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Three-Dimensional Tomographic Analysis of the Mandibular Condyle in People with Sickle Cell Disease

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ABSTRACT

***Introduction*: The literature reports joint changes in the knee and hip of people with sickle cell disease (SCD), but morphological modifications in mandibular condyles are little described.**

***Objective*: This study aimed to evaluate, by computed tomography (CT), morphological mandibular condyle modifications in people with SCD.**

***Material and methods*: We have selected 70 mandibular CT of people with SCD (test group) and 70 CT, matched for age and sex, in a 1:1 ratio, of people without SDC or other diseases affecting bone metabolism (control group). Linear and volumetric measurements and qualitative assessment of the mandibular condyles were performed.**

***Results*: The results showed a lower height of the mandibular condyles in men (p=0.05) and the presence of erosion and flatting of the condylar bone surface in women with SCD. Conclusions: The lower height of the mandibular condyle in men may contribute to the more retrusive positioning of the mandible in this group.**

Keywords: Computed tomography, sickle cell anemia, sickle cell disease, temporomandibular joint.

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I. INTRODUCTION

A mutation in the beta-globin gene, resulting in S hemoglobin (SHb) formation, causes sickle cell disease (SCD). SHb, in hypoxic situations, changes the shape of red blood cells that assume the aspect of a sickle. This unconventional configuration determines its early hemolysis and, consequently, chronic anemia. Vaso-occlusive phenomena and multisystem inflammatory changes characterize SCD [1], [2]. Several skeletal bones maintain erythrocyte production to compensate for the anemia. This bone activity determines bone marrow expansion and modifies its morphology. This expansion frequently occurs in facial bones, with a tendency for maxillary protrusion, mandibular retrusion, and dental malocclusion [3]-[5].

The literature also describes joint changes, especially in the knee and hip joints [6]-[9]. The pathophysiology of bone-joint involvement in SCD is not fully understood, and the most critical event is the occlusion of the small vessels with consequent ischemia [6]. Vascular occlusion results in medullary infarctions and osteonecrosis, which affect the epiphyses. Both processes can result in morbidity and pain in patients with SCD, with the most common locations being the head of the femur, followed by the humerus and knee. Epiphyseal infarctions occur in the subchondral bone, followed by eventual collapses. Radiographs may show radiolucent areas. Initially, the weird images appear on the epiphyses after it occurs faceting, sclerosis, and subsequent subchondral collapse [10].

Despite this, the possible modifications of the mandibular condyle in people with SCD are little discussed. Considering the importance of the mandible in facial growth, mastication, dental balance, and aesthetics, this study aimed to evaluate, by computed tomography (CT), morphological mandibular condyle modifications in people with SCD.

II. Methods

This cross-sectional study was approved by the Research Ethics Committee of the Professor Edgard Santos University Hospital of the Federal University of Bahia, Salvador-BA, Brazil. The test group comprised 70 mandibular CT from people diagnosed with SCD. The control group consisted of mandibular CT of 70 people without SCD, paired by sex and age, in a 1:1 ratio. The inclusion criteria in both groups were people at the final stage of maturation of the cervical vertebrae (SMCV) [11], without other systemic diseases affecting bone metabolism, and who agreed to participate in the study by signing the Informed Consent Form.

CT was performed in a 64-channel multidetector device (Aquilion 64®, Toshiba, Japan). Axial volumetric sections were obtained (voxels 0.5 x 0.5 x 0.3 mm; 150 mA, 120 kV, FOV 22cm), with no gantry tilt, and no intravenous iodinated contrast injection.

The images were exported at DICOM (Digital Imaging and Communication in Medicine) files to electronic media (CD-ROM). The CT files were processed in Dolphin Imaging software® v. 11.5 Premium (Dolphin Imaging & Management Solutions, Chatsworth, California, USA) to obtain the multiplanar and three-dimensional (3D) reconstructions.

The volume of the condyles was measured by the Volume Sculpting tool. Considering the mandibular notch as the reference point, a line parallel to Frankfort's Horizontal Plane (FHP) was determined. The condyle was segmented, and this line was its lower limit, and the volume in cm^3 was automatically calculated (Fig. 1).

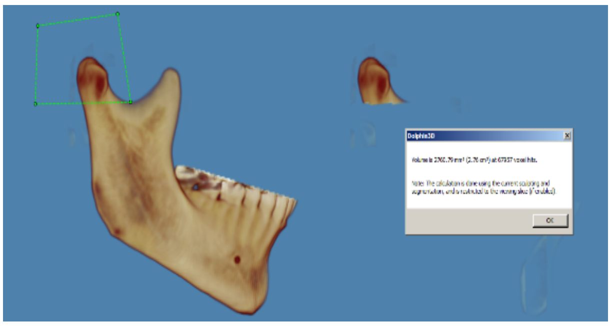

Fig. 1. Condylar volume calculation using the Volume Sculpting tool, at Dolphin® software.

After the following cephalometric points were located:

a) Posterior Condylium (pCo): posterior point of the mandibular condyle;

b) Upper Condylium (uCo): highest point of the mandibular condyle.

A line parallel to the FHP was drawn through the mandibular notch, and then the following measurements were performed (Fig. 2):

a) Condyle height: distance between uCo and its orthogonal projection in the line parallel to the FHP;

b) Anteroposterior width of the condyle: distance between pCo and the most anterior point of the condyle, parallel to the FHP;

c) The transverse width of the condyle: measured in a posterior view of the mandible, between the most mesial and distal points of the condyle.

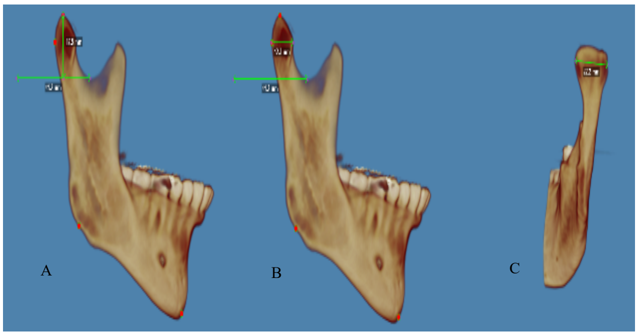

Fig. 2. Linear measurements of the mandibular condyles (A: height of the condyle, B: anteroposterior width; C: cross width).

In multiplanar reconstructions, the following condylar degeneration signs were analyzed: erosion of the cortical condylar and/or joint eminence; flatting of the condyle head with loss of its convexity; and presence of osteophytes.

A single examiner performed all analyses, and both condyles were evaluated in each CT. The Kolmogorov-Smirnov test tested the normal distribution, and the student's t-test compared the data. The comparison of qualitative data was performed using the chi-square test. The significance level of 5% was adopted for the analysis.

III. Results

There were 38 women (54.3%) and 32 men (45.7%), in each experimental group, with a mean age was 35 years. The evaluation of the condyles volume showed no significant difference between the groups (p= 0.32), as well as their transverse width (p= 0.08) and their anteroposterior dimension (p= 0.07). However, the height of the condyles (p= 0.01) was statistically lower in the SCD group. When assessing sex, no significant difference was observed for any of the parameters in women. There was a statistical difference between the groups only in men's condyle height (p= 0.05). Tables I and II show these data.

TABLE I: Condylar Dimensions, according to the Experimental Group

	SCD	Control	P-value
Condyle volume (cm^3)	1.89	1.86	0.32
Condyle height (mm)	16.3	17.0	0.01*
Condyle dimension AP (mm)	11.3	11.5	0.07
Condyle width (mm)	18.4	18.7	0.08

SCD= sickle cell disease; AP= anteroposterior; *statistical difference

The qualitative evaluation of the mandibular condyles revealed that in the SCD group, there were significantly more cases of erosion of the condylar surface (p= 0.03). Regarding the presence of flatting or osteophytes, no difference was found between the groups evaluated (p= 0.12 and p= 0.74, respectively). Table III shows these results. According to sex, we observed that in women, there was a significantly higher number of cases of flatting (p= 0.0007) and bone surface erosion (p= 0.05) in the SCD group. For men, no difference was observed between the groups in these parameters.

TABLE II: CONDYLAR DIMENSIONS, ACCORDING TO THE EXPERIMENTAL GROUP AND SEX

	WOMEN			MEN		
	SCD	Control	P-value	SCD	Control	P-value
Condyle volume (cm^3)	1.8	1.8	0.35	2.0	1.9	0.40
Condyle height (mm)	16.2	16.9	0.06	16.3	17.1	0.05 *
Condyle dimension AP (mm)	10.9	11.2	0.12	11.7	11.9	0.20
Condyle width (mm)	17.8	18.2	0.19	19.0	19.4	0.15

SCD= sickle cell disease; AP= anteroposterior; *statistical difference

TABLE III: FREQUENCY OF THE MORPHOLOGICAL CONDYLAR MODIFICATIONS, ACCORDING TO THE EXPERIMENTAL GROUP

		Groups		P-value
		SCD	Control	
Flatting	Present	67 (47.9%)	54 (38.6%)	0.12
	Absent	73 (52.1%)	86 (61.4%)	
Osteophytes	Present	22 (15.7%)	20 (14.3%)	0.74
	Absent	118 (84.3%)	120 (85.7%)	
Erosion	Present	73 (52.1%)	67 (47.9%)	0.03*
	Absent	55 (47.9%)	85 (52.1%)	

SCD= sickle cell disease; *statistical difference

IV. DISCUSSION

Although the condylar volume was not different between the groups, its height was lower in men with SCD. This lower height may result from vascular impairment inherent to the SCD and is responsible for the shorter length of the mandibular branch, as described by [12]. This could lead to a more retrusive positioning of the mandible, as already described in previous studies [3]-[5]. According to [12] and [13], ischemia and bone infarction compromise the epiphyseal growth and shortening of long bones in people with SCD.

Thus, it is possible to postulate that morphological condylar modifications may contribute to the Class II facial profile. Moreover, maxillary compensatory medullary expansion, more significant than in the mandible due to the lower thickness of its cortical, determines the maxillary protrusion characteristic of SCD [4], [5], [14].

We also evaluated the presence of signs that characterize degeneration of temporomandibular joints. In the SCD group, there were significantly more cases of condylar surface erosion, which agree with [10].

When we evaluated sex, women with SCD also presented more cases of condylar flatting. A systematic review found a positive correlation between estrogen levels and temporomandibular dysfunctions [15]. This finding, therefore, suggests that women with SCD may present more significant joint degeneration by a combination of SCD and hormonal characteristics.

In this study, the genotypic variations of SCD were unavailable, which may determine substantial variations in the clinical manifestations of the disease.

V. CONCLUSION

We concluded that men with SCD have lower condylar height, and women with SCD have a greater frequency of condylar erosion and flatting. The lower condylar height may contribute to the more retrusive positioning of the mandible in SCD people.

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CONFLICT OF INTEREST

Authors declare that they do not have any conflict of interest.

REFERENCES

[1] Kato GJ, Piel FB, Reid CD, Gaston MH, Ohene-Frempong K, Krishnamurti L, et al. Sickle cell disease. *Nat Rev Dis Primers*. 2018; 4: 18010.

[2] Sundd P, Gladwin MT, Novelli EM. Pathophysiology of Sickle Cell Disease. *Annu Rev Pathol*. 2019: 263-292.

[3] Pithon MM, Palmeira LMV, Barbosa AAL, Pereira R, Andrade ACDV, Coqueiro RS. Craniofacial features of patients with sickle cell anemia and sickle cell trait. *Angle Orthod*. 2014; 4(5): 825-829.

[4] Basyouni A, Almasound NN, Al-Khalifa KS, Al-Jandan BA, Al Sulaiman OA, Nazir MA. Malocclusion and craniofacial characteristics in saudi adolescents with sickle cell disease. *Saudi J Med Med Sci*. 2018; 6(3): 149-154.

[5] Santos HLR, Barbosa IS, Oliveira TFL, Sarmento VA, Trindade SC. Evaluation of the maxillomandibular positioning in subjects with sickle-cell disease through 2- and 3- dimensional cephalometric analyses: A retrospective study. *Medicine*. 2018; 7(25): e11052.

[6] Silva Junior GB, Daher EF, da Rocha FA. Osteoarticular involvement in sickle cell disease. *Rev Bras Hematol Hemoter.* 2012; 34(2): 156-64.

[7] Caracas MS, Jales SP, Jales Neto LH, da Silva Castro JC, Suganuma LM, Fonseca GH, *et al.* Temporomandibular joint arthritis in sickle cell disease: a case report. *Oral Surg Oral Med Oral Pathol Oral* Radiol. 2013; 5(2): e31-5.

[8] Braimah RO, Oladejo T, Olarinoye TO, Adetoye AO, Osho PO. A multidisciplinary approach to the management of temporomandibular joint ankylosis in a sickle-cell anemia patient in a resource-limited setting. *Ann Maxillofac Surg*. 2016: 130-4.

[9] Adesina OO, Neumayr LD. Osteonecrosis in sickle cell disease: an update on risk factors, diagnosis, and management. *Hematology Am Soc Hematol Educ Program*. 2019; 351-358.

[10] Vanderhave KL, Perkins CA, Scannell B, Brighton BK. Orthopaedic manifestations of sickle cell disease. *J Am Acad Orthop Surg.* 2018; 6(3): 94-101.

[11] Baccetti T, Franchi L, Mcnamara JA. The cervical vertebral maturation (CVM) method for the assessment of optimal treatment timing in dentofacial orthopedics. *Semin Orthod*. 2005; 11: 119-29.

[12] Ganguly A, Boswell W, Aniq H. Musculoskeletal manifestations of sickle cell anaemia: a pictorial review. *Anemia*. 2011; 2011: 794283.

[13] Ejindu VC, Hine AL, Mashayekhi M, Shorvon PJ, Misra RR. Musculoskeletal manifestations of sickle cell disease. *Radiographics*. 2007; 4: 1005-21.

[14] Brown DL, Sebes JI. Sickle cell gnathopathy: radiologic assessment. *Oral Surg Oral Med Oral Pathol*. 1986; 1(6): 653-656.

[15] Berger M, Szalewski L, Bakalczuk M, Bakalczuk G, Bakalczuk S, Szkutnik J. Association between estrogen levels and temporomandibular disorders: A systematic literature review. *Prz Menopauzalny*. 2015; 4(4): 260-70.

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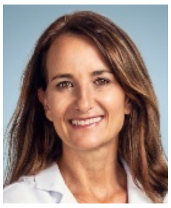

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