



The evaluation of superior semicircular canal in patients with unilateral cleft lip and palate using CBCT

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Abstract

Objectives The present study aims to evaluate the thickness and radiological patterns of the superior semicircular canal (SSC) in patients with unilateral cleft lip and palate (CL/P).

Methods Cone beam computed tomography (CBCT) images of the patients were evaluated in axial and Pöschl planes. CBCT images of 84 patients with unilateral CL/P and 168 healthy individual controls were included in the study. Three study groups were established: the CS–CL/P group (cleft side temporal bones of the CL/P patients), NCS–CL/P (non-cleft side temporal bones of the CL/P patients) and the control group. The radiological patterns of SSCs were categorized as dehiscence, papyraceous, normal, pneumatized and thick. The minimum bone thickness of SSC was measured.

Results It was found that the CS–CL/P group had a higher prevalence for SSCD compared to both the NCS–CL/P group and the control group. CS–CL/P group had a higher prevalence of dehiscence type and papyraceous type compared to the control group. The SSC thickness on the CS–CL/P patients was thinner than the NCS–CL/P patients and the control group sides ($p = 0.033$ and $p < 0.001$, respectively).

Conclusions The mean thickness of SSC was found significantly lower in the CS–CL/P group compared to both the NCS–CL/P group and the control group. The elevated prevalence of dehiscence and papyraceous types in the CS–CL/P group compared to the control group implies that the presence of a cleft may be a predisposing factor for these types.

Keywords Cleft lip and palate · Cone beam computed tomography · Dehiscence · Semicircular canal · Temporal bone

Introduction

Cleft lip and palate (CL/P) is a common congenital defect that causes feeding and speech difficulties. It is more common in males compared to females. Dysfunction of palatal muscles and Eustachian tube and middle ear problems are seen in patients with cleft lip and palate [1]. Previous studies highlighted that patients with CL/P are commonly associated with middle ear diseases and hearing loss [2, 3]. Some

authors stated that it is important to examine the associated conditions with CL/P for a better understanding of the etiology of this congenital defect [4, 5].

Superior semicircular canal dehiscence (SSCD) is a phenomenon that has been the focus of many studies in recent years. Minor et al. [6] stated that the bony dehiscence of the superior semicircular canal is associated with the Tullio phenomenon, oscillopsia, changes in middle ear pressure and vestibular disorders. Previous studies highlighted the otologic problems of patients with CL/P [1, 7]. Altun et al. found that CL/P patients had a higher incidence of SSCD compared to normal patients [7]. However, there is still a necessity to heighten focus and conduct a more comprehensive examination pertaining to correlation between CL/P and the structure of the SSC.

The use of cone-beam computed tomography (CBCT) for maxillofacial imaging has become widespread in recent years. CBCT provides lower scanning time and radiation dose compared to computed tomography (CT) [8] and it can ensure repeatable and reliable measurements [9, 10]. In

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addition, there has been a growing preference among otolaryngologists for the use of CBCT due to its ability to provide a comprehensive assessment of the temporal bone while also being compact in size [11].

The study aims to explore the association between unilateral CL/P and thickness and radiological patterns of the superior semicircular canal (SSC), considering its potential impact on otologic issues and middle ear problems in individuals with CL/P. Understanding these associations is crucial for gaining insights into the etiology of CL/P and improving the overall comprehension of how a congenital defect-like CL/P may be linked to specific otologic conditions, such as SSCD.

Materials and methods

The present study was approved by the Ethics Committee of University (2021–116.) From the pool of 479 unilateral CL/P patients with available CBCT scans in archive of the Dentomaxillofacial Radiology Department, 33 females and 52 males were randomly selected. One CBCT scan of a female individual was excluded due to its low-quality. Among the 809 healthy individuals with available CBCT scans in our archives, individuals who were matching in age and gender with the CL/P patients were initially sorted. For each CL/P patient, the selection of two healthy controls among the corresponding age–gender matched individuals was randomize (control patients: 64 females and 104 males). Three study groups were established: the CS–CL/P group

(cleft side temporal bones of the CL/P patients), NCS–CL/P (non-cleft side temporal bones of the CL/P patients) and the control group.

High-quality CBCT images with visualization of the temporal region were included in the study. Patients with a history of surgery or trauma in the craniofacial region were excluded from the study, as well as low-quality images caused by artifacts, such as metal or movement artifacts.

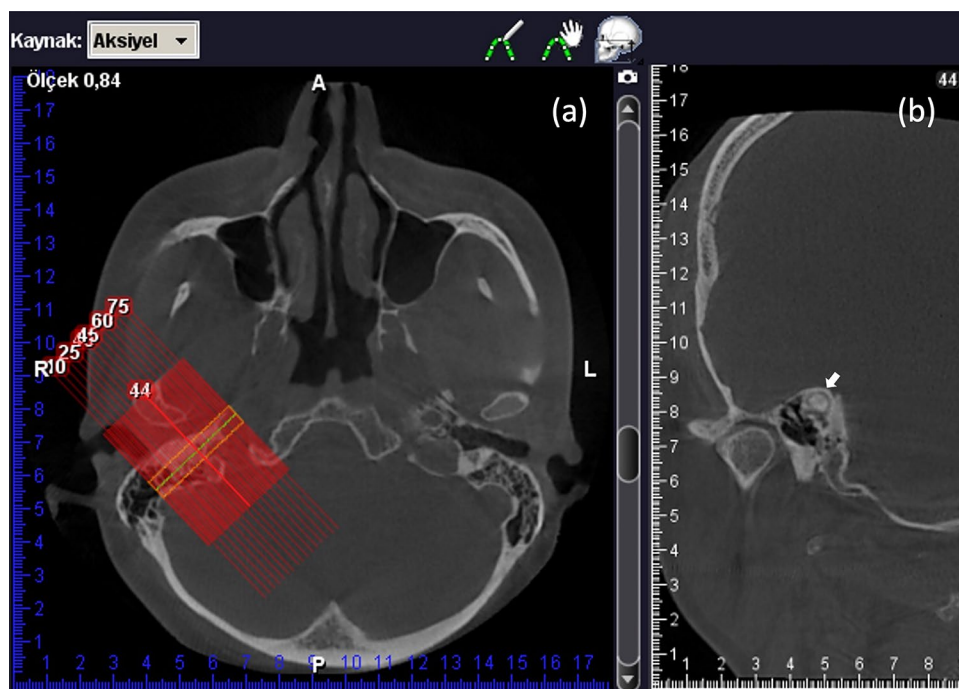
Image analysis

The maxillofacial imaging of the patients was performed with a CBCT device (Planmeca ProMax® 3D Mid, Helsinki, Finland; exposure parameters: 90 kV, 10 mA, 27 s scan time, voxel size: 0.4 mm³). Image analysis was performed using the Planmeca, Romexis viewer software in a dark and quiet room.

CBCT images were reformatted according to the previous studies evaluating SSC [7, 12]. The images were examined in the axial plane. The Pöschl projection was prepared perpendicular to the long axis of the petrous bone, at approximately an angle of 45 degrees with the sagittal and coronal planes (Fig. 1a). It was specifically drawn parallel to the SSC. SSC was observed as a ring in the Pöschl projection (Fig. 1b). SSCs were examined in the axial and Pöschl planes. The slice thickness was 0.4 mm. The minimum bone thickness of SSC was measured from the inner surface of the roof of SSC to the inner surface of the middle cranial fossa.

Cisneros et al. [13] delineated the boundaries of the normal pattern based on a study conducted by Crovetta [14]

Fig. 1 Pöschl projection with reformatted cone beam computed tomography images. **a** Angle of reformation showed on axial plane, **b** ring view of SSC indicated by arrow in Pöschl projection



et al. The normal pattern of SSC was characterized by bone thickness between 0.6 and 1.7 mm. Papyraceous (≤ 0.5 mm) and thick (≥ 1.8 mm) patterns were determined depending on whether they were thinner or thicker than the normal pattern. Dehiscent pattern was described as an absence of bone covering upon the SSC. Pneumatized pattern was described as a SSC with multiple supralabyrinthine cells presenting as a woven structure (Fig. 2a).

The images were evaluated by two experienced dentomaxillofacial radiologists. Three weeks later, 25% of all measurements were repeated to evaluate intra- and inter-observer reliability. Different assessments were discussed and consensus was reached for categorical variables.

Statistical analysis

Power analysis was performed to determine the sample size to be used in the study (G*Power 3.1.9.4). For this purpose, the sample size was calculated with the data of the only similar study [12] in the literature, as far as we know and the sample size was determined as 336 (effect size = 0.24; $\alpha = 0.05$; power = 0.94). Data were evaluated with the SPSS software package 20.0 (SPSS, Chicago, IL, USA). $p < 0.05$ was accepted as statistically significant. In tables summarizing categorical data, frequencies (n) and column percentages (%) were used to compare independent groups (Tables 1, 3, 5), while frequencies (n) and row percentages (%) were used to compare dependent groups (Table 2). Numerical data were presented with tables using mean \pm SD (standard deviation) and median (min–max). Whether the numerical measurements provided the assumption of normal distribution was tested with the Kolmogorov–Smirnov test. The Wilcoxon signed rank and Mann–Whitney U tests were used to compare SSC measurements. The Chi-square test/Fisher's exact test (for independent groups) and the McNemar–Bowker test (for dependent groups) were used to evaluate the relationships between categorical variables. The relationships between age and SSC thickness were evaluated by Spearman's correlation coefficients. For categorical variables, Cramer's V coefficients were calculated for the intra- and inter-observer agreement. Intraclass correlation coefficients

Table 1 Demographic data according to groups

	CL/P	Control
<i>Age</i>		
Mean \pm std. dev	17.4 \pm 3.7	17.4 \pm 3.7
Median (min–max)	16 (14–26)	16 (14–26)
<i>p</i>	1.000 [†]	
<i>Gender</i>		
Female	32 (38.1)	64 (38.1)
Male	52 (61.9)	104 (61.9)
Total	84 (100)	168 (100)
<i>p</i>	1.000 [‡]	

Data are shown as mean \pm standard deviation, median (minimum–maximum) and frequencies (column %). [†]Mann–Whitney U test. [‡]Chi-square test. CL/P: cleft lip and palate

(ICC) were calculated for intra- and inter-observer agreement in thickness measurements.

Results

According to the power analysis, the total sample size was decided as 336 sides. Eighty-four cleft sides and 84 non-cleft sides of 84 CL/P patients and 168 sides of 168 control patients (84 right sides of 84 control patients and 84 left sides of 84 control patients) were included in the study. Demographic information is shown in Table 1. Cramer's V coefficients for categorical variables were greater than 0.94 for intra- and inter-observer ($p < 0.001$). ICCs for thickness measurements were greater than 0.85 for intra- and inter-observer ($p < 0.001$).

The study population exhibited a relatively low frequency of thick and pneumatized SSC patterns. In addition, the pneumatized patterns that were observed tended to be thick, with a thickness of ≥ 1.8 mm. Given these findings, the authors combined the thick and pneumatized patterns into a single category, which is referred to as the "pneumatized/thick" type.

In 64.3% of the patients with CL/P, the superior semicircular canal (SSC) types were symmetrical on both the

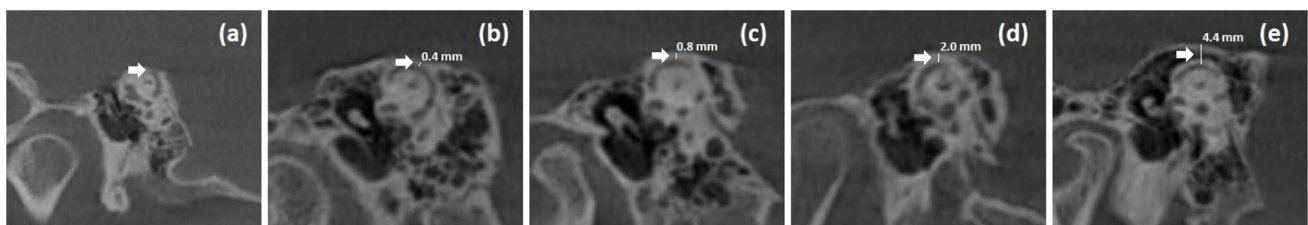


Fig. 2 Radiological patterns of SSCs with reformatted cone beam computed tomography images. **a** Dehiscent pattern, **b** papyraceous pattern, **c** normal pattern, **d** thick pattern, **e** pneumatized pattern

Table 2 SSC types on the cleft and non-cleft sides of patients with cleft lip and palate

SSC types		CS–CL/P				Total
		Dehiscent	Papyraceous	Normal	Pneum/thick	
NCS– CL/P	Dehiscent	9 (81.8)	0 (0)	2 (18.2)	0 (0)	11 (100)
	Papyraceous	2 (50)	2 (50)	0 (0)	0 (0)	4 (100)
	Normal	6 (9.4)	14 (21.9)	40 (62.5)	4 (6.2)	64 (100)
	Pneum/thick	0 (0)	2 (40)	0 (0)	3 (60)	5 (100)
	Total	17 (20.2)	18 (21.4)	42 (50)	7 (8.3)	84 (100)
<i>p</i>		<0.001*				

SSC types		CS–CL/P		Total
		Dehiscent/Papyraceous	Normal/Pneum/Thick	
NCS– CL/P	Dehiscent/Papyraceous	13 (86.7)	2 (13.3)	15 (100)
	Normal/Pneum/Thick	22 (31.9)	47 (68.1)	69 (100)
	Total	35 (41.7)	49 (58.3)	84 (100)
<i>p</i>		<0.001*		

Data are shown as frequencies(row %). McNemar–Bowker test. * $p < 0.05$ is indicated in bold. CL/P: cleft lip and palate, CS–CL/P: cleft side of CL/P, NCS–CL/P: non-cleft side of CL/P. Pneum: pneumatised, SSC: superior semicircular canal

Table 3 SSC types by groups

SSC types	CS–CL/P	Control	NCS–CL/P	Control
	Dehiscent	17 (20.2) [†]	14 (8.3)	11 (13.1)
Papyraceous	18 (21.4) [†]	9 (5.4)	4 (4.8)	9 (5.4)
Normal	42 (50) [†]	115 (68.5)	64 (76.2)	115 (68.5)
Pneumatized/Thick	7 (8.3) [†]	30 (17.9)	5 (6) [†]	30 (17.9)
Total	84 (100)	168 (100)	84(100)	168 (100)
<i>p</i>	<0.001		0.057	

SSC types	CS–CL/P	Control	NCS–CL/P	Control
	Dehiscent/Papyraceous	35 (41.7)	23 (13.7)	15 (17.9)
Normal/Pneum/Thick	49 (58.3)	145 (86.3)	69 (82.1)	145 (86.3)
Total	84 (100)	168 (100)	84 (100)	168 (100)
<i>p</i>	<0.001*		0.384	

Data are shown as frequencies(column %). Chi-square test. [†]indicates a statistically significant difference between the columns ([†]*p* values corrections with Bonferroni method). * $p < 0.05$ is indicated in bold. CL/P: cleft lip and palate, CS–CL/P: cleft side of CL/P, NCS–CL/P: non-cleft side of CL/P, Pneum: pneumatised, SSC: superior semicircular canal

cleft and non-cleft sides. The most frequently observed type was the normal type, which was present in 50% of the cleft sides and 76.2% of the non-cleft sides. Among the non-cleft sides classified as normal type SSC, 62.5% had corresponding cleft sides that were also classified as normal. However, among those with normal type SSC on the non-cleft side, 21.9% had papyraceous type on the cleft side and 9.4% had dehiscent type on the cleft side. A McNemar–Bowker test revealed that the SSC types of the cleft and non-cleft sides in CL/P patients were asymmetrical ($p < 0.001$). The agreement level was found to be fair ($\kappa = 0.38$, $p < 0.001$).

Furthermore, the authors classified the SSC patterns into two distinct groups based on their potential clinical significance. The first group included dehiscent and papyraceous types, while the second group comprised normal, pneumatised, and thick types. Among those with normal/pneumatized/thick type SSC on the non-cleft side, 68.1% had the same type on the cleft side. Similarly, among those with dehiscent/papyraceous type SSC on the non-cleft side, 86.7% had the same type on the cleft side. The McNemar–Bowker test indicated a lack of symmetry in the SSC types between the cleft and non-cleft sides of CL/P patients ($p < 0.001$). Furthermore,

a fair level of agreement was observed ($\kappa=0.36, p < 0.001$) (Table 2).

There was a significant difference in terms of SSC types between the cleft side of CL/P (CS–CL/P) patients and the control group sides ($p < 0.001$). The frequency of dehiscence and papyraceous SSC patterns was found to be significantly higher on the CS–CL/P patients compared to the control sides. There was a slightly statistically significant difference in terms of SSC types between the non-cleft side of CL/P (NCS–CL/P) patients and the control group sides ($p=0.057$). Accordingly, pneumatised/thick types were significantly less common on the NCS–CL/P patients compared to the control sides (Table 3).

Dehiscence/papyraceous types were significantly higher, while normal/pneumatised /thick types were significantly less common on the CS–CL/P patients compared to the control group sides ($p < 0.001$). However, there was no statistically significant difference between the NCS–CL/P patients and control group sides ($p=0.384$) (Table 3).

When SSC thicknesses were compared between the groups, the SSC thickness on the CS–CL/P patients was thinner than the NCS–CL/P patients and the control group sides ($p=0.033$ and $p < 0.001$, respectively). In addition,

SSC thickness on the NCS–CL/P patients was also thinner than the control group sides ($p=0.001$) (Table 4).

There was no statistically significant difference observed in the SSC patterns between males and females between all groups ($p > 0.05$) (Table 5).

There was no significant relationship between age and SSC thickness in all groups (CS–CL/P patients: $p=0.551; r = -0.066$; NCS–CL/P patients: $p=0.255; r = -0.126$; control patients: $p=0.090; r=0.131$).

Discussion

Due to its complex anatomy, the middle ear is a challenging area for radiologists. The previous imaging studies of SSCD were mostly performed with CT [13–16]. Although CT provides valuable information about the complex anatomy of the middle ear, a few studies pointed out that it has some limitations [15, 17, 18]. Sequeira et al. highlighted that CT should not be taken as the only indicator for SSCD [15]. Bremke et al. compared CT and CBCT for the evaluation of SSCD. They concluded that CBCT had a higher potential to detect SSCD, especially in thin temporal bones [17]. Tavassolie et al. [16] and Mondina et al. [18] also stated that SSCD can be overestimated with CT imaging. Cloutier et al. emphasized that CT is not an appropriate screening tool for SSCD. They suggested that it should only be used to confirm a strong clinical suspicion [19]. Thabet et al. stated that they only used CT for the dehiscences which are larger than 2 mm [20]. In the present study, CBCT images were used to evaluate SSC thickness and patterns in patients with CL/P. Previous studies reported that the Pöschl plane has the diagnostic value for the evaluation of SSCD [17, 19]. A recent study by Duman & Doğan suggested that the Pöschl plane should be created for the evaluation of temporal bones [21]. The present study also used the Pöschl plane to evaluate the temporal bone.

Table 4 SSC thicknesses by groups

	CS–CL/P	NCS–CL/P	Control
SSC thicknesses			
Mean \pm std. dev	0.69 \pm 0.6	0.79 \pm 0.53	1.09 \pm 0.91
Median(min–max)	0.57 (0–2.88)	0.57 (0–2.83)	0.89 (0–5.44)
<i>p</i>	0.033 [†]	<0.001 [‡]	0.001 [§]

Data are shown as mean \pm standart deviation and median (minimum–maximum). [†] Wilcoxon signed rank test, [‡], [§] Mann–Whitney *U* test. * $p < 0.05$ is indicated in bold. Comparisons: [†] CS–CL/P and NCS–CL/P; [‡] CS–CL/P and control; [§] NCS–CL/P and control. CL/P: cleft lip and palate, CS–CL/P: cleft side of CL/P, NCS–CL/P: non-cleft side of CL/P, SSC: superior semicircular canal

Table 5 SSC types by gender

	CS–CL/P		NCS–CL/P		Control	
	Female	Male	Female	Male	Female	Male
<i>SSC types</i>						
Dehiscence pattern	6 (18.8)	11 (21.2)	2 (6.3)	9 (17.3)	4 (6.2)	10 (9.6)
Papyraceous pattern	6 (18.8)	12 (23.1)	-	4 (7.7)	4 (6.2)	5 (4.8)
Normal pattern	14 (43.8)	28 (53.8)	28 (87.5)	36 (69.2)	40 (62.5)	75 (72.1)
Pneum/thick pattern	6 (18.8)	1 (1.9)	2 (6.3)	3 (5.8)	16 (25)	14 (13.5)
Total	32 (100)	52 (100)	32 (100)	52 (100)	64 (100)	104 (100)
<i>p</i>	0.075 [†]		0.163 [†]		0.461 [‡]	

Data are shown as frequencies(column %). [†] Fisher’s exact test, [‡] Chi-square test. CL/P: cleft lip and palate, CS–CL/P: cleft side of CL/P, NCS–CL/P: non-cleft side of CL/P, Pneum: pneumatised, SSC: superior semicircular canal

Altun et al. evaluated 258 temporal bones using CBCT. They included both unilateral and bilateral CL/P cases in their study. They found that CL/P patients had a higher prevalence for SSCD [7]. A recent study by Paknahad et al. evaluated the SSC in unilateral CL/P, bilateral CL/P, and control groups. They found that the prevalence of SSCD was significantly higher in both the unilateral and bilateral CL/P groups compared to the control group [12]. In the present study, it was found that temporal bones on the cleft side had a higher prevalence for SSCD compared to both the non-cleft side and control group. Our findings are in line with the literature.

Cisneros et al. stated that the prevalence of SSCD was 1.84% in their CT study [13]. In a previous cadaver study, the prevalence of SSCD was reported as 8% [18]. Kurt et al. reported that the prevalence of SSCD was 6.28% in their CBCT study [22]. Akay et al. also evaluated the SSCD using CBCT, they reported the prevalence of SSCD as 16.5% [23]. Paknahad et al. reported the prevalence of SSCD as 6.8% in their healthy control group [12]. In the present study, the prevalence of SSCD was found as 8.3% in the control group. These differences can be attributed to the fact that the individuals included in the studies consisted of populations with various age, gender and ethnic distributions. Akay et al. [23] also highlighted that the various slice thicknesses used in the studies may affect the results. In the present study, the slice thickness was determined as 0.4 mm.

The present study reported a prevalence of 20.2% for SSCD in the CS–CL/P group and 13.1% for the NCS–CL/P group. This finding is consistent with the previous studies. Altun et al. also used CBCT to evaluate SSCD in patients with CL/P, and reported a prevalence of 18% [7]. Paknahad et al. [12] reported the SSCD prevalences as 17.6% and 36.8% for the unilateral and bilateral CL/P patients, respectively. However, it should be noted that both of the aforementioned studies evaluated the SSC patterns of the cleft side and non-cleft side together in unilateral CL/P patients. Minor differences observed between their findings and those of the present study may be attributable to differences in methodology.

Significant differences in SSC thickness were observed. The mean thickness of SSC for the CS–CLP group was found significantly lower than both NC–CLP and control groups. In addition, the mean thickness of SSC for the NC–CLP group was significantly lower than the control group. Our findings suggest that CL/P have a significant impact on the thickness of the SSC on both the cleft side and non-cleft side, with a greater effect observed on the cleft side.

In contrast to our findings, Paknahad et al. [12] found no significant differences in SSC patterns between the cleft side and the non-cleft side of unilateral CL/P patients. They reported significantly lower SSC thickness values for unilateral and bilateral CL/P patients compared to the controls,

similar to the presented study. However, they did not observe a significant difference in SSC thickness between the cleft side and the non-cleft side of unilateral CL/P patients. The inconsistency between the findings may be attributed to differences in sample size. Paknahad et al. [12] had a smaller number of unilateral CL/P patients compared to the current study, which could have influenced their results.

The effect of age on SSCD is controversial. Crovetto et al. evaluated the influence of age on SSCD [24]. They concluded that the bone layer covering the SSC thins slightly with age. They suggested that these thin papyraceous-type SSCs may cause dehiscence with advancing age. Nadgir et al. also advocated that SSCD is an acquired condition [25]. However, Mahulu et al. [26] and Akay et al. [23] found no significant difference between young and elder individuals for the SSC thickness. Our study population included individuals between 14 and 26 ages. It can be considered as a limitation of the study. The SSCD status of the elder patients with CL/P requires further investigation.

SSCD syndrome is characterized by auditory, vestibular and clinical symptoms which could be treated by surgical interventions [6]. A few studies suggested SSCs with papyraceous patterns may lead to dehiscence [13, 27–29]. Therefore, it is believed that, from a clinical standpoint, the dehiscence and papyraceous types are of primary importance [13, 27]. In the present study, the elevated prevalence of dehiscence and papyraceous types in the CS–CL/LP group compared to the control group implies that the presence of a cleft may be a predisposing factor for these types. Consequently, there is an increased necessity for radiological evaluations, especially in individuals with CL/P whom may be suspected to have thinner SSC, to proactively address these potential concerns.

The present study has some limitations. The patient population consists of relatively young individuals. The radiological pattern of elderly CLP patients should be investigated in further studies. Another limitation of this study is that it was conducted retrospectively, and therefore, there was no information available regarding any clinical complaints or symptoms experienced by the individuals included in the study. This information could have provided a more comprehensive understanding of the clinical implications of SSCD in CL/P patients.

Conclusions

The mean thickness of SSC was found significantly lower in CS–CL/P group compared to both NCS–CL/P group and control group. CL/P is considered to affect the osseous structures of the temporal bone. CS–CL/P group had a higher prevalence of SSCD and papyraceous compared to control group. Papyraceous type SSC is also considered to

be clinically important; therefore, its frequent occurrence within the CL/P group should not be overlooked. For this reason, CL/P patients should also be evaluated in terms of SSC radiological pattern. The effect of CL/P on the temporal bone structures should be investigated in further studies.

Author contributions HDY: Substantial contributions to the conception and design of the work; acquisition of data, statistical analysis and interpretation of the data; and approval of the final version of the manuscript. DŞÇ: Substantial contributions to the conception and design of the work; acquisition of data, and interpretation of data; drafting the manuscript; critical revision of the manuscript for important intellectual content. AC: Substantial contributions to the conception and design of the work and interpretation of the data; and approval of the final version of the manuscript.

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Data availability The datasets generated and analyzed during the current study are available from the corresponding author on reasonable request.

Declarations

Conflict of interest The authors have no competing interests to declare that are relevant to the content of this article.

Ethics approval The Ethical Committee of Cukurova University's Medical School approved the study (date: 2021, meeting no: 97, decision no: 116).

Informed consent The standard protocol at our department contains acquiring signed Informed Consent from all patients or their parents for evaluation of their CBCT records for any scientific reasons.

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