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The significance of the clavicle on shoulder girdle function

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Background: Patients with cleidocranial dysplasia (CCD) can have a congenital partial or total absence of the clavicle. The aim of this study was to evaluate the functional shoulder score in these patients.

Material and methods: Patients with CCD who were members of a social media group were invited to take part in an Internet-based survey. The questionnaire was composed of 3 general questions, a question concerning partial or total absence of the clavicle, and 3 patient-based shoulder scores (American Shoulder and Elbow Surgeons score; shortened Disabilities of the Arm, Shoulder, and Hand score; and patient-based Constant-Murley score).

Results: Thirty-six patients (27 women, 9 men) with a mean age of 40 years participated in the survey; 17 patients had a bilateral absence, 16 patients had a bilateral partial absence, and 3 patients had a total absence on one side and a partial absence of the clavicle on the other side. The average patient-based Constant-Murley score was 79; shortened Disabilities of the Arm, Shoulder, and Hand score, 11; and American Shoulder and Elbow Surgeons score, 93. There was no significant difference in the scores between left and right. There was no correlation between partial and total absences and the shoulder scores.

Conclusion: Patients with self-reported CCD are subjectively normal to their peers when they are evaluated with 3 common shoulder scores. There was no correlation between partial and total absences and the shoulder scores.

Level of evidence: Survey Study, Patients.

Keywords: Shoulder function scores; cleidocranial dysplasia; clavicle; absence; social media

The clavicle is unique, being the only long bone to ossify by the intramembranous process and the first bone in the body to ossify (fifth week of fetal life). Its configuration presents a double curvature, convex forward in the medial half and concave forward in the lateral half.[2] This shape, resembling that of the clavicula (Latin diminutive of *clavis*, key) of the musical score, accounts for its name.

The clavicle can be seen only in a small percentage of mammals.[22] During evolution, all sea mammals and those adapted for running lost their clavicle because their movement occurs only in the anteroposterior plane and there is no need for lateral swinging. In contrast, prehensile forelimbs (primates) and bats have a clavicle.[14]

The importance of the clavicle to the functionality of the human shoulder girdle has been the subject of debate and has been evaluated on the basis of the functional outcome in patients after a total claviculectomy.[3,18,25] In contrast, to our

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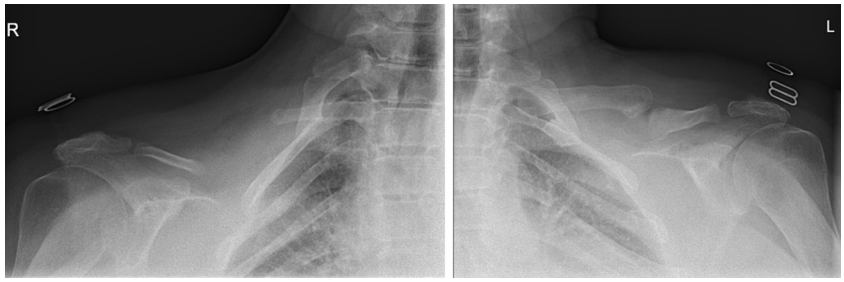

Figure 1 Right and left partial absence in a patient with cleidocranial dysplasia (CCD).

knowledge, functional shoulder scores in patients with congenital partial or total absence of the clavicle and without any operative treatment have not been described before.

Cleidocranial dysplasia (CCD) is a general skeletal condition so named from the collar bone (cleido-) and cranium deformities.[1,4,20] It is usually autosomal dominant, but in some cases the cause is not known. In these patients, the clavicle can be partly missing (Fig. 1). In 10% of cases, they are completely missing. If the collar bones are completely missing or reduced to small vestiges, this allows hypermobility of the shoulders, including the ability to touch the shoulders together in front of the chest. The defect is bilateral 80% of the time.

The aim of this study was to evaluate patient-based shoulder scores in adult patients with CCD and a partial or total absence of the clavicle.

Material and methods

Study subjects

Patients with CCD who were members of a social media group were invited to take part in an Internet-based survey. Inclusion criteria were patients diagnosed with CCD, minimum age of 18 years, and partial or total absence of a minimum of one shoulder or both shoulders.

Survey design and distribution

A questionnaire was developed for the current study composed of 3 general questions (age, gender, dominant side), a question concerning partial or total absence of the right and left clavicle, and 3 patient-based shoulder scores (American Shoulder and Elbow Surgeons [ASES] score[16]; shortened Disabilities of the Arm, Shoulder, and Hand [QuickDASH] score[6]; and patient-based Constant-Murley score [CS][11]) for both arms. Last, the participants were able to write a comment on the following question: What are things you can and you cannot do compared with people with a total clavicle? The answers were classified by difference in range of motion, difference in strength, or others.

The questionnaire was trialed on 3 fellowship-trained orthopedic sports medicine surgeons not involved in the study, revised accordingly, and then distributed. Survey distribution and analysis followed a similar process as was outlined in a previous survey study on the surgical treatment of anterior sternoclavicular dislocation.[23] A reminder was sent to the members of the group at 1 month after the initial invitation. Responses were de-identified with study numbers.

Statistical analysis was performed with SPSS Statistics for Windows (version 21.0; IBM, Armonk, NY, USA). The evaluation between the side and the scores/power was performed with the Mann-Whitney *U* test, between the gender and the scores/power also with the Mann-Whitney *U* test, and between the age and the scores/power with the Spearman rank correlation.

Results

Thirty-six patients (27 women, 9 men) participated in the survey (Table I). The mean age was 40 years (range, 22-68 years). Thirty-four patients were right dominant, and 2 patients were left dominant; 17 patients (2 men, 15 women) had a bilateral absence of the clavicle, 16 patients had a bilateral partial absence of the clavicle (7 men, 9 women), and 3 patients (3 women) had a total absence on one side and a partial absence on the other side. The average patient-based CS on the right side was 79 (standard deviation [SD] ± 15). The average patient-based CS on the left side 79 (SD ± 15).

The average QuickDASH score on the right side was 11 (SD ± 16). The average QuickDASH score on the left side was 11 (SD ± 18). The average ASES score on the right side was 93 (SD ± 15). The average ASES score on the left side was 93 (SD ± 15). The average strength in the right arm was 4 kg (SD ± 2.9 kg). The average strength in the left arm was 4 kg (SD ± 2.9 kg).

The oldest patient (68 years) scored very low on all scores (right ASES score, 25; left ASES score, 28; right QuickDASH score, 70; left QuickDASH score, 90; right patient-based CS, 8; left patient-based CS, 11) and mentioned that she did not have any problems before the age of 40 years. She had a total absence of function at both shoulders.

There was no significant difference in scores between left and right.