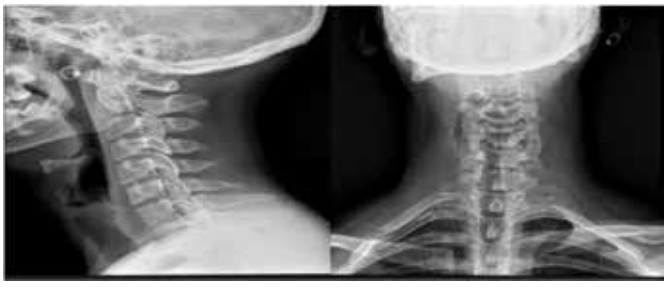

Figure 5: Chest X-ray in both views, showing shortening of the first rib

After the diagnosis, the patient improved with the use of NSAIDs (non-steroidal anti-inflammatory drugs), muscle relaxants, and chiropractic intervention, without the need for invasive procedures.

DISCUSSÃO

A FC, também conhecida como tumor esternocleidomastoídeo da infância ou torcicolo muscular congênito, tem sua definição equivocada de malignidade. Entretanto, massas cervicais tem origens de diversas condições patológicas e de desenvolvimento, sendo importante distinguir das mais graves [1].

Apesar dessa patologia ser mais comuns em recém nascidos, em qualquer porção do ECM, mais raramente bilateralmente, o caso supracitado foi em um jovem após um contato contínuo e direto de sua primeira costela com o músculo em questão durante um longo período [1-2].

Costelas rudimentares ou hipoplásicas refere-se a uma variação anatômica em que uma ou mais costelas, geralmente sendo as primeiras, apresentam um desenvolvimento reduzido, atrofiado ou incompleto em comparação com as costelas normais. Muitas vezes, sua presença costuma ser assintomática e pode ser detectada incidentalmente através de exames de imagens. Em alguns casos, essa variação pode estar associada a condições genéticas ou anomalias no desenvolvimento embrionário. Um estudo foi feito, onde foram encontradas em 79 homens (0.2%) do estudo. Vale lembrar que não se deve confundir uma primeira costela rudimentar com uma costela cervical [3-4].

O diagnóstico se baseia tanto na clínica do paciente, como também na ultrassonografia com a presença de sinais típicos: músculo ECM espessado e fusiforme com manutenção do padrão fibrilar das fibras musculares, às vezes pode ser circundado por uma borda hipoecóica focal, representando a compressão do músculo afetado, por isso a importância de fazer a comparação com o lado não afetado. A US pode descartar outros diagnósticos diferenciais como linfadenopatias ou higroma císticos em recém nascidos [5-7].

A punção aspirativa por agulha fina (PAAF) ou a histopatologia podem ser usadas em adultos quando o diagnóstico não é claro se tal massa possa ser maligna ou benigna, pois mostram uma atrofia das fibras musculares esqueléticas e células musculares gigantes em regeneração8-10. Dependendo dos resultados do diagnóstico, em adultos a FC costuma ser conservador na ausência de sintomas graves ou complicações. O tratamento pode envolver observação regular para monitorar o crescimento da massa, fisioterapia, AINES, quiropraxia ou cirurgia nos últimos casos se necessário [5].

CONCLUSÃO

A FI é uma causa rara de edema cervical em recém nascidos, e ainda mais rara em adultos. A US é o método diagnóstico de escolha, evitando assim a necessidade de outros procedimentos invasivos e intervenções terapêuticas. Na presença de características ultrassonográficas típicas, a PAAF não é necessária. Sendo importante uma diferenciação na malignidade para descartar outros diagnósticos diferenciais na dúvida com o US. Por ser uma condição autolimitada, sendo necessário apenas sintomáticos, fisioterapia e observação.

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CASE REPORT

A CHALLENGING PRENATAL CASE: UNRAVELING THE RARE AND LETHAL SACROCOCCYGEAL TERATOMA

THIAGO LARA ROCHA, SAMUEL CAVALCANTE REIS, VALÉRIA MARÇAL VIEIRA, MARCELLO BRAGA VIGGIANO

ABSTRACT

This is a case report referring to prenatal care and follow-up in the third trimester of pregnancy, which was referred to confirm the initial diagnostic hypothesis of Myelomeningocele. After morphological ultrasound an even rarer and lethal pathology was diagnosed: Sacrococcygeal Teratoma (SCT). Sacrococcygeal teratoma is a germ cell tumor, and such fetal neoplasia is associated with complications such as prematurity, hydrops and fetal death, and may also lead to maternal complications such as maternal mirror syndrome, where the mother presents the same fetal complications such as generalized edema and pleural effusion.

The case in question evolved with a favorable outcome for the pregnant woman and an unfavorable one for the fetus, culminating in death, even with the neonatal ICU apparatus, thus reinforcing the severity of the malformation.

KEYWORDS: SACROCOCCYGEAL TERATOMA, GERM CELL NEOPLASM, NEURAL TUBE DEFECTS, HIGH RISK PRENATAL CARE, FETAL MALFORMATION.

INTRODUCTION

Sacrococcygeal teratoma is a type of tumor that originates from germ cells and is most common in childhood, representing approximately 40% of all tumors of this type. It can also be identified during the fetal stage, with an approximate incidence of 1 in every 27,000 cases.

In this specific case, the diagnosis was made late through an ultrasound performed at a specialized and high-risk service. Obstetric ultrasound was used with morphological parameters, even though the ideal gestational age for this type of evaluation had already passed.

Despite being a relatively common neoplasm, its diagnosis is complex, and when detected, it allows for a more active approach to prevent unfavorable outcomes, as occurred in this case. Therefore, the aim of this study is to present ultrasound images suggesting the presence of this tumor, with the goal of early diagnosis of this condition. Additionally, it highlights possible complications associated with this condition, both during the prenatal and postnatal periods.

CASE REPORT

A 28-year-old patient, in her third pregnancy and with two previous cesarean sections, was referred to her first high-risk prenatal consultation at the Hospital da Mulher de Goiânia (HEMU). The initial suspicion was a possible myelomeningocele, due to changes detected in a previous ultrasound. In this initial assessment, the patient did not present specific complaints, having sought assistance only due to changes in the previous ultrasound examination.

The previous ultrasound examination showed a mild increase in the amniotic fluid index (AFI) and the presence of a hypoechoic oval-shaped image in the lumbar region, which could correspond to a myelomeningocele. Laboratory tests and the patient's physical examination did not show significant abnormalities. Therefore, the patient was advised to undergo an obstetric ultrasound with morphological parameter evaluation, although it was outside the ideal timeframe for the second-trimester morphological examination (typically between 20 and 24 weeks). The objective was to confirm the diagnosis and determine the appropriate course of action for prenatal care and delivery.

The second ultrasound was performed seven days after the first consultation and showed an amniotic fluid index (AFI) with values above the normal range, confirming the diagnosis of polyhydramnios. Additionally, an image consistent with a sacrococcygeal teratoma measuring 11.5 x 9.5 cm was identified, changing the diagnostic suspicion to this rare condition with a worse prognosis (Figure 1).

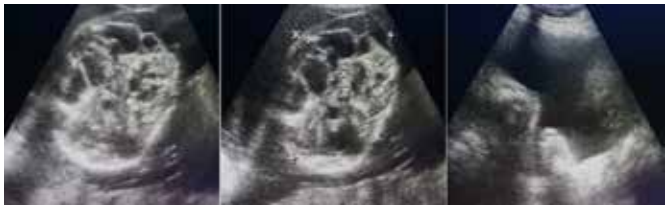

Figure 1. Ultrasound shows a mixed tumor image in the fetal sacral region and polyhydramnios.

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Six days after the first ultrasound, during a new fetal assessment, significant fetal anemia was observed due to the large accumulation of fetal fluids caused by the tumor. At this time, the middle cerebral artery peak systolic velocity was measured at 60.3cm/s (> 1.5MoM), indicating moderate anemia. Given the gestational age of 32 weeks and 3 days, the patient was admitted for lung maturation and, subsequently, the pregnancy was planned to be resolved by cesarean section.

The patient remained in the hospital ward for another three days to stabilize before the cesarean section was performed. The indication for cesarean section was related to fetal malformation, moderate anemia and worsening Doppler flowmetry results.

During delivery, a single female fetus was extracted, which presented an APGAR score of 3 in the first minute and 6 after 5 minutes. The fetus presented a large mass in the sacral region, without ulcerations, with several nodulations and hardness to the touch, in addition to a lower ear implantation.

The newborn (NB), despite not crying or breathing immediately after birth, received resuscitation, including positive pressure ventilation, and was transferred to a stabilization room. The pediatric team described a 15-20cm lesion in the sacral region, without ulcerations, with several hardened nodules and significant collateral circulation (figure 2). Additionally, a low auricular implantation was observed.

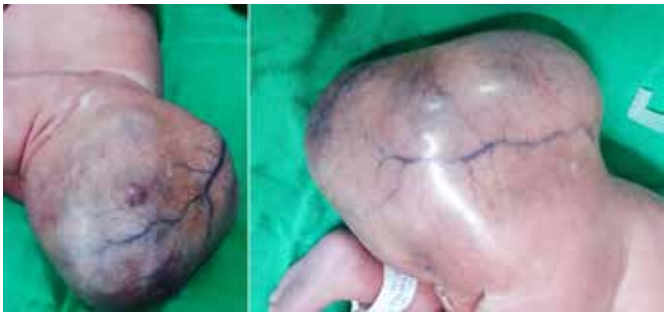

Figure 2. Neonatal image shows tumor mass in the sacral region with exuberant collateral circulation.

The NB remained in the stabilization room before being transferred to the neonatal ICU. However, after 28 hours of life, the newborn presented a significant worsening in herr overall condition, including hemodynamic instability, cold extremities and slow peripheral perfusion, indicating shock (figure 3). Non-invasive ventilation, orotracheal intubation, central venous access puncture and administration of adrenaline were necessary to stabilize her. Measures were adopted for hemodynamic compensation, such as expansion with saline solution and transfusion of packed red blood cells.

Figure 3. Neonatal image 28 hours after birth shows a significant increase in the tumor mass.

After these interventions, the NB underwent a chest X-ray, which revealed a bilateral pneumothorax. Chest drainage was performed, but the NB did not show hemodynamic or respiratory improvement. Unfortunately, the NB went into cardiorespiratory arrest, with unsuccessful cardiopulmonary resuscitation attempts, leading to the patient's death.

This case report highlights the complexity of diagnosing and managing sacrococcygeal teratoma, as well as the serious complications associated with this condition, both prenatally and postnatally.

DISCUSSION

Sacrococcygeal tumor is the most common germ cell tumor of childhood. In the pediatric population they account for 40% of all germ cell tumors, rarely in adulthood. It is the most frequently recognized fetal neoplasm, with an estimated incidence of approximately 1 in 27,000, and more common in females [1].

Teratomas are generally composed of cells representing all three germ cell layers, and present in utero as a mass extending outward from the caudal end of the fetus, or as a childhood tumor that may be asymptomatic, or show signs of obstruction of the rectum or bladder [1,2].

The Altman classification describes the extent, whether the tumor is external and/or internal. Type I tumors are mainly external, while type IV lesions are entirely internal. Type I and II tumors are the most evident on prenatal ultrasound and clinical examination. Type IV tumors are generally found later, in early childhood, compared to tumors with an external component (types I-III) [1]. Compared with the case in question, it is clear that the corresponding type is type I.

Prenatal diagnosis typically occurs during the second trimester, which underscores the difficulty in diagnosing the case in question, as it was identified at the beginning of the third trimester. Most SCTs diagnosed prenatally are solid or mixed, with both cystic and solid components; calcifications are often present, consistent with the reported case, which corresponds to a mixed tumor with solid and cystic components and interspersed calcifications[2,3].

Associated structural abnormalities may include bladder

outlet obstruction and hydronephrosis, rectal stenosis or atresia and cardiomegaly secondary to vascular shunt and high-output heart failure, characteristics that were not observed in the case analyzed [3].

Fetal magnetic resonance imaging (MRI) is recommended when available. Compared to ultrasound, MRI more accurately characterizes the intrapelvic and abdominal extent of the tumor and the compression of adjacent organs. This information can assist in prenatal counseling and preoperative planning for surgical resection. Although these options were considered, due to the rapid progression of the condition, it was not possible to obtain a more precise diagnosis through MRI, and the primary course of action was based on the ultrasound diagnosis.

The most important differential diagnosis of an exophytic cystic sacral mass in the fetus is a distal neural tube defect (such as myelomeningocele or myelocystocele). This fact was confirmed when analyzing the reason for referring the case to the referral hospital, and the initial suspicion of the investigation was indeed this highly relevant differential diagnosis. Therefore, it is essential to keep in mind that both pathologies should be considered when encountering such abnormalities[4]. It is important to note that SCTs always have a portion near the coccyx; they can extend to the sacrum, but the mass effect is usually pre-sacral and not posterior, as would be the case with a neural tube defect[4].

Serial ultrasound evaluation of the fetus, placenta, and tumor throughout pregnancy is an essential component of the overall treatment plan. The primary goal is to identify fetuses at increased risk of fetal death due to hydrops resulting from high-output cardiac failure related to vascularization and tumor size and to intervene as appropriate. This is another crucial point highlighted in the article by Egler et al [1], which also inherently connects with the case reported in this article. Through fetal middle cerebral artery Doppler, fetal anemia can be identified, which was consistent with cardiac failure and hydrops that led to fetal death in the outcome of the case in question. Therefore, it emphasizes the importance of early identification and proper management of this fetal pathology.

The size of the tumor should be measured in each ultrasound examination, and the solid portions of the tumor should be assessed with Doppler ultrasound to evaluate vascular flow 1,5. Large lesions (>10cm), such as the one in this case, which was around 15-20cm, especially vascular ones, are associated with a high rate of perinatal mortality, consistent with the unfavorable outcome in this case. Relatively cystic lesions with absent or mild vascularity tend to exhibit slow growth and a favorable outcome, even when large (>10 cm) 1,5.

The amniotic fluid volume and placental thickness should also be assessed because polyhydramnios and placental thickening are markers of hydrops, and oligohydramnios can result from bladder obstruction due to SCT [4]. In the analyzed case, there were significant polyhydramnios and signs of hydrops.

The frequency of ultrasound examinations depends on the composition of the tumor (i.e., cystic or solid), its vascularity, and any associated findings. Follow-up imaging can be as often as twice a week for high-risk tumors or as infrequently as every two weeks for small or predominantly cystic lesions [4]. This point could not be analyzed in the case due to its severity, which required a more rapid and decisive resolution.

Fetal echocardiography is recommended in fetuses with predominantly solid and/or vascular tumors. Echocardiography is used to identify a high-output cardiac state that precedes the onset of hydrops. A fetal cardiac profile consisting of the evaluation of the cardiothoracic ratio, cardiac dimension scores, combined ventricular output, and valvular regurgitation can be used to identify fetuses with a poor prognosis [4].

Therefore, it can be observed that several factors described in the specialized literature were consistent with what was found in the present case. Even though some important points could not be applied, it became evident that early diagnosis is essential for a favorable outcome and increased survival of fetuses with such malformation.

CONCLUSION

This case report highlights the severity of this fetal malformation, which should be diagnosed early, aiming for individualized prenatal care with the support of a tertiary hospital due to its potential lethality. It is also essential to provide appropriate assistance to the pregnant woman, offering counseling and medical interventions to minimize both her physical and mental distress.

The fetal tumor in this reported case could be classified as type I or II, given its large volume in the sacral region, likely greater than 10cm, and its highly vascular nature, which contributes to increased severity and higher perinatal mortality, as detailed in the outcome of this case.

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CASE REPORT

MALFORMATIONS AND INTRAUTERINE GROWTH RESTRICTION IN TWINNING: A CASE REPORT

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ABSTRACT

Twin pregnancies have higher fetal mortality and morbidity compared to singleton pregnancies, especially monochorionic pregnancies, as they are associated with a higher risk of malformations, fetal growth disorders and prematurity.

The objective of the present work is a case report on diamniotic monochorionic twins associated with a selective growth restriction due to a malformation of the digestive system. The definitive diagnosis of tracheoesophageal fistula is established with the direct visualization of the fistulous path through esophagoscopy and/or tracheoscopy, radiological visualization by imaging methods, surgeries and autopsies, demonstrating a diagnostic difficulty, since the ultrasound does not have specific signs, being discovered only during childbirth.

Therefore, the determination of chorionicity is fundamental for the adequate planning of prenatal and ultrasound follow-up, since it is directly related both to the increased risk of complications and to alterations exclusive to monochorionicity.

KEYWORDS: MALFORMATION; IUGR; TWINS; PREMATURITY; ULTRASOUND

INTRODUCTION

With continuous technological advances and improvements in the quality of healthcare, the diagnosis of multiple pregnancies has become more frequent, assuming a prominent role in the knowledge of human reproduction. This scenario is justified due to the challenges inherent to this type of pregnancy, which include a greater risk of prematurity, fetal deaths, growth restrictions, morphological changes, among other complexities.

The frequency of monozygotic pregnancies is presented as a constant rate of 4/1,000, with small variations in relation to other factors such as maternal age, and may correspond to 20-30% of all twin pregnancies. While dizygotic ones respond around 70-80%, all being dichorionic types[1]. Monozygotic pregnancies can result in monochorionic or dichorionic pregnancies, with a smaller proportion being dichorionic, with embryonic division occurring up to the fourth day. When division occurs after this period, it will always result in the formation of a single placenta, regardless of the number of amnions[1].

The differentiation between zygocity and chorionicity is extremely important in the evaluation of multiple pregnancies. Zygocity refers to the identification of the number of fertilized eggs that are present in a multiple pregnancy. There are two main categories of zygocity: monozygotic and dizygotic. Chorionicity refers to the number of chorionic membranes present in multiple pregnancies, and is relevant to assess the risk of obstetric complications. Chorionic membranes are structures that surround gestational sacs and contain developing babies. Chorionicity can be classified into two main types: monochorionic and dichorionic [1].

Ultrasonography plays a crucial role in determining chorionicity in multiple pregnancies. Using ultrasound, it is possible to visualize and analyze the number of gestational sacs, chorionic membranes and the position of babies in the uterus. This information is essential to properly monitor the pregnancy and provide an individualized obstetric care plan, ensuring the health and safety of both mother and babies throughout pregnancy [1-2].

Therefore, this work aims to report a case of a twin with congenital malformation associated with growth restriction, evaluating the long-term prognosis in twins affected or not by selective growth restriction and examining the clinical and fetal development results.

CASE REPORT

Patient F.B.F.M.S, 33 years old, Caucasian, primigravida, at 12 weeks and 3 days of gestation in her first ultrasound (US) showed a monochorionic diamniotic twin pregnancy, with twin 1 having a crown-rump length (CRL) of 71.1mm and twin 2 measuring 63.0mm (Figure 1). Both twins had the same nuchal translucency (NT) measurement of 1.8mm. The pregnancy was classified as monochorionic and diamniotic due to the presence of the T-sign of the membranes on the chorionic plate (Figure 2).

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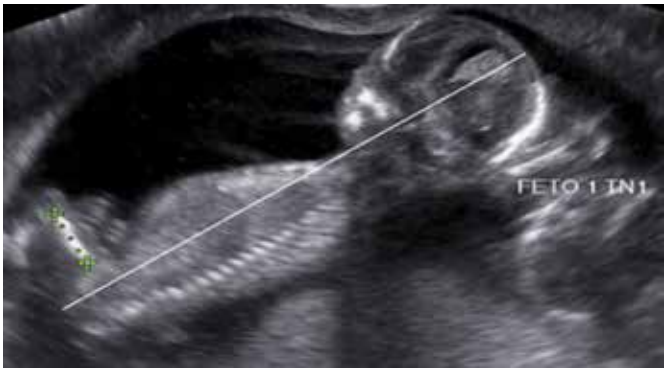

Figure 1: Crown-Rump Length of Twin 1

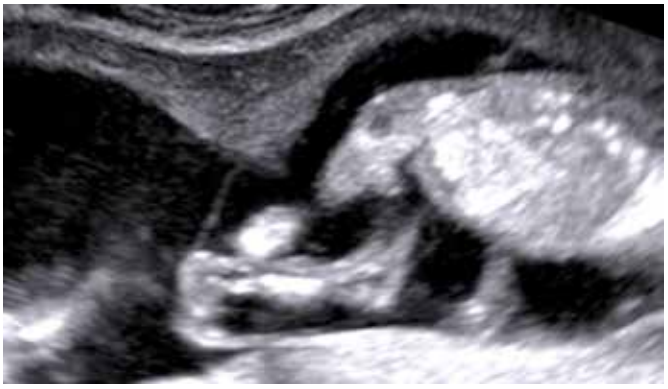

Figure 2: T-sign in Monochorionic Diamniotic Pregnancy

A Doppler ultrasound of the uterine arteries was also performed, which showed a mean PI of 1.77 (p62), indicating an increased risk stratification for preeclampsia and growth restriction, especially considering it was a twin pregnancy. As a result, a 150mg dose of aspirin was initiated. In the second-trimester morphological ultrasound, cardiac screening was found to be within normal limits for both fetuses; however, a single umbilical artery was identified in fetus [2].

Table 1 illustrates the longitudinal follow-up of fetal growth through biometric data and amniotic fluid measurements. It shows evidence of growth restriction in fetus [2].

Feto 1	Consulta 1	Consulta 2	Consulta 3	Consulta 4	Consulta 5
CC	24,2	29,1	28	29,5	28,1
CA	21,5	22,3	23,8	27	28,3
CF	4,3	4,7	5,8	6	5,7
MB	3,9	4,6	4,1	5	5
PFE	820	1034	1222	1607	1552

Feto 2	Consulta 1	Consulta 2	Consulta 3	Consulta 4	Consulta 5
CC	22	23,4	24,9	27,3	27,8
CA	19	20	21,1	23	23,8
CF	4	4,3	5,4	5,2	5,2
MB	3	3	3,1	3	3
PFE	495	786	958	1191	1270

Table 1: Relationship of biometric parameters between fetuses, demonstrating a restriction in fetus 2, but with amniotic fluid with values within the normality parameter. HC – head circumference AC – abdominal circumference FL – femur length LP – largest pocket EFW- estimated fetal weight.

On 12/29/2017, at 32 weeks of gestation, the patient went into labor due to a ruptured pouch, presenting a 3cm dilated cervix, and both fetuses with cephalic and breech presentation, which is why a surgical approach was necessary.

After the cesarean section, it was found that fetus 1 weighed 1685g and fetus 2 weighed 1200g, confirming growth restriction and the presentation of malformations. (tracheoesophageal fistula and agenesis of the thumb on the right hand).

During neonatal assessment, fetus 1 had a hyaline membrane, a respiratory disorder in which the alveoli of the newborn's lungs do not remain open due to high surface tension resulting from insufficient production of surfactant due to prematurity, but was discharged after 20 days in good conditions. Fetus 2 underwent gastrostomy and correction of the tracheoesophageal fistula (TEF), fistula dehiscence, developed aspiration pneumonia, septicemia and died on the 39th day.

DISCUSSION

Ultrasonography in the first trimester has emerged as an effective method for early determination of chorionicity and amniocity. Performing ultrasound between the 11th and 14th week of pregnancy provides detailed images of the gestational sac, enabling the identification of distinctive characteristics between monochorionic and dichorionic pregnancies. In the case report, it was a diamniotic monochorionic pregnancy, the growth of the gestational sacs results in the obliteration of the extraembryonic space, allowing contact between them and forming a thin membrane. From the 10th week onwards, it fuses with the chorion forming a 90° T-shaped angle (T sign), with a higher frequency of complications such as fetal deaths, anomalies, growth restriction and prematurity. Monochorionics have a risk of fetal transfusion syndrome, polycythemia anemia sequence, reverse arterial perfusion syndrome and selective fetal growth restriction caused by vascular anastomoses present in the single placenta shared by the two fetuses [1-6].

In cases of selective growth restriction, the difference in weight between the fetuses is at least 25%, and one of the fetuses has an estimated weight below the 10th percentile for gestational age. This restriction can be classified as type [1], despite the difference in size between babies, the flow pattern in the umbilical cord is normal, despite being unique, with the chance of birth occurring earlier, as in this case. In addition to having a 30% rate of presenting other malformations, a change in genetic etiology is excluded as it is a monozygotic pregnancy and has the same genetic load [4,5].

An important finding in the case was the amount of amniotic fluid that was within normal limits in both fetuses, thus ruling out other differential diagnoses such as feto-fetal transfusion and vascular anastomoses (figure 3).

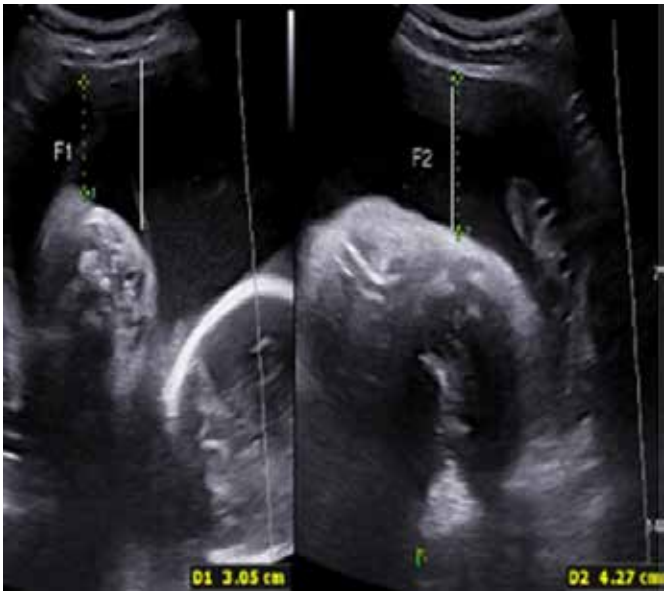

Figure 3. Illustrates the measurement of the largest pocket of amniotic fluid in fetuses 1 and 2.

At birth, it was seen that fetus 1, despite being prematurity, was discharged in good condition, but fetus 2, in addition to the restriction, had a tracheoesophageal fistula, a congenital malformation in which there is an abnormal communication between the trachea and the esophagus. TEF can occur alone or be associated with other congenital anomalies.

The exact causes of TEF are not yet fully understood, but it is believed that a combination of genetic and environmental factors may play a role in its development. Certain risk factors, such as smoking and exposure to toxic substances during pregnancy, have also been associated with a higher risk of TEF. It is confirmed by videofluoroscopy or bronchoscopy, allowing visualization of the fistula or abnormal contrast communication. Tracheoesophageal fistula treatment usually involves corrective surgery. However, the ideal time to perform surgery may vary depending on the severity of TEF and other associated medical conditions [7-10].

CONCLUSION

The study of fetal twinning and selective growth restriction reveals a complex picture in the field of obstetrics and perinatal medicine. This clinical case shows the importance of defining chorionicity, reinforcing its screening in the first trimester of pregnancy, and adequate follow-up throughout the pregnancy to evaluate fetal biometry and amniotic fluid.

Fetal twinning itself is a unique phenomenon that can present different challenges and medical considerations. However, when selective growth restriction is associated with a difficult-to-diagnose malformation in one of the fetuses, without other signs that can help with the diagnosis, the complexity increases and more specialized care is required.

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REVIEW ARTICLE

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IMAGING DIAGNOSIS OF SPLENIC TRAUMA: NARRATIVE REVIEW

THAMIRES DA SILVA SANTOS, LEONARDO DE SOUZA PIBER

ABSTRACT

INTRODUCTION: The spleen has an important role in the running of the human blood defense system, removing old red blood cells and holds a reserve of blood. However, this function can be compromised if occours an splenic trauma, that is the most commom kind of abdominal trauma, it can be classified as penetranting or blunt. The blunt splenic trauma can be caused, for example, by sporting injuries. Whilst the penetrating splenic trauma is caused by, for example, by gunshot wound. So, there is the gold standard diagnosis, the computed tomography, wich leaves room for non-operative management.

OBJECTIVE: Review, identify and describe the imaging characterization of splenic traumas.

METHODOLOGICAL PROCEDURES: This study can be characterized as a narrative review with emphasis on a collection of images. The databases were MEDLINE via PubMed, LILACS via BIREME, Scielo and Academic Google. The health descriptors (MeSH terms) in English are "splenic rupture", "spleen", "wounds and injuries" and "diagnostic imaging. Studies (clinic trials, pictorial essays, literature reviews, among others) that had images of diagnostic methods that were in accordance with the research objective and available online in full text published in the last 10 years, in english, spanish and portuguese.

RESULTS AND DISCUSSION: Splenic trauma presents as an imaging finding mainly the spleen laceration, seen as a hypodense line, which may or may not be irregular. Such a condition corresponds to the splenic hematoma and hemiperitoneum, as well as the fluid adjacent to the liver and in the paracolic, related to hemorrhage. Subcapsular and parenchymal hematoma can also be seen, as well as the presence of hypo-anechoic fluid collection in the subcapsular or perisplenic space. In addition, the computed tomography has a better performance when contrast is used.

CONCLUSION: The imaging diagnosis of splenic trauma should be done using preferably computed tomography, but focused assessment with sonography in trauma and ultrasonography also can be used with further confirmation by computed tomography.

KEYWORDS: SPLENIC TRAUMA. DIAGNOSTIC IMAGING. COMPUTED TOMOGRAPHY. SPLEEN.

INTRODUCTION

The spleen is one of the various organs that make up the human immune system, protecting the body against the risk of severe infectious diseases[1]. In general, the spleen removes old red blood cells and acts as a blood reservoir. However, its different regions, such as the splenic marginal zone, white pulp, and red pulp, have some essential functions[2]. The marginal zone contains B cells that produce many of the IgM and IgG antibodies, which react with blood-borne pathogens. The B follicular cells of the white pulp produce highly specific antibodies to combat these pathogens, an action known as adaptive immunity. Lastly, the red pulp of the spleen has intravascular macrophages that eliminate bacteria and other external materials. Therefore, the spleen can be considered the center of the immune system[2,3].

Due to its importance, splenic trauma puts the maintenance of human life at risk, as the injury or removal of this organ hinders the action of the immune system. Such trauma is the most common in relation to the abdomen, and can be perforating, caused for example by firearms, or blunt, caused mainly by car accidents, sports or even falls from great heights[2,4]. Its rupture can also occur after surgical procedures and infections2. Furthermore, it can affect all age groups and genders and has a high mortality rate of 7-18% when diagnosed late[5].

The advancement of radiology has greatly facilitated both the diagnosis and decision-making regarding the treatment of splenic trauma[2]. In addition to this, pain in the upper left quadrant of the abdomen, pain in the left shoulder, diffuse abdominal pain, and pain in the lower left chest are also indicative for diagnosis[2]. However, a splenic rupture can be masked by other injuries, and if it is a contained rupture, its initial symptoms may be minimal[2].

The Extended Focused Assessment with Sonography for Trauma (eFAST) and computed tomography (CT) are two imaging tests that can be used to diagnose such traumas[2]. The first reveals free abdominal fluids, such as blood or gastrointestinal contents, being very useful in hemodynamically unstable patients, as it is accessible, portable and non-invasive[2,5]. The latter is recommended for hemodynamically stable patients, with sensitivity and specificity of 96-100%[5]. CT is also considered the gold standard for blunt splenic trauma,

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having been fundamental for the development of Non-Operative Treatment (NOT) in this type of spleen trauma, even reducing the need for exploratory laparotomies. The correct use of these diagnostic methods reduces the chances of worsening the lesions, which could lead to extreme situations, such as splenectomy[2].

The American Association for the Surgery of Trauma (AAST) has formulated an organ injury scale, graded from I to V, with increasing severity, ranging from less than 10% involvement to complete rupture of the spleen, taking into account the extent of hematoma and the depth of laceration[2]. Based on this classification in conjunction with CT imaging, it is possible to determine the need for surgical intervention[2].

In cases of blunt splenic trauma, after correct classification of the injury, most cases I and II can be treated through NOM5. In grade III cases, the success percentage is 50%, reducing even further in IV and V cases6. Furthermore, for this method to be successful, there must be no peritonitis and associated injuries that require laparotomy, or hemodynamic instability. Therefore, angiography of the splenic artery may be used, which uses catheters guided with the aid of radiography with the aim of finding possible obstructed vessels, and angioembolization, which uses mechanical or chemical agents to reach obstructed vessels close to or distant from the bleeding point [2,4]. Such methods increase in need depending on the increase in the degree of the injury. The NOM failure rate is low, 4-15% of cases[7].

Regarding perforating splenic trauma, laparotomy is the gold standard of treatment[5]. More common in military areas or areas with endemic urban violence, it accounts for around 5-14% of cases of splenic trauma[8]. However, currently, there is an attempt to avoid splenectomy and invasive treatments as much as possible, resorting to non-operative treatments, as is the case of injuries caused by stabbing, in which if there is hemodynamic stability accompanied by the necessary diagnostic tests, the use of NOM can be indicated[1]. In hemodynamically unstable patients, perforating splenic traumas present fewer foci of blood loss than blunt traumas, which makes it possible to reduce the need for rapid removal of the spleen[1]. Thus, the use of laparotomy increases according to the degree of severity of the injury, with splenectomies occurring in cases of degree equal to or greater than III5.

OBJECTIVES

Review, identify and describe the imaging characteristics of splenic trauma.

METHODOLOGY

This is a narrative review with an emphasis on the collection of images. The databases were MEDLINE via PubMed, LILACS via BIREME, Scielo and Google Scholar. The health descriptors (MeSH term) in English are "splenic rupture", "spleen", "wounds and injuries" and "diagnostic imaging" in the following search strategy: (((spleen AND (wounds and injuries)) OR (splenic rupture) AND (diagnostic imaging)). Studies were included (clinical trials, pictorial essays, literature reviews, case reports, among others) that addressed the topic, that had images of diagnostic methods, that were in accordance with the objective of the research and that were available online in full text, published in the last 10 years in English, Spanish and Portuguese.

RESULTS AND DISCUSSION

Splenic trauma presents as imaging findings mainly the laceration of the spleen, which will be graded according to its severity, seen as a hypodense line, which may be irregular or not accordingly. This condition is accompanied by splenic hematoma and hemiperitoneum, as well as fluid adjacent to the liver and in paracolic gutters, related to hemorrhage. Subcapsular and parenchymal hematoma may also be observed. The identification of such findings allows the diagnosis to be made quickly. Furthermore, computed tomography performs better in terms of an assertive diagnosis when performed using contrast.

Computed tomography, ultrasound (US) and FAST 1-9 images display the features found in splenic trauma.

The images below show the portal venous displaying two examples of splenic lacerations. The left image depicts a small laceration (AAST classification level I), indicated by the arrow, appearing as a hypodense line extending from the spleen's surface with a small subcapsular hematoma (arrowhead). The right image shows a wide laceration with irregular margins (arrowhead), complicated by a ruptured subcapsular hematoma, with hemoperitoneum extending beyond the boundaries of the splenic capsule (arrows), classified as AAST level III4 - Figure 1.

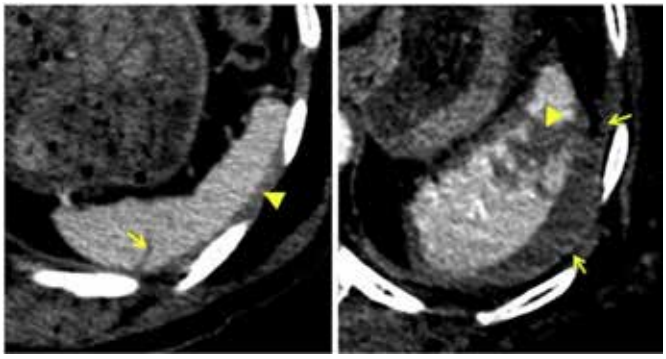

Figura 1 – Trauma esplênico contuso com laceração. Imagens de tomografia computadorizada[4].

In the figure below it is indicated in the portal venous phase: small (AACT level I) subcapsular hematoma (arrow) seen as a growing slightly hypodense collection with a smooth border in the portal venous phase – Figure 2. The following image displays the venous phase of the portal: intra-parenchymal hematoma (arrow) seen as a very globular hypodense "mass" in the spleen in the venous phase of the portal. It is <5 cm in size, a level II lesion on AACT4 – Figure 3.

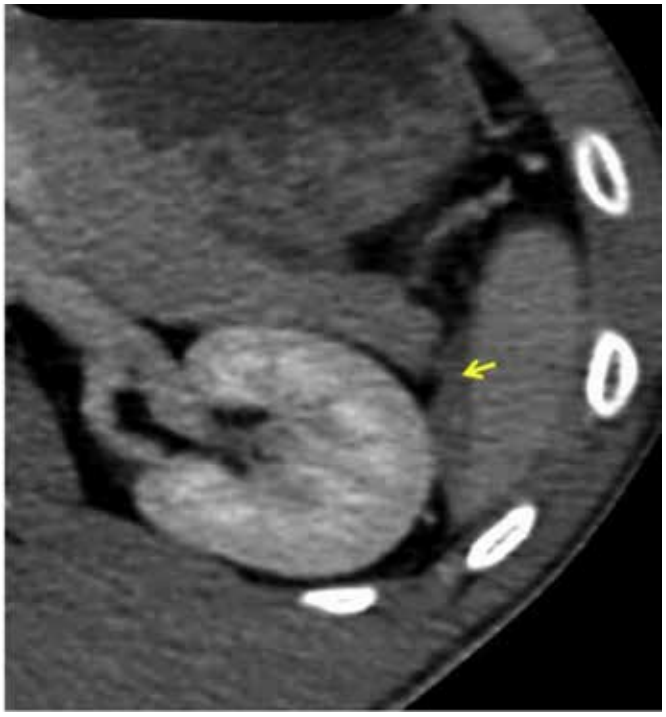

Figure 2 – Blunt splenic trauma: subcapsular hematoma. Computed tomography image[4].

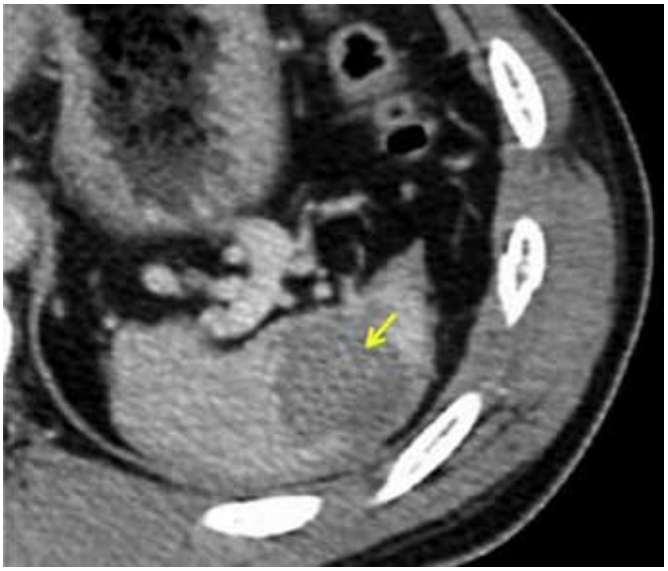

Figure 3 – Blunt splenic trauma: intra-parenchymal hematoma. Computed tomography image[4].

The CT image below of a 60-year-old woman after colonoscopy that was performed without contrast, showing a heterogeneous, hyperattenuating lesion in the spleen, 11.7 cm in maximum diameter, consistent with a large splenic hematoma, with only a small amount of normal splenic parenchyma being visible along the medial margin[9] – Figure 4.

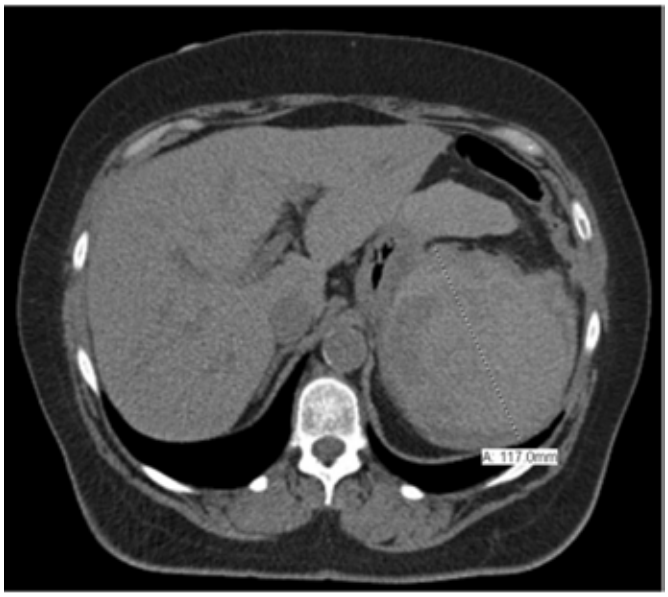

Figure 4 – Computed tomography of a 60-year-old woman after colonoscopy with splenic rupture performed without contrast[4].

The image below shows a FAST, which revealed heterogeneous echogenicity of the spleen consistent with splenic hemorrhage. Red arrowheads show hyperechoic areas and yellow arrowhead shows hypohechoic area. The curve of the diaphragm is indicated by D and the splenic trauma was confirmed by CT. The patient was treated with laparotomy[10] – Figure 5.

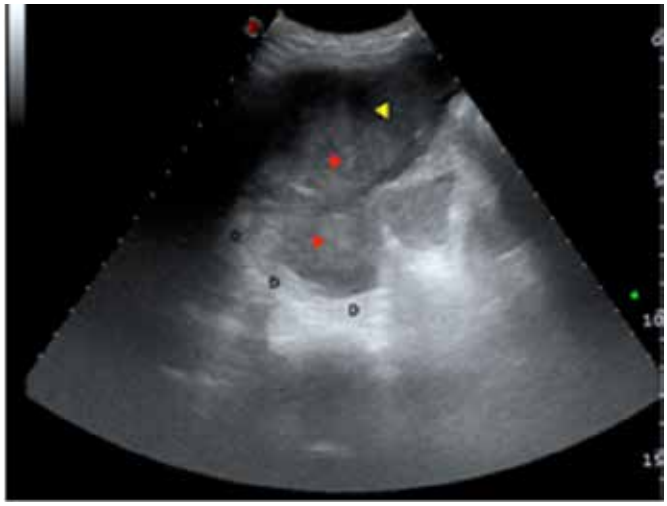

Figure 5 - FAST of a splenic trauma, later confirmed by computed tomography, and the patient treated with laparotomy[10].

The figure below shows an US and a CT of a 64-year-old man who is admitted to the emergency room close to syncope, with pain in his left flank and shoulder. Two days earlier he had undergone a colonoscopy due to a tortuous colon. Additionally, he reported significant left upper quad-

rant abdominal pain with guarding. During examinations it is possible to observe hemoperitoneum and splenic laceration. He developed hypovolemic shock and was treated surgically[11] – Figure 6.

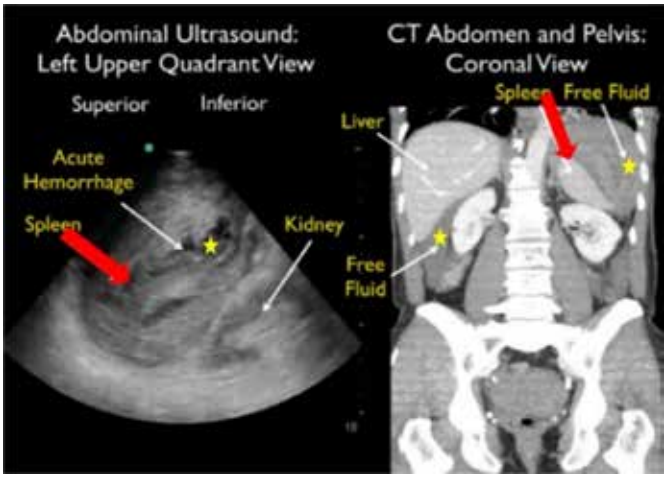

Figure 6 - Ultrasound image (left) and computed tomography (right) showing splenic trauma in a 64-year-old man who presented to the emergency room, developed hypovolemic shock, and was treated surgically[11].

The image below shows in A a longitudinal US of a spleen after blunt trauma, which demonstrates an intra-parenchymal laceration with a linear hypoechoic area (white arrow). In B, a post-contrast CT, which confirms the splenic injury (white arrow) and shows a laceration of the left hepatic lobe (black arrowhead) [12] – Figure 7.

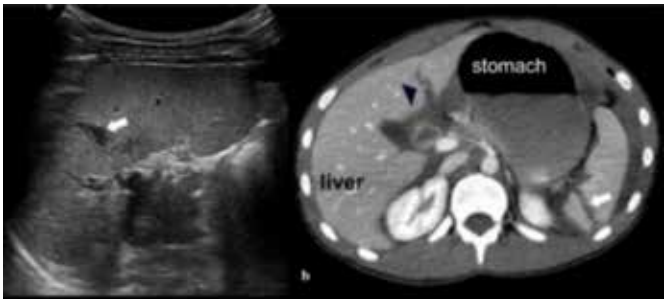

Figure 7 - Ultrasound and computed tomography images of splenic trauma in a child[12].

The figure below shows a splenic trauma. In ("A") there is a gray scale and in ("B") a transabdominal Doppler examination technique, which show a smooth heterogeneity of the lower pole of the spleen (thin white arrow) associated, in ("C"), with the presence of rupture of the splenic capsule (thin white arrow) and resulting perisplenic hematoma [12] – Figure 8.

Figure 8 – Gray scale ultrasound ("a"), transabdominal Doppler examination technique ("b") and presence of rupture of the splenic capsule ("c") [12].

Finally, the figure below shows a contrasted US. In "a" a longitudinal sonography after blunt trauma that shows an area of poorly defined echogenicity in the middle of the spleen and in "b", a hypoechoic lesion along the surface[12] – Figure 9.

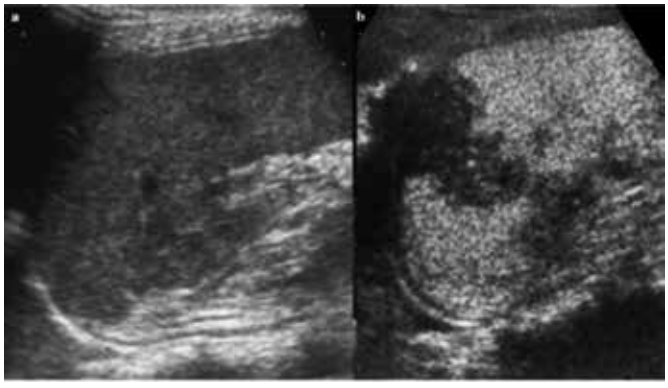

Figure 9 – Contrast ultrasound of splenic trauma. In "a" longitudinal sonography and in "b", a hypoechoic lesion [12].

CONCLUSION

The imaging diagnosis of splenic trauma should preferably be made using computed tomography, and FAST and US can also be used with subsequent confirmation by CT. Knowledge of the specific imaging findings of this abdominal trauma by the health professional provides greater agility in treating the patient, which may result in a better outcome of the case.

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