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CASE REPORT

SELSTOC: a new entity to bear in mind – case report

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Abstract

Introduction: SELSTOC (self-limiting sternal tumor of childhood) is a rapidly growing, self-limiting sternal/parasternal lesion of unknown etiology, first described in 2010. It has a median age of 16 months. It may present with local inflammation, but there is no systemic involvement. Mildly elevated inflammatory markers may be observed. Ultrasound shows a well-defined, non-infiltrative, dumbbell-shaped subcutaneous lesion. Additional investigations are unnecessary and conservative management is recommended. **Case report:** we report three cases (2023 – 2024) of healthy infants (aged seven, 13, and 27 months) with sudden sternal/parasternal swelling, with no trauma or fever. One had localized pain. Two showed mildly elevated inflammatory markers. Ultrasound imaging revealed well-defined, hypoechoic, dumbbell-shaped subcutaneous lesions in every case. MRI (two cases) confirmed these findings. All received antibiotics due to diagnostic uncertainty, with complete resolution within six months. **Discussion:** the clinical presentation and favorable outcome support the diagnosis of SELSTOC. It is important to recognize this condition and choose conservative management with ultrasound monitoring, avoiding unnecessary medications and additional examinations.

Keywords: Sternum. Self-limiting diseases. Bone neoplasms. Diagnostic imaging.

SELSTOC: uma nova entidade a ter em conta – relato de caso

Resumo

Introdução: SELSTOC (Self-limiting sternal tumors of childhood) é uma lesão esternal/paraesternal autolimitada, de crescimento rápido e etiologia desconhecida, descrita em 2010. Idade mediana de 16 meses. Pode haver inflamação local, sem envolvimento sistémico, e ligeira elevação dos parâmetros inflamatórios. Ecograficamente descrita como lesão subcutânea bem delimitada, não infiltrativa, em forma de ampulheta. Não estão recomendados outros exames. A abordagem é conservadora. **Relato do caso:** Descrevemos três casos (2023 – 2024) de crianças saudáveis (7, 13, 27 meses) com tumefação esternal/paraesternal súbita, sem trauma ou febre. Um com dor; dois com elevação dos parâmetros inflamatórios. Em todos, ecografia com lesões subcutâneas hipoecogenicas, bem delimitadas, em forma de ampulheta. RMN (dois casos) confirmatória. Todos medicados com antibiótico por dúvida diagnóstica. Resolução completa em seis meses. **Discussão:** A apresentação clínica e evolução favorável sustentam o diagnóstico de SELSTOC. Reconhecer esta entidade é fundamental para adotar uma atitude conservadora, com vigilância ecográfica, evitando terapêuticas e exames desnecessários.

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Keypoints

What is known

– SELSTOC is an entity that was first described only in 2010, which, when considered, can help avoid unnecessary tests and treatments.

What is added

– it is a relatively newly recognized entity, with only a limited number of cases described in the literature. We present three cases that we identified over the course of a year in our hospital.

Introduction

Chest wall tumors, particularly in the sternum, are rare. The extensive differential diagnosis includes both benign (bone cysts, fibrous dysplasia and chronic non-bacterial osteomyelitis) and malignant (Ewing's sarcoma, chondrosarcoma and rhabdomyosarcoma)[1,2]. Because a sternal mass is often alarming, it is important to identify the clinical features of benign processes.

In 2010, Winkel et al.[1] reported a new entity known as self-limiting sternal tumors of childhood (SELSTOC) for the first time. A retrospective study was conducted in a pediatric oncology center in the Netherlands between 2001 and 2009, where 14 children presented with a presternal or parasternal rapidly growing lesion with spontaneous regression. The etiology was most likely an aseptic inflammatory reaction of a currently unknown origin. The median age was 16 months (range: seven – 50 months). There was no history of infection, trauma, or neoplasia. Physical examination revealed solid tumors with a median diameter of 3 cm (range: 1 – 4.5 cm), some with local pain or local heat and red/blue colored skin. Increased body temperature was sometimes observed, with no signs of systemic illness. Chest X-rays revealed soft tissue swelling with no bone involvement. They suggest avoiding this examination given its limited diagnostic yield. The most informative examination was ultrasonography, which typically showed a lesion with a hypoechoic and inhomogeneous appearance, usually dumbbell shaped, involving (or in contact with) the sternal and/or costal cartilages, with presternal/parasternal and retrosternal/mediastinal components, without involving the skin or infiltrating bone or muscle. Color Doppler ultrasound examination showed no significant internal vascularity. Computed tomography (CT) and magnetic resonance imaging (MRI) showed an increase in the distance between the centers of ossification of the sternum due to inflammation of the cartilage. However, these imaging tests are neither particularly informative nor essential for this diagnosis. Laboratory inflammatory parameters were not, or only slightly, raised and microbiologic cultures were negative in all patients. Both biopsy and histopathology samples were negative for malignancy. All tumors decreased in size within one month, in patients both with (six patients) and without antibiotics. On physical examination, the tumors disappeared within six months. To prevent invasive diagnostic interventions and unnecessary treatment, the recommendation is to adopt a wait-and-see approach, potentially treating the patients with anti-inflammatory medication and performing ultrasound follow-up until the lesion has regressed.

Sánchez et al.[3] reported five cases (2012 – 2019) of rapidly growing sternal lesions for which the clinical and radiological features were compatible with SELSTOC. Ultrasound was the diagnostic test performed in each case, showing the typical dumbbell-shaped lesion but no lesions in the bone or underlying cartilage in the sternum, ribs, or sternocostal joints. In contrast with benign masses, malignant sternal/parasternal lesions show aggressive signs of destruction and invasion of neighboring structures and tend to increase in size over time. They confirmed that in the absence of alarming symptoms and laboratory markers, watchful waiting could be an appropriate therapeutic approach. They suggested that the first ultrasound should be performed two weeks to one month after the initial presentation. Preventive routine medical examinations can be spaced out after the lesion has decreased in size and stopped after it disappears.

SELSTOC is a rare entity described in very few published studies around the world and especially in our country. Thus, the incidence and prevalence in the pediatric population remain unknown. Therefore, we consider it important to present three clinical cases compatible with this diagnosis, occurring over the past year (2023 – 2024), in the pediatric department of a hospital serving a broad population in the Lisbon Metropolitan Area (approximately 930,000 children), which may contribute to a better understanding of this disease.

Case reports

Case 1

January 2024: a healthy seven-month-old female infant presenting with a sudden onset of painless sternal swelling, which developed in one day. No history

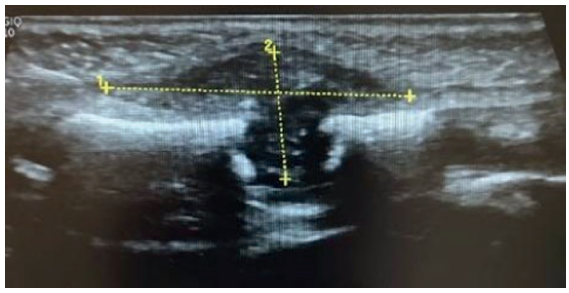

Fig. 1. Case 1: ultrasound of sternal lesion with dumbbell-shaped subcutaneous lesion.

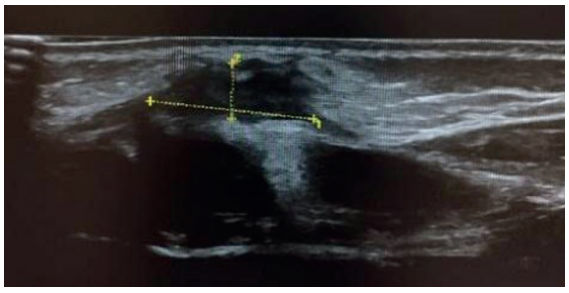

Fig. 2. Case 2: ultrasound of sternal lesion with dumbbell-shaped subcutaneous lesion.

of trauma or fever. Diagnosed with bronchiolitis the previous week. No relevant personal or family medical history. On physical examination, a 2 x 3 cm elastic swelling was observed in the sternal manubrium. No tenderness upon palpation and no signs of associated inflammation. The laboratory evaluation showed a slight increase in C-reactive protein (1.76 mg/dL), with no other abnormalities. A chest X-ray revealed slight edema of the subcutaneous tissue and a widening of the space between the segments of the sternum, with no bone involvement. An initial ultrasound performed on the day of admission showed a "space-occupying lesion in the subcutaneous tissue measuring 19.4 x 25.9 x 10 mm, with regular borders, hypoechoic with a small, more anechoic central area, dumbbell-shaped" (Fig. 1). The case was discussed with the radiology team regarding the possibility of a SELSTOC. The images were reviewed and compared with descriptions in the literature, supporting this as a diagnostic hypothesis. However, due to diagnostic uncertainty as to a potential abscess or infected dermoid cyst, cefuroxime was initiated for two weeks. A follow-up ultrasound was performed after two weeks, showing ultrasound findings similar to those of the initial examination. After one month, the patient was re-evaluated, with the swelling almost imperceptible, and an MRI was conducted, confirming the diagnosis of SELSTOC: "edematous changes centered at the cartilaginous junction between the sternal manubrium and the body of the sternum, with a dumbbell-shaped morphology, with no signs of periosteal reaction, suggestive of SELSTOC." Two months after its onset, the lesion had completely resolved.

Case 2

January 2024: a healthy 13-month-old female child presenting with a sudden onset of swelling in the lower third of the sternum, which developed in one day. No history of trauma or fever, and no relevant personal or family medical history. On physical examination, a 2 x 2 cm swelling was noted, located 1 cm from the left inferior lateral margin of the sternum, with a firm consistency and tenderness on palpation. No other signs of associated inflammation. The laboratory evaluation showed no changes. An ultrasound was performed, revealing "thickening and increased echogenicity of the soft tissues at the left parasternal region, measuring 20 x 8 mm, that did not extend to the underlying costal cartilage or the joint space. The described semiological findings suggest a probable local inflammatory process, which could be consistent with the clinical hypothesis of SELSTOC" (Fig. 2). Due to diagnostic uncertainty regarding costal chondritis, treatment with amoxicillin and clavulanic acid was initiated for one week. A follow-up ultrasound was performed after two weeks, showing a slight reduction in size (14 x 4 mm). After one month, clinical and imaging resolution was observed.

Case 3

November 2023: given these two recent cases, we reviewed a previous case of a 27-month-old male child, previously healthy, who was referred to General Pediatrics due to a mass in the right anterior chest wall that had been developing for three to four weeks, with no associated pain, warmth, or redness (Fig. 3). No relevant personal or family medical history. The laboratory evaluation showed a slight increase in C-reactive protein (1.06 mg/dL), with no other abnormalities. An ultrasound was performed, revealing a space-occupying lesion at the right-sided chondro-sternal junction, measuring 39 x 35.4 x 18 mm (Fig. 4). The lesion appeared to be an expansive cartilaginous mass, heterogeneous in nature, with apparent cortical rupture on its anterior aspect, extending into the adjacent soft tissues, with edema observed in the pectoralis major

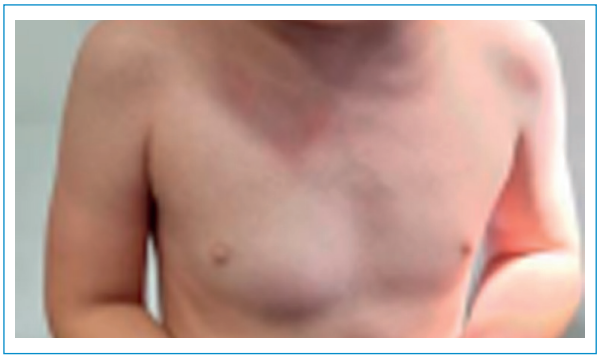

Fig. 3. Case 3: physical examination of a child with right-sided parasternal lesion.

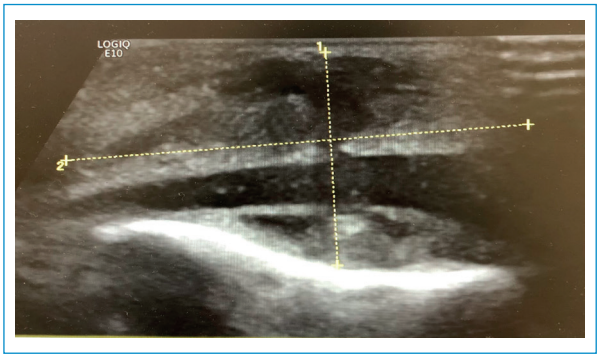

Fig. 4. Case 3: ultrasound of sternal lesion with dumbbell-shaped subcutaneous lesion.

and subcutaneous adipose tissue. Given the diagnostic hypotheses of a tumor or an infectious process, an MRI was performed for etiological diagnosis. The MRI revealed a mass in the middle portion of the fourth right-sided chondro-sternal joint, with a focal transverse disruption, resembling a fistulous tract, and an area of liquefaction within the cartilage extending outside the cartilage, simulating a microabscess. Suspecting chondritis with a microabscess, it was decided that he should be hospitalized for further evaluation and antibiotic therapy with cefuroxime and clindamycin. The patient was hospitalized for five days, with a gradual reduction in the size of the swelling, measuring 2 cm at the time of discharge. Antibiotic therapy was continued for two weeks, and the patient was re-evaluated in a follow-up consultation, where the swelling was nearly imperceptible and no signs of associated inflammation were present. An ultrasound was performed six months later, showing no changes.

This case was reviewed at the time of cases 1 and 2. A comparison of the available images was made, and after a clinical discussion with the radiology team, the diagnosis of SELSTOC was confirmed, contrary to the initial clinical hypothesis of chondritis with a microabscess, as the seemingly fistulous shape corresponded to the dumbbell-shaped morphology typically observed in this entity on ultrasound.

Discussion

According to the literature published in this regard, our findings were similar to those of Winkel et al.[1], Adri and Kreindel[2], Sánchez et al.[3], Yamane et al.[4], Moreira and Marchiori[5], Fuente-Lucas et al[6] and Blouet et al.[7]. Since this entity was initially not well-known to our medical team, it was not considered as a hypothesis in the first case in November 2023 (case 3), which led to the patient being hospitalized and another diagnosis being assumed. However, in the cases from January 2024 (cases 1 and 2), this diagnosis was immediately considered as a possibility and was discussed with the radiology team at an early stage, who had to review the literature and ultimately confirmed the diagnosis. Nonetheless, once again due to inexperience in the management of these lesions, antibiotic therapy was initiated in each case due to diagnostic uncertainty. In each case, an ultrasound was performed, and upon comparison of the images, the typical dumbbell-shaped, non-infiltrative lesions were detected. In two of the cases, MRI was also performed, which would have been unnecessary given the benign characteristics presented. Ultrasound monitoring was performed in all cases, showing a reduction in the size of the lesion after the first month and complete resolution within less than six months.

In summary, SELSTOC are rapidly growing benign sternal masses in which the etiology remains unknown. It is crucial to add this disease to the list of differential diagnoses for sternal masses, since it is a self-limiting disease with no need for treatment. A diagnosis of SELSTOC should be considered in cases of a sudden onset of presternal or parasternal lesions, with no history of trauma, infection, or neoplasia, in young children. Mild local signs of inflammation signs may be present, but with no systemic involvement. Slightly elevated C-reactive protein and white blood cell counts may be observed in SELSTOC; however, not to the extent that they are raised in other infectious diseases. In children with rapidly growing chest wall tumors, the differential diagnoses are malignant tumors including Ewing sarcoma, osteosarcoma, malignant lymphoma, metastatic tumors, and infections such as tuberculous tumors. However, these diseases often affect cortical bone, which are distinct from SELSTOC, which rarely

affect the bone itself. Characteristic imaging findings include widening of the ossification centers and a dumbbell-shaped mass. The first choice in imaging is ultrasound, which is cost-effective and free from radiation. CT and MRI should be reserved for ambiguous cases[4].

Pediatricians should be aware of this new entity and consider this etiology in the differential diagnosis of sternal lesions to avoid unnecessary diagnostic and treatment methods.

Author contributions

C. Picão de Carvalho: conceptualization; writing (original draft); writing (review and editing); E. Freitas: conceptualization; J. Sousa: validation, visualization, writing (review and editing). S. Bacalhau: validation, visualization, writing (review and editing); H. Neves: validation, visualization, writing (review and editing); H. de Castro Faria: validation, visualization, writing (review and editing).

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Conflicts of interest:

None.

Ethical considerations

Protection of humans and animals. The authors declare that no experiments involving humans or animals were conducted for this research.

Confidentiality, informed consent, and ethical approval. The authors have followed their institution's confidentiality protocols, obtained informed consent from patients, and received approval from the Ethics Committee. The SAGER guidelines were followed according to the nature of the study.

Declaration on the use of artificial intelligence. The authors declare that no generative artificial intelligence was used in the writing of this manuscript.

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