Bilateral cervical chondrocutaneous branchial remnants

A case report and a review of the literature

Han Shin Lee, MD^a, Tae Han Kim, MD^a, Jae Yool Jang, MD^a, Jung Woo Woo, MD^a, JinKwon Lee, MD^a, Sang Ho Jeong, MD^a, Eun Jung Jung, MD^a, Hyo Jung An, MD^b, Taejin Park, MD^{a,*}

Abstract

Rationale: Cervical chondrocutaneous branchial remnants are rare, benign, congenital anomalies, frequently seen bilaterally. **Patient concerns:** Here, we report the case of a 4-month-old female infant who presented with bilateral lower neck skin tag since birth.

Diagnosis and Interventions: The patient underwent mass excision. The final pathological diagnosis was bilateral cervical chondrocutaneous branchial remnants with hyaline cartilage.

Outcomes: No complications were observed after excision. One-year follow-up revealed no recurrence.

Lessons: Bilateral chondrocutaneous branchial remnants are rare anomalies. They are often associated with cardiac or genitourinary abnormalities. Therefore, additional preoperative imaging of the abdomen and heart are recommended.

Abbreviations: CCBR = congenital chondrocutaneous branchial remnants, SCM = sternocleidomastoid.

Keywords: branchial remnant, chondrocutaneous, neck, skin tag

1. Introduction

Cervical mass is a relatively common pathological condition in the neonatal period. Neonatal neck masses are mostly congenital malformations that occur during transformation into adult derivatives, so branching malformations often occur due to the persistence of the part of the branching device, which should generally disappear.^[1] First reported in 1858, benign neck tumors, formerly called "cervical skin tags," "accessory tragus," "wattle," and "cervical auricle," were retermed "chondrocutaneous branchial remnants (CCBR)" by Altan et al in 1997.^[2,3] Generally, CCBRs appear unilaterally or bilaterally. To date, 117

The authors have no funding and conflicts of interest to disclose.

Data sharing not applicable to this article as no datasets were generated or analyzed during the current study.

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

^a Department of Surgery, ^b Department of Pathology, Gyeongsang National University Changwon Hospital, Gyeongsang National University School of Medicine, Changwon, Korea.

^{*} Correspondence: Taejin Park, Department of Surgery, Gyeongsang National University Changwon Hospital, Gyeongsang National University School of Medicine, 11, Samjeongja-ro, Seongsan-Gu, Changwon 51472, Korea (e-mail: taejin6393@gmail.com).

Copyright © 2020 the Author(s). Published by Wolters Kluwer Health, Inc. This is an open access article distributed under the Creative Commons Attribution License 4.0 (CCBY), which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.

How to cite this article: Lee HS, Kim TH, Jang JY, Woo JW, Lee J, Jeong SH, Jung EJ, An HJ, Park T. Bilateral cervical chondrocutaneous branchial remnants: a case report and a review of the literature. Medicine 2020;99:28(e21114).

Received: 25 November 2019 / Received in final form: 19 May 2020 / Accepted: 4 June 2020

http://dx.doi.org/10.1097/MD.00000000021114

cases have been reported (34 with bilateral lesions) in medical literature.^[4] Bilateral CCBR are rare, and multiple differential diagnoses should be considered while diagnosing it according to the location. Herein, we present a case of bilateral CCBR presenting as a neonatal neck mass at the sternocleidomastoid (SCM) muscle level and investigated previous medical literatures about bilateral CCBR case reports (Table 1).

Medicine

2. Methods

Because this case report is not a prospective or retrospective study, the consent of the patient's parents was sufficient, and ethical approval was provided done by the IRB. Thus, we decided to publish only the age, image findings, and pathologic pictures in the case report, and we received written consent from the patient's parents.

3. Case report

3.1. Clinical summary

A 4-month-old girl was referred to our hospital because of bilateral neck skin lesions since birth. The skin lesions on the neck were covered with normal skin and each lesion measured 1 cm in length (Fig. 1). The lesions were located in the lower third of the neck, anterior to the SCM muscle. The lesions were stiff and elastic. There were no opening pits, discharge, or inflammatory changes. Physical exam was unremarkable. There were no cardiac or urogenital anomalies on ultrasonography. There was no family history of this condition. After obtaining the consent of the parents, surgical excision under general anesthesia was performed. Cartilaginous remnants extended to the fascia of the SCM muscles. There was no fistula tract to deep neck structures (Fig. 2). After mass excision, no recurrence or complications were seen at 1-year follow-up.

Editor: Maya Saranathan.

Table 1

Case reports of bilateral cervical chondrocutaneous branchial remnants.

NoAuthorSexAgeHeightLocationsize (cm)UepinPathologyanomalesManagementCountyYear1Lindgren ^{1[4]} F9 yr0SurgeryUSA19563Punyamuthy ¹⁷¹ M12 yrLover thirdAnt. to SCMNormal cartilage0SurgeryIndia19754Clarke ¹⁶¹ F-Lover thirdAnt. to SCM0.3-1.5Plastysma-0SurgeryUK19765F-Lover thirdAnt. to SCM0.3-1.5Plastysma-1SurgeryUSA19866F-Lover thirdAnt. to SCM0.3-1.5Plastysma-1SurgeryLUSA19868Doi at al. ^[19] 0SurgeryAustralia198690SurgeryKanea199710Atan et al ^[61] M7 moInf. thirdAnt. to SCM1.2SCMElastic0SurgeryRomana200011Kim et al ^[71] M25 yrSurgersteinalAnt. to SCM3.5 / 1.2-Halaic0SurgeryRomana200012Fraud et al ^[62] M4 mLover thirdAnt. to SCM1.0 / 1.5PlastysmaElastic0SurgeryLonead <t< th=""><th></th><th></th><th></th><th></th><th></th><th></th><th>e: ()</th><th></th><th></th><th>No. of associated </th><th></th><th><u> </u></th><th></th></t<>							e : ()			No. of associated 		<u> </u>	
1 Lindgren ^[16] F 9 yr - - - - - 0 Surgery USA 1956 2 Purganuthy ¹⁷¹ R 12 yr Lower third Ant to SCM - - Normal cartilage 0 Surgery Ivia 1972 4 Clarke ¹⁶¹ F - Lower third Ant to SCM 0.3-1.5 Plastysma - 0 Surgery Ivia 1972 6 - F 66 yr Lower third Ant to SCM 0.3-1.5 Plastysma - 1 Surgery UX 1986 8 Doi at al. ^[10] - - - - - - 0 Surgery Quardental 1986 9 - - - - - - - 0 Surgery Canada 1997 11 Kint et al ^[21] M 4 Tro Int to SCM 3.5 / 1.2 - Hyaline Surgery	NO	Author	Sex	Age	Height	Location	Size (cm)	Depth	Pathology	anomalies	Management	Country	Year
2 - - - - - - Normal cartiage 0 Surgery India 1972 3 Punyamuthy ¹⁷¹ M 12 yr Lower third Ant. to SCM - - Normal cartiage 0 Surgery India 1972 5 F - Lower third Ant. to SCM 0.3-1.5 Plastysma - 0 Surgery UK 1976 7 Spering ^[0] F 13 yr Lower third Ant. to SCM 0.5 / 0.5 Plastysma - 0 Surgery Australia 1986 8 Doi at al. ^[19] - - - - - - 0 Surgery Australia 1986 8 Doi at al. ^[19] M 7.mo Inf. third Ant. to SCM 1.2 SCM Eastic 0 Surgery Karaa 1997 11 Kim et al ^[20] M 2.5 yr Surgarey Lower third Ant. to SCM 3.5 / 1.2 - Hyaline 2 Surgery Garaa 2002 <td< td=""><td>1</td><td>Lindgren^[16]</td><td>F</td><td>9 yr</td><td>_</td><td>_</td><td>-</td><td></td><td></td><td>0</td><td>Surgery</td><td>USA</td><td>1956</td></td<>	1	Lindgren ^[16]	F	9 yr	_	_	-			0	Surgery	USA	1956
3 Puryanuthp ⁽¹⁷⁾ M 12 yr Lower third Ant. to SCM - - Morral cartilage 0 Surgery India 1977 4 Clarke ¹¹⁶ F - Lower third Ant. to SCM 0.3-1.5 Plastysma - 0 Surgery UK 1976 6 - F 66 yr Lower third Ant. to SCM 0.3-1.5 Plastysma - 1 Surgery USA 1986 7 Spering ¹⁰ F 13 yr Lower third Ant. to SCM 0.5 Plastysma - 1 Surgery USA 1986 9 - - - - - - - 0 Surgery Los T Plastysma - - 0 Surgery Los T Surgery Los T Surgery Los T Surgery Lower T Surgery Los T <	2		-	-	-	-	_			0	Surgery		
4 Clarkel ¹⁸ F - Lower third Ant. to SCM 0.3-1.5 Plastysma - 0 Surgery UK 1976 5 F 6 F 66 yr Lower third Ant. to SCM 0.3-1.5 Plastysma - 0 Surgery USA 1986 8 Doi at al. ¹¹⁰ - - - - - - 0 Surgery USA 1986 9 - - - - - - - 0 Surgery Caaad 1997 10 Attan et al ^{[61} M 7 mo Inf. third Ant. to SCM 2.5 - Elastic 0 Surgery Caaad 1997 12 Braun et al ^{[61} M 4 mo Lower third Ant. to SCM 2.5 - Elastic 0 Surgery Kaata 2006 13 Fuad et al ^[20] M 4 mo Lower third Ant. to SCM 1.0 / 1.5 Plast	3	Punyamurthy ^[17]	Μ	12 yr	Lower third	Ant. to SCM	-	-	Normal cartilage	0	Surgery	India	1972
5 F - Lower third Ant. to SCM $0.3-1.5$ Plashyma - 0 Surgery User string 6 - F 66 yr Lower third Ant. to SCM $0.3-1.5$ Plashyma - 1 Surgery USA 1986 8 Doi at al. ^[19] - - - - - - 0 Surgery Australia 1986 9 - - - - - - 0 Surgery Australia 1987 10 Atta et al ^[61] M 7 mo Inf. third Ant. to SCM 1.0 - Elastic 0 Surgery Korea 1997 12 Braun et al ^[62] M 4 yr Ower third Ant. to SCM 3.5 / 1.2 - Hyaline 0 Surgery Boarda 2002 14 Coras et al ^[61] M 4 yr Inf. to SCM 1.0 / 1.5 Plastysma Elastic 0 Surgery <	4	Clarke ^[18]	F	-	Lower third	Ant. to SCM	0.3-1.5	Plastysma	-	0	Surgery	UK	1976
6 F 66 for for lower third Ant. to SCM $0.3 - 1.5$ Plastysma - 1 Surgery USA 1986 7 Spering ^[6] F 13 yr Lower third Ant. to SCM $0.5 / 0.5$ Plastysma Hyaline 0 Surgery USA 1986 8 Doi at $A^{[19]}$ - - - - - 0 Surgery Australia 1986 9 - - - - - - - 0 Surgery Australia 1986 10 Attan et al ^{[71} M 7 mo Inf. third Ant. to SCM 1.0 - Elastic 0 Surgery Koraa 1997 12 Braun et al ^{[61} M 4 mo Lower third Ant. to SCM 2.5 - Elastic 0 Surgery Australia 1986 14 Coras et al ^{[211} - 4 yr Inf. third Ant. to SCM 1.0 / 1.5 Plastit Surgery	5		F	-	Lower third	Ant. to SCM	0.3-1.5	Plastysma	-	0	Surgery		
7 Spering ^[9] F 13 yr Lower third Ant to SCM $0.5 / 0.5$ Plastysma Hyaline 0 Surgery USA 1986 8 Doi at al. ^[19] - - - - - - 0 Surgery Australia 1986 9 - - - - - - 0 Surgery Canada 1997 10 Attan et al ^[21] M 7 mo Inf. third Ant. to SCM 1.0 - Elastic 0 Surgery Canada 1997 11 Kim et al ^[71] M 22 yr Lower third Ant. to SCM 2.5 - Elastic 0 Surgery Busita 2003 12 Graun et al ^[221] M 4 yr - <	6		F	66 yr	Lower third	Ant. to SCM	0.3-1.5	Plastysma	-	1	Surgery		
8 Doi at al. ^[19] - - - - - - - 0 Surgery Australia 1986 9 - - - - - - - 0 Surgery Canada 1997 10 Atla et al ^[2] M 7 mo Int. third Ant. to SCM 1.0 - Elastic 0 Surgery Canada 1997 11 Kim et al ^[6] M 4 mo Lower third Ant. to SCM 2.5 - Elastic 0 Surgery Australia 2003 12 Braun et al ^{[60} M 2.9 Lower third Ant. to SCM 3.5 / 1.2 - Hyaline 2 Surgery Basina 2003 14 Coras et al ^[21] - 4 yr - - - - Elastic 0 Surgery Turkey 2006 15 Ozturk et al ^[22] M 4 yr - - 1.7 - Elastic 0 Surgery India 2002 16 Giboa et al ^[24] <	7	Sperling ^[9]	F	13 yr	Lower third	Ant. to SCM	0.5 / 0.5	Plastysma	Hyaline	0	Surgery	USA	1986
9 -	8	Doi at al. ^[19]	-	_	_	_	_	_	_	0	Surgery	Australia	1988
10 Atlan et al ^[7] M 7 mo Inf. third Ant. to SCM 1.2 SCM Elastic 0 Surgery Canada 1997 11 Kim et al ^[7] M 25 yr Surgarstemal Ant. to SCM 1.0 - Elastic 0 Surgery Korea 1997 12 Braun et al ^[20] M 4 mo Lower third Ant. to SCM 2.5 - Elastic 0 Surgery Basina 2003 13 Fuad et al ^[20] M 2 yr Lower third Ant. to SCM 3.5 / 1.2 - Hyaline 2 Surgery Basina 2003 14 Coras et al ^[21] - 4 yr - - - - Elastic 0 Surgery Basina 2007 15 Ozturk et al ^[26] M 4 mo Lower third Ant. to SCM 1.0 / 1.5 SCM Elastic 0 Surgery India 2007 16 Giboa et al ^[26] M 4 mo Lower third Ant. to SCM 1.0 / 1.3 - Hyaline Surger	9		-	-	-	_	_	_	-	0	Surgery		
11Kim et alM25 yrSuprasternal SuprasternalAnt. to SCM1.0-Elastic0SurgeryKorea199712Braun et alM4 moLower thirdAnt. to SCM2.5-Elastic0SurgeryAustria200313Fuad et alM22 yrLower thirdAnt. to SCM3.5 / 1.2-Hyaline2SurgeryBosnia200314Coras et alM4 yr0.5 / 0.5-Elastic0SurgeryGermany200615Ozturk et alM4 yrInf. thirdAnt. to SCM1.0 / 1.5PlastysmaElastic0SurgeryTurkey200616Gilboa et alSM4 moLower thirdAnt. to SCM1.0 / 1.5SCMElastic0SurgeryLebanon200717Barahe et alFPrenatal1.7-Hastic0SurgeryLebanon200718Dayal De talDesiM4 moLower thirdAnt. to SCM1.0 / 1.3-Hyaline0SurgeryIabano200719Asanina et alF1 moLower thirdAnt. to SCM2.5 / 2.0SCMElastic0SurgeryIabano200720Tarrier alM4 yr0.70SurgeryIabano201121Nasser et alM <td>10</td> <td>Atlan et al^[2]</td> <td>М</td> <td>7 mo</td> <td>Inf. third</td> <td>Ant. to SCM</td> <td>1.2</td> <td>SCM</td> <td>Elastic</td> <td>0</td> <td>Surgery</td> <td>Canada</td> <td>1997</td>	10	Atlan et al ^[2]	М	7 mo	Inf. third	Ant. to SCM	1.2	SCM	Elastic	0	Surgery	Canada	1997
12Braun et alBraun et alM4 moLower thirdAnt. to SCM2.5-Elastic0SurgeryAustria200313Fuad et alM22 yrLower thirdAnt. to SCM3.5 / 1.2-Hyaline2SurgeryBosnia200314Coras et alM4 yr0.5 / 0.5-Elastic0SurgeryGermany200515Ozturk et alM4 yrInf. thirdAnt. to SCM1.0 / 1.5PlastysmaElastic0SurgeryTurkey200716Gilboa et alM1 yr2SurgeryLebanon200717Rameh et alM1 yr2SurgeryLebanon200718Dayal D et alM4 moLower thirdAnt. to SCM1.0 / 1.5SCMElastic0SurgeryLebanon200718Dayal D et alF1 4 dMid third-0.5 / 0.70SurgeryLebanon200720Tamir et alF5 yrLower thirdAnt. to SCM2.5 / 2.0SCMElastic0SurgeryLebanon201721Nasser et alF1 moLower thirdAnt. to SCM2.0 / 1.2SCMElastic0SurgeryHann201222Pham et alF <td>11</td> <td>Kim et al^[7]</td> <td>М</td> <td>25 yr</td> <td>Suprasternal</td> <td>Ant. to SCM</td> <td>1.0</td> <td>_</td> <td>Elastic</td> <td>0</td> <td>Surgery</td> <td>Korea</td> <td>1997</td>	11	Kim et al ^[7]	М	25 yr	Suprasternal	Ant. to SCM	1.0	_	Elastic	0	Surgery	Korea	1997
13 Fuad et al ^[20] M 22 yr Lower third Ant. to SCM $3.5 / 1.2$ - Hyaline 2 Surgery Bosnia 2003 14 Coras et al ^[21] - 4 yr - - 0.5 / 0.5 - Elastic 0 Surgery Germany 2005 15 Ozturk et al ^[23] M 4 yr - - - - - 2 Surgery Burgery Iurkey 2007 16 Gilboa et al ^[23] M 4 yr - - - - - 2 Surgery Iurkey 2007 17 Rameh et al ^[26] M 1 yr - - 1.7 - Elastic 0 Surgery Iurkey 2007 18 Dayal D et al ^[26] M 4 mo Lower third Ant. to SCM 1.0 / 1.3 - + Hyaline 0 Surgery Iurkey 2002 19 Asabina et al ^[27] M 4 yr - - 0.7 - - 0 Surgery Iurkey	12	Braun et al ^[6]	М	4 mo	Lower third	Ant. to SCM	2.5	_	Elastic	0	Surgery	Austria	2003
14 Coras et al ^[21] - 4 yr - - 0.5 / 0.5 - Elastic 0 Surgery Germany 2005 15 Ozturk et al ^[22] M 4 yr Inf. third Ant. to SCM 1.0 / 1.5 Plastysma Elastic 0 Surgery Turkey 2006 16 Gilboa et al ^[23] F Prenatal - - - - - 2 Surgery turkey 2006 17 Rameh et al ^[24] M 1 yr - - 1.7 - Elastic 0 Surgery Lebanon 2007 18 Dayal D et al ^[25] M 4 mo Lower third Ant. to SCM 1.0 / 1.3 - Hyaline 0 Surgery Japan 2006 19 Asahina et al ^[26] F 14 d Mid third - 0.5 / 0.7 - - 0 Surgery Japan 2006 20 Tamir et al ^[14] F 5 yr Lower third Ant. to SCM 2.0 / 1.2 SCM Elastic 0 Surgery Japan	13	Fuad et al ^[20]	Μ	22 yr	Lower third	Ant. to SCM	3.5 / 1.2	_	Hyaline	2	Surgery	Bosnia	2003
15 Ozturk et al ^[22] M 4 yr Inf. third Ant. to SCM 1.0 / 1.5 Plastysma Elastic 0 Surgery Turkey 2006 16 Gilboa et al ^[23] F Prenatal - - - - - - 2 Surgery Israel 2007 17 Rameh et al ^[24] M 1 yr - - 1.7 - Elastic 0 Surgery Lebanon 2007 18 Dayal D et al ^[26] F 14 d Mid third - 0.5 / 0.7 - - 0 Surgery Japan 2006 20 Tamir et al ^[14] F 5 yr Lower third Ant. to SCM 1.0 / 1.3 - Hyaline 0 Surgery Japan 2006 21 Nasser et al ^[4] F 1 mo Lower third Ant. to SCM 2.0 / 1.2 SCM Elastic 0 Surgery Japan 2012 22 Nasser et al ^[14] F 10 yr Halfway Ant. to SCM 1.2 / 1.2 SCM Elastic 0 <td>14</td> <td>Coras et al^[21]</td> <td>-</td> <td>4 yr</td> <td>_</td> <td>_</td> <td>0.5 / 0.5</td> <td>-</td> <td>Elastic</td> <td>0</td> <td>Surgery</td> <td>Germany</td> <td>2005</td>	14	Coras et al ^[21]	-	4 yr	_	_	0.5 / 0.5	-	Elastic	0	Surgery	Germany	2005
16 Gilboa et al ^[23] F Prenatal - - - - - - 2 Surgery at 6m Israel 2007 17 Rameh et al ^[24] M 1 yr - - 1.7 - Elastic 4 Surgery Lebanon 2007 18 Dayal D et al ^[25] M 4 mo Lower third Ant. to SCM 1.0 / 1.5 SCM Elastic 0 Surgery India 2006 19 Asahina et al ^[26] F 14 d Mid third - 0.5 / 0.7 - - 0 Surgery Lebanon 2007 20 Tamir et al ^[104] F 5 yr Lower third Ant. to SCM 2.5 / 2.0 SCM Elastic 0 Surgery Lebanon 2007 21 Nasser et al ^[41] F 1 mo Lower third Ant. to SCM 2.0 / 1.2 SCM Elastic 0 Surgery Japan 2012 22 Oiso et al ^[27] M 4 yr - - - - - - Surgery	15	Ozturk et al ^[22]	М	4 yr	Inf. third	Ant. to SCM	1.0 / 1.5	Plastysma	Elastic	0	Surgery	Turkey	2006
17 Rame et al ^[24] M 1 yr - - 1.7 - Elastic 4 Surgery Lebanon 2007 18 Dayal D et al ^[25] M 4 mo Lower third Ant. to SCM 1.0 / 1.5 SCM Elastic 0 Surgery India 2006 19 Asahina et al ^[26] F 14 d Mid third - 0.5 / 0.7 - - 0 Surgery Japan 2006 20 Tamir et al ^[14] F 5 yr Lower third Ant. to SCM 1.0 / 1.3 - Hyaline 0 Surgery Lebanon 2017 21 Nasser et al ^[41] F 1 mo Lower third Ant. to SCM 2.0 / 1.2 SCM Elastic 0 Surgery Lebanon 2017 22 Oiso et al ^[27] M 4 yr - - 0.7 - - 0 Surgery Lebanon 2012 23 Pham et al ^[101] F 10 yr Halfway Ant. to SCM 2.0 / 1.2 SCM Elastic 0 Surgery <td>16</td> <td>Gilboa et al^[23]</td> <td>F</td> <td>Prenatal</td> <td>_</td> <td>_</td> <td>_</td> <td>-</td> <td>_</td> <td>2</td> <td>Surgery at 6m</td> <td>Israel</td> <td>2007</td>	16	Gilboa et al ^[23]	F	Prenatal	_	_	_	-	_	2	Surgery at 6m	Israel	2007
18 Dayal D et al ^[25] M 4 mo Lower third Ant. to SCM $1.0/1.5$ SCM Elastic 0 Surgery India 2006 19 Asahina et al ^[26] F 14 d Mid third - $0.5/0.7$ - - 0 Surgery Japan 2006 20 Tamir et al ^[14] F 5 yr Lower third Ant. to SCM $1.0/1.3$ - Hyaline 0 Surgery Israel 2006 21 Nasser et al ^[4] F 1 mo Lower third Ant. to SCM $2.5/2.0$ SCM Elastic 0 Surgery Lebanon 2017 22 Oiso et al ^[27] M 4 yr - - 0.7 - - 0 Surgery France 2013 23 Pham et al ^[111] F 10 yr Halfway Ant. to SCM $2.0/1.2$ SCM Elastic 0 Surgery France 2013 24 F - - - - - - Surgery Morcco 2014	17	Rameh et al ^[24]	М	1 yr	_	-	1.7	_	Elastic	4	Surgery	Lebanon	2007
19Asahina et alAsahina et al I^{261} F14 dMid third- $0.5/0.7$ 0SurgeryJapan200620Tamir et al I^{141} F5 yrLower thirdAnt. to SCM $1.0/1.3$ -Hyaline0SurgeryIsrael200621Nasser et alF1 moLower thirdAnt. to SCM $2.5/2.0$ SCMElastic0SurgeryLebanon201122Oiso et al I^{271} M4 yr0.70SurgeryJapan201223Pham et alf1 0 yrHalfwayAnt. to SCM $2.0/1.2$ SCMElastic0SurgeryFrance201324FSCMElastic0SurgeryFrance201324FSCMElastic0SurgeryMorocco201324FSCMElastic0SurgeryMorocco201325FSCMElastic0SurgeryMorocco201426Hemmaoui et al I^{120} M3 yrLower thirdAnt. to SCM $1.2/1.2$ SCMElastic0Surgery <t< td=""><td>18</td><td>Dayal D et al^[25]</td><td>Μ</td><td>4 mo</td><td>Lower third</td><td>Ant. to SCM</td><td>1.0 / 1.5</td><td>SCM</td><td>Elastic</td><td>0</td><td>Surgery</td><td>India</td><td>2008</td></t<>	18	Dayal D et al ^[25]	Μ	4 mo	Lower third	Ant. to SCM	1.0 / 1.5	SCM	Elastic	0	Surgery	India	2008
20Tamir et alImage: Second sec	19	Asahina et al ^[26]	F	14 d	Mid third	_	0.5 / 0.7	_	_	0	Surgery	Japan	2008
21Nasser et alII<	20	Tamir et al ^[14]	F	5 yr	Lower third	Ant. to SCM	1.0 / 1.3	_	Hyaline	0	Surgery	Israel	2008
22 Oiso et al $[27]$ M 4 yr - - 0.7 - - 0 Surgery Japan 2012 23 Pham et al F 10 yr Halfway Ant. to SCM 2.0 / 1.2 SCM Elastic 0 Surgery France 2013 24 F - - - - - - - Surgery France 2013 24 F - - - - - - - Surgery France 2013 25 F - - - - - - - Surgery Morocco 2014 26 Hemmaoui et al I ^[28] M 3 yr Lower third Ant. to SCM 1.2 / 1.2 SCM Elastic 0 Surgery Morocco 2014 27 Begovic et al M 8 mo - - - - Elastic 0 Surgery Serbia 2014 28 M 8 mo - - -	21	Nasser et al ^[4]	F	1 mo	Lower third	Ant. to SCM	2.5 / 2.0	SCM	Elastic	0	Surgery	Lebanon	2011
23Pham et alF10 yrHalfwayAnt. to SCM $2.0 / 1.2$ SCMElastic0SurgeryFrance 2013 24FSurgeryFrance 2013 25FSurgeryFrance 2013 26Hemmaoui et alISyrLower thirdAnt. to SCM $1.2 / 1.2$ SCMElastic0SurgeryMorocco 2013 27Begovic et alF7 moElastic1SurgerySerbia 2014 28M8 moElastic0SurgerySerbia 2014 28M7 yrElastic0SurgerySerbia 2014 29M7 yrNot operateSurgery $1.2 / 1.2$ ScMElastic0Surgery $1.4 / 1.2$ 20M7 yrNot operate $1.2 / 1.2$ ScMElastic0Surgery $1.4 / 1.2$ 20M7 yrNot operate $1.3 / 1.2$ $1.2 / 1.2$ ScMElastic0SurgeryIndia 2014 32Klockars et alInfant </td <td>22</td> <td>Oiso et al^[27]</td> <td>М</td> <td>4 vr</td> <td>_</td> <td>_</td> <td>0.7</td> <td>_</td> <td>_</td> <td>0</td> <td>Surgery</td> <td>Japan</td> <td>2012</td>	22	Oiso et al ^[27]	М	4 vr	_	_	0.7	_	_	0	Surgery	Japan	2012
24 F - - - - - - - Surgery 25 F - - - - - - - Surgery 26 Hemmaoui et al ^[28] M 3 yr Lower third Ant. to SCM $1.2 / 1.2$ SCM Elastic 0 Surgery Morocco 2013 27 Begovic et al ^[3] F 7 mo - - - - Elastic 1 Surgery Serbia 2014 28 M 8 mo - - - - Elastic 0 Surgery Serbia 2014 28 M 8 mo - - - - Elastic 0 Surgery Serbia 2014 29 M 7 yr - - - - - Not operate - - - Not operate - - - - Not operate - - - - - - - - - - - - <td>23</td> <td>Pham et al^[11]</td> <td>F</td> <td>10 yr</td> <td>Halfway</td> <td>Ant. to SCM</td> <td>2.0 / 1.2</td> <td>SCM</td> <td>Elastic</td> <td>0</td> <td>Surgery</td> <td>France</td> <td>2013</td>	23	Pham et al ^[11]	F	10 yr	Halfway	Ant. to SCM	2.0 / 1.2	SCM	Elastic	0	Surgery	France	2013
25FSurgery26Hemmaoui et alM3 yrLower thirdAnt. to SCM $1.2 / 1.2$ SCMElastic0SurgeryMorocco201327Begovic et alF7 moElastic1SurgerySerbia201428M8 moElastic0SurgerySerbia201429M7 yrElastic1SurgerySerbia201430MNot operate31Chander et alM6 yr-0.6 / 0.5-Elastic0SurgeryIndia201432Klockars et alInfantNot operateFinland201533FInfant334Kono et alF6 moLower thirdAnt. to SCM1.3 / 1.0SCMElastic0SurgeryJapan201535Nielsen et alM5 yrNot operatePlotter201636Nielsen et alM5 yr36Nielsen et alM	24		F	_	_ `	_	_	_	_	_	Surgery		
26Hermmaoui et alImage: all stress of the stress of	25		F	_	_	_	_	_	_	_	Surgery		
27Begovic et alBegovic et al <td>26</td> <td>Hemmaoui et al^[28]</td> <td>М</td> <td>3 vr</td> <td>Lower third</td> <td>Ant. to SCM</td> <td>1.2 / 1.2</td> <td>SCM</td> <td>Elastic</td> <td>0</td> <td>Surgery</td> <td>Morocco</td> <td>2013</td>	26	Hemmaoui et al ^[28]	М	3 vr	Lower third	Ant. to SCM	1.2 / 1.2	SCM	Elastic	0	Surgery	Morocco	2013
28M8 moElastic0Surgery29M7 yrElastic1Surgery30MNot operate31Chander et al ^[10] M6 yr0.6 / 0.5-Elastic0SurgeryIndia201432Klockars et al ^[15] MInfant0Not operateFinland201533FInfant334Kono et al ^[29] F6 moLower thirdAnt. to SCM1.3 / 1.0SCMElastic0SurgeryJapan201535Nielsen et al ^[13] M5 yrHyaline0SurgeryDemark201636Otest et al ^[13] M5 yrHyaline0SurgeryDemark2016	27	Begovic et al ^[3]	F	7 mo	_	_	_	_	Elastic	1	Surgery	Serbia	2014
29M7 yrElastic1Surgery30MNot operate31Chander et al ^[10] M6 yr $0.6 / 0.5$ -Elastic0SurgeryIndia201432Klockars et al ^[15] MInfant0Not operateFinland201533FInfant0Not operateFinland201533FInfant334Kono et al ^[29] F6 moLower thirdAnt. to SCM1.3 / 1.0SCMElastic0SurgeryJapan201535Nielsen et al ^[12] M5 yrHyaline0SurgeryDemark201636Circut et al ^[13] NIsfeetLower thirdAnt. to SCM35Nielsen et al ^[13] M5 yr	28	- 3	М	8 mo	_	_	_	_	Elastic	0	Surgery		
30MNot operate31Chander et alM6 yr0.6 / 0.5-Elastic0SurgeryIndia201432Klockars et alMInfant0Not operateFinland201533FInfant3-34Kono et alF6 moLower thirdAnt. to SCM1.3 / 1.0SCMElastic0SurgeryJapan201535Nielsen et alM5 yrHyaline0SurgeryDenmark201636Offer et alM5 yrHyaline0SurgeryDenmark201637Nielsen et alM5 yrHyaline0SurgeryDenmark201638Nielsen et alM5 yrHyaline0SurgeryDenmark201639Offer et alM5 yr <t< td=""><td>29</td><td></td><td>М</td><td>7 vr</td><td>_</td><td>_</td><td>_</td><td>_</td><td>Elastic</td><td>1</td><td>Surgery</td><td></td><td></td></t<>	29		М	7 vr	_	_	_	_	Elastic	1	Surgery		
31Chander et alM6 yr $0.6 / 0.5 -$ Elastic0SurgeryIndia201432Klockars et alMInfant0Not operateFinland201533FInfant33-34Kono et alF6 moLower thirdAnt. to SCM1.3 / 1.0SCMElastic0SurgeryJapan201535Nielsen et alM5 yr435Nielsen et alM5 yrHyaline0SurgeryDenmark201636OSurgeryLower thirdAnt. to SCM1.3 / 1.0SCMElastic0SurgeryDenmark201637Nielsen et alM5 yr38Nielsen et alM5 yr	30		М	_	_	_	_	_	_	_	Not operate		
32Klockars et al M Infant $ 0$ Not operateFinland201533FInfant $ 3$ $ 3$ $-$ 34Kono et alF6 moLower thirdAnt. to SCM1.3 / 1.0SCMElastic0SurgeryJapan201535Nielsen et alM5 yr $ -$ Hyaline0SurgeryDenmark201636Oliver et alM5 yr $ -$ Hyaline0SurgeryDenmark2016	31	Chander et al ^[10]	М	6 vr			0.6 / 0.5	_	Elastic	0	Surgery	India	2014
$\begin{array}{cccccccccccccccccccccccccccccccccccc$	32	Klockars et al ^[15]	М	Infant	_	_	_	_	_	0	Not operate	Finland	2015
34Kono et al E 6 moLower thirdAnt. to SCM1.3 / 1.0SCMElastic0SurgeryJapan201535Nielsen et alM5 yrHyaline0SurgeryDenmark201536Oliver et alM5 yrHyaline0SurgeryDenmark201536Oliver et alM5 yr	33		F	Infant	_	_	_	_	_	3	_		
35 Nielsen et al ^[12] M 5 yr $ -$ Hyaline O Surgery Denmark 2016	34	Kono et al ^[29]	F	6 mo	Lower third	Ant. to SCM	1.3/1.0	SCM	Flastic	0	Surgery	Japan	2015
	35	Nielsen et al ^[12]	M	5 vr	_	_	_	_	Hvaline	0	Surgery	Denmark	2016
36 Glant et al ^{ers} – Intant Lower third Ant, to SUM – SUM Flastic 2 Surgery USA 2018	36	Giant et al ^[13]	_	Infant	Lower third	Ant. to SCM	_	SCM	Flastic	2	Surgery	USA	2018
37 Lee et al ^[this case] F 4 mo Lower third Ant. to SCM 1.0 / 1.0 SCM Hyaline 0 Surgery Korea 2019	37	Lee et al [this case]	F	4 mo	Lower third	Ant. to SCM	1.0 / 1.0	SCM	Hyaline	0	Surgery	Korea	2019

SCM = sternocleidomastoid.

3.2. Pathological findings

Histologic examination showed hyaline cartilage cores covered by normal skin consisting of epidermis and dermis with subcutaneous fat compatible with CCBR (Fig. 3).

4. Discussion

Neonatal tumors in cervical area are referred when tumors present before the 28th day of life.^[5] Tumor located in a newborn's neck includes, differential diagnosis of teratoma, embryoma, hamartoma, dermoid cyst and choristoma.^[6] CCBRs are choristomas of the cervical area, and 2 suggestions have been proposed for the embryologic source, although these are yet to be verified. One theory suggests that they arise from ectopic auricular tissue.^[7] The other suggests that CCBRs originated from the branchial tissues contributing to the formation of most cervical tissues.^[8,9] The core of CCBR is either elastic cartilage or hyaline cartilage. The presence of elastic cartilage may suggest an auricular origin from the first or second branchial arch, whereas the presence of hyaline cartilage excludes an auricular origin and

suggests a cervical origin from the second or lower branchial arches.^[9] The widely accepted theory of origin is that CCBRs are the result of incomplete obliteration of the branchial apparatus, leaving cells behind in the neck during embryonic migration that differentiate into cartilage. Others suggest that it is rather the presence of pluripotent cell rests, much like the presence of supernumerary nipples, which proliferate into cartilage.^[10] These lesions are similar or analogous to preauricular tags, but are located in the lower neck. Most lesions present unilaterally; bilateral lesions as seen in our case are extremely rare.^[3,11]

In 1997 Altan et al described CCBR as follows:

- (1) predominance in male (11 of 17);
- (2) high incidence of associated anomalies (76%) involving the auditory (neurosensory deafness, serous otitis media, and malformation of the external ear), respiratory (tracheomalacia), oro-gastrointestinal (cleft palate, oronasal reflux and inguinal hernia), genitourinary (hydronephrosis), cardiovas-cular (atrial septal defect), musculoskeletal and visual systems;
 (3) presence of a cartilage core;



Figure 1. A 4-month-old girl gross findings of bilateral chondrocutaneous branchial remnants (CCBR).

- (4) a scarcity of bilateral lesions (1 of 17);
- (5) located in the middle or lower third of the neck; and
- (6) increased prevalence anterior to the SCM muscle.^[2]



Figure 2. Intraoperative picture of bilateral chondrocutaneous branchial remnants.

CCBR can have either rod-shaped elastic or hyaline cartilage core surrounded by normal skin and subcutaneous tissues. They are located in the middle or lower third of the neck, anterior to or over the SCM muscle. The lesion presents at birth and has no or very slow growth. The lesion has no connection with deep structures but adherence to the fascia of the SCM muscle is often reported. There is no report of underlying sinuses and cysts.^[12] Ultrasonography can be useful for describing the lesions, which have the characteristic presence of a tubular cartilage that extends



Figure 3. Gross finding and histopathologic findings of excised bilateral chondrocutaneous branchial remnants. (a) The gross finding of excised and cut in half of bilateral chondrocutaneous branchial remnants. The whitish glistening cartilaginous mass is covered by normal skin tissue. (b) On microscope, a polypoid skin lesion with underlying subcutaneous tissue and hyaline cartilage. (c) The overlying skin is composed of many hair follicles, dermal collagen, and adipose tissue. (d) On higher magnification, extracellular matrix of hyaline cartilage and evenly distributed bland-looking chondrocytes.

to the SCM muscle. CCBR is often associated with cardiac or genitourinary abnormalities, which have been reported in 11% to 76% of cases.^[13] Therefore, preoperative additional imaging studies of the abdomen and heart are recommended.

In our study, we examined the previous medical literatures and inferred that there was no correlation between anomalies on unilateral CCBR and bilateral CCBR (Table 1).

As treatment for CCBR and to obtain an accurate histologic diagnosis, complete surgical excision is recommended. If the patient has problems with tolerating anesthesia, the excision can be postponed.

Tarmir et al suggested surgical treatment of CCBR just before starting school, which allows minimization of surgical complications; however, it can be postponed to a suitable and safe age.^[14,15]

5. Conclusions

Bilateral CCBR is a rare condition. CCBR should be included in the differential diagnosis for congenital neck lesions in pediatric patients. The treatment of choice of CCBR is complete surgical excision. Further, careful preoperative assessments are needed for investigating associated lethal anomalies by abdominal ultrasound and cardiac examination.

Author contributions

Conceptualization: Taejin Park, Han Shin Lee.

Data curation: Taejin Park, Hyo Jung An, Han Shin Lee.

Formal analysis: Han Shin Lee.

Investigation: Tae Han Kim.

Methodology: Jae Yool Jang, Jung Woo Woo.

Project administration: JinKwon Lee.

Resources: Sang Ho Jeoung.

Supervision: Eun Jung Jung.

Validation: Han Shin Lee.

Visualization: Han Shin Lee.

Writing – original draft: Han Shin Lee.

Writing – review & editing: Taejin Park.

References

- Moore KL, Persaud TVN, Torchia MG. The Developing Human-E-Book: Clinically Oriented Embryology. Saunders, Philadelphia, USA: Elsevier Health Sciences; 2018.
- [2] Atlan G, Egerszegi EP, Brochu P, et al. Cervical chrondrocutaneous branchial remnants. Plast Reconstr Surg 1997;100:32–9.
- [3] Begovic N, Simic R, Vlahovic A, et al. Cervical chondrocutaneous branchial remnants – report of 17 cases. Int J Pediatr Otorhinolaryngol 2014;78:1961–4.
- [4] Nasser HA, Iskandarani F, Berjaoui T, et al. A case report of bilateral cervical chondrocutaneous remnants with review of the literature. J Pediatr Surg 2011;46:998–1000.

- [5] Halperin EC. Neonatal neoplasms. Int J Radiat Oncol Biol Phys 2000;47:171–8.
- [6] Braun H, Hofmann T, Wolfgruber H, et al. Case report of bilateral cervical chondrocutaneous branchial remnants. Int J Pediatr Otorhinolaryngol 2003;67:89–92.
- [7] Kim SW, Moon SE, Kim JA. Bilateral accessory tragi on the suprasternal region. J Dermatol 1997;24:543–5.
- [8] Hogan D, Wilkinson RD, Williams A. Congenital anomalies of the head and neck. Int J Dermatol 1980;19:479–86.
- [9] Sperling LC. Congenital cartilaginous rests of the neck. Int J Dermatol 1986;25:186–7.
- [10] Chander B, Dogra SS, Raina R, et al. Chondrocutaneous branchial remnants or cartilaginous choristoma: terminology, biological behavior and salience of bilateral cervical lesions. Turk Patoloji Derg 2014; 30:195–200.
- [11] Pham Dang N, Chevaleyre A, Troude B, et al. Bilateral cervical chondrocutaneous remnants: a familial observation. Br J Oral Maxillofac Surg 2013;51:e288–90.
- [12] Nielsen LJ, Von Rosen K, Jakobsen LP. Cervical chondrocutaneous branchial remnants: a case report. Eplasty 2016;16:ic17.
- [13] Ginat DT, Johnson DN, Shogan A, et al. Cervical chondrocutaneous branchial remnants. Head Neck Pathol 2018;12:244–6.
- [14] Tamir S, Nidal M, Constantin R, et al. Bilateral cervical chondrocutaneous branchial remnants. Int J Pediatr Otorhinolaryngol Extra 2008; 3:117–9.
- [15] Klockars T, Kajosaari L. Cervical chondrocutaneous branchial remnants. Cleft Palate Craniofac J 2017;54:223–6.
- [16] Lindgren VV. Bilateral cartilaginous branchial remnants (branchial appendages): case report. Plast Reconstr Surg 1946;17:304-8.
- [17] Punyamurthy M. Accessory auricles in the neck. J Laryngol Otol 1972;86:173–4.
- [18] Clarke JA. Are wattles of auricular or branchial origin? Br J Plast Surg 1976;29:238–44.
- [19] Doi O, Hutson JM, Myers NA, et al. Branchial remnants: a review of 58 cases. J Pediatr Surg 1988;23:789–92.
- [20] Fuad B, Elmir C, Samir DD. Neck auricles with microtia and low position of the right ear: a case report. Auris Nasus Larynx 2003;30:283–5.
- [21] Coras B, Hafner C, Roesch A, et al. Congenital cartilaginous rests of the neck (wattles). Dermatol Surg 2005;31:1349–50.
- [22] Ozturk H, Ozdemir T, Demirbag S, et al. Bilateral cervical chondrocutaneous remnants: a case report and review of the literature. Turk J Pediatr 2006;48:175–7.
- [23] Gilboa Y, Achiron R, Zalel Y, et al. Prenatal diagnosis of cervical chondrocutaneous vestige. Ultrasound Obstet Gynecol 2007;30: 1010–2.
- [24] Rameh C, Sidani C, Arabi M, et al. Bilateral cervical chondrocutaneous branchial remnants associated with cardiac anomalies. J Otolaryngol 2007;36:E79–81.
- [25] Dayal D, Menon P. Bilateral cervical chondrocutaneous branchial remnants. Indian Pediatr 2008;45:221.
- [26] Asahina A, Mitomi H, Sakurai N, et al. Multiple accessory tragi without cartilage: relationship with hair follicle naevi? Acta Derm Venereol 2009;89:316–7.
- [27] Oiso N, Kawada A. Cervical auricles in a family. Eur J Dermatol 2012;22:395-6.
- [28] Hemmaoui B, Fejjal N, Nadour K, et al. Cervical chondrocutaneous branchial remnants: two case reports. J Paediat Dermatol 2013;14: 33–5.
- [29] Kono T, Ro H, Murakami N, et al. Accessory auricles affecting the tragus and cheek occurring with cervical chondrocutaneous branchial remnants: A case report. An International Open Access Journal of Surgical Reconstruction (JPRAS OPEN) 2015;6:20–4.