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Anatomy-based diagnostic criteria for complex body wall anomalies (CBWA)

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Abstract

Background: Precise diagnosis and classification of CBWA cases can be challenging. BSA are considered when there is a body wall anomaly, skeletal abnormalities, and the umbilical cord is anomalous, absent or rudimentary, and LBWC when there is a body wall and structural limb anomalies with or without craniofacial abnormalities.

Methods: PubMed was searched for body stalk anomalies, limb body wall complex, body stalk anomalies and amniotic band syndrome, and limb body wall complex and amniotic band syndrome. Sixty nine articles were selected and reviewed. This article systematically classifies the variants of CBWA in 218 cases, the study is based on the embryological and anatomical criteria established by Martín-Alguacil and Avedillo to study BSA in the pig.

Results: Eight different BSA presentation were defined. One hundred and eighty nine cases were classified as BSA, from which five were Type I, nine Type II, 20 Type III, 57 Type IV, 11Type V, 24 Type VI, 11 Type VII, and 52 Type VIII. Twenty six cases presented cranial phenotype, 114 abdominal phenotype, 42 cranio/abdominal overlapping phenotype, and five without defined phenotype. In addition, 52 BSA cases presented some kind of spinal dysraphism (SPDYS) and were classified as BSA/SPDYS, most of these cases did not show structural limb anomalies, except for three cases and were classified as LBWC/SPDYS.

Conclusion: This morphology-based classification represents a useful tool for clinical diagnosis, it helps to quantify and to evaluate CBWA in a precise, objective manner.

KEYWORDS

ABS, BSA, classification, definition, LBWC

1 | INTRODUCTION

There is no consensus on the definition and classification of complex body wall anomalies (CBWA). Recently an anatomical and embryological criteria was used to classify body wall anomalies with multiple congenital anomalies in the pig (Martín-Alguacil & Avedillo, 2020a). Body stalk anomalies (BSA) were considered when there is a body wall anomaly, skeletal abnormalities, and the umbilical cord is anomalous, absent or rudimentary, and limb body wall complex (LBWC) when there is a body wall and structural limb anomalies with or without craniofacial abnormalities. The proposed

TABLE 1 Summary of anomalies described in the studied cases

	BWA	SPA	UCA	Limb anomaly	AA	CRA and/or FAA	SPDYS	UA and/or GA	AB	Proposed classification	Authors' diagnosis
Arici, Perçin, Ozer, Aslan, & Cetin, 2004	Ab	+	+ SUA	LLSA	+			+		BSA Type III LBWC Type II ABD phenotype	LBWC
Baruah & Choudhur, 2013	Ab	+	+	LLNSA	+			+		BSA Type IV ABD phenotype	LBWC
Bhat, Ilyas, & Dev, 2016 Case 1	Ab	+	+ SUA	LLNSA			+			BSA Type VIII ABD and SPDYS phenotype	LBWC
Case 2	Ab	+	+ SUA				+			BSA Type VIII SPDYS phenotype	LBWC
Case 3	Ab	+	ND				+			BSA Type VIII SPDYS phenotype	LBWC
Bohîlţea et al., 2017	Ab	+	+	ULSA LLNSA		+				BSA Type VII LBWC Type II CR phenotype	BSA
Bugge, 2012 Case 1	ThAb		+	LLNSA				+		BSA Type II ABD phenotype	BSA
Case 2	Ab	+	+ SUA	LLSA	+			+		BSA Type III LBWC Type II ABD phenotype	BSA
Case 3	Ab		+ SUA	LLSA LLNSA	+			+		BSA Type III LBWC Type II ABD phenotype	BSA
Case 4	Ab		+ SUA	LLNSA	+			+		BSA Type IV ABD phenotype	BSA
Case 5	Ab	+	+ SUA	LLNSA				+		BSA Type IV ABD phenotype	BSA
Case 6	ThAb	+	+ SUA	LLNSA	+		+	+		BSA Type II ABD and SPDYS phenotype	BSA
Case 7	Ab	+	+	LLNSA	+	+		+		BSA Type IV CR/ABD phenotype	BSA

(Continues)

TABLE 1 (Continued)

	BWA	SPA	UCA	Limb anomaly	AA	CRA and/or FAA	SPDYS	UA and/or GA	AB	Proposed classification	Authors' diagnosis
Case 8	Ab	+	+	LLNSA				+		BSA Type IV ABD phenotype	BSA
Case 9	Ab	+	+	LLSA	+	+		+		BSA Type III LBWC Type II CR/ABD phenotype	BSA
Case 10	Ab	+	+ SUA	LLSA	+	+		+		BSA Type III LBWC Type II CR/ABD phenotype	BSA
Case 11	Ab	+	+ SUA	LLNSA	+					BSA Type IV ABD phenotype	BSA
Case 12	Ab	+	+ SUA	LLNSA				+		BSA Type IV ABD phenotype	BSA
Case 13	Ab	+	+ SUA	ULNSA		+				BSA Type VIII CR phenotype	BSA
Case 14	Ab	+	+ SUA	LLNSA	+	+	+	+		BSA Type IV CR/ABD phenotype	BSA
Case 15	Ab	+	+	LLNSA				+		BSA Type IV ABD phenotype	BSA
Case 16	ThAb	+	SUA	ULNSA LLNSA	+			+		BSA Type II ABD phenotype	BSA
Chen, 2001	ThAb	+	+	ULSA	+	+		+	+	BSA Type I LBWC Type I CR/ABD phenotype	LBWC
Chen et al., 2007 Case 1	ThAb	+	SUA	LLSA LLNSA	+			+		BSA Type I LBWC Type I ABD phenotype	LBWC
Case 2	Ab	+	+ SUA	LLNSA	+			+		BSA Type IV ABD phenotype	LBWC
Case 3	Ab	+	+ SUA	LLNSA	+			+		BSA Type IV ABD phenotype	LBWC

(Continues)

TABLE 1 (Continued)

	BWA	SPA	UCA	Limb anomaly	AA	CRA and/or FAA	SPDYS	UA and/or GA	AB	Proposed classification	Authors' diagnosis
Case 4	Ab	+	+ SUA	LLSA	+			+		BSA Type III LBWC Type II ABD phenotype	LBWC
Case 5	Ab	+	+	LLSA						BSA Type VII LBWC Type II ABD phenotype	LBWC
Case 6	ThAb		+	LLNSA	+			+		BSA Type II ABD phenotype	LBWC
Case 7	Ab	+	+ SUA	LLSA LLNSA	+			+		BSA Type III LBWC Type II ABD phenotype	LBWC
Case 8	Ab	+	+	LLNSA	+			+		BSA Type IV ABD phenotype	LBWC
Case 9	Ab	+	+ SUA	LLSA						BSA Type VII LBWC Type II ABD phenotype	LBWC
Case 10	Ab	+	N	ULSA LLNSA		+			+	ABS/LBWC Type II CR phenotype	LBWC
Case 11	Ab	+	N	ULSA LLNSA		+			+	ABS/LBWC Type II CR phenotype	LBWC
Case 12	Ab	+	N	ULSA LLNSA		+			+	ABS/LBWC Type II CR phenotype	LBWC
Chen et al., 2009	Ab	+	+ SUA	LLSA				+	+	BSA Type III LBWC Type II ABD phenotype	LBWC
Chen, Chen, Su, & Wang, 2011	Ab	+	N	ULSA LLNSA		+			+	ABS/LBWC Type II CR phenotype	LBWC
Chen et al., 2018	Ab	+	+	LLNSA					+	BSA Type VIII ABD phenotype	LBWC

(Continues)

TABLE 1 (Continued)

	BWA	SPA	UCA	Limb anomaly	AA	CRA and/or FAA	SPDYS	UA and/or GA	AB	Proposed classification	Authors' diagnosis
Chikkannaiah, Dhumale, Kangle, & Shekar, 2013	Ab	+	+	LLNSA	+	+		+		BSA Type IV CR/ABD phenotype	LBWC
Coleman et al., 2018 Case 1	Ab	+					+	+		OEIS	OEIS
Case 2	Ab	+	+	LLSA			+	+		BSA Type III LBWC Type II ABD and SPDYS phenotype	OEIS
Case 3	Ab		+ SUA	LLNSA			+	+		BSA Type IV ABD and SPDYS phenotype	OEIS
Case 4	Ab	+	+ SUA	LLNSA			+	+		BSA Type IV ABD and SPDYS phenotype	OEIS LBWC
Case 5	Ab		+ SUA				+	+		OEIS	OEIS
Case 6	Ab	+	+	LLNSA			+			BSA Type VIII ABD and SPDYS phenotype	OEIS
Colpaert et al., 2000 Case 1	ThAb	+	+	ULNSA						BSA Type VI CR phenotype	LBWC
Case 2	Ab	+	+ SUA	LLNSA	+		+	+		BSA Type IV ABD and SPDYS phenotype	LBWC
Case 3	Ab	+	+ SUA	LLNSA	+			+		BSA Type IV ABD phenotype	LBWC
Case 4	Ab	+	+	LLNSA	+			+		BSA Type IV ABD phenotype	LBWC
Craven, Carey, & Ward, 1997 Case 1	Ab	+	+ SUA	LLSA	+			+		BSA Type III LBWC Type II ABD phenotype	LBWD
Case 2	Ab	+	+	LLNSA	+		+	+		BSA Type IV ABD and SPDYS phenotype	LBWD
Case 3	Ab	+	+ SUA	LLNSA	+		+	+		BSA Type IV ABD and SPDYS phenotype	LBWD
Case 4	Ab	+	+	LLNSA	+		+	+		BSA Type IV ABD and SPDYS phenotype	LBWD

(Continues)

TABLE 1 (Continued)

	BWA	SPA	UCA	Limb anomaly	AA	CRA and/or FAA	SPDYS	UA and/or GA	AB	Proposed classification	Authors' diagnosis
Case 5	Ab	+	+ SUA	LLNSA	+		+	+		BSA Type IV ABD and SPDYS phenotype	LBWD
Crespo, Pinar, & Kostadinov, 2012 Case 1	ThAb	+	+ SUA	ULNSA		+		+	+	BSA Type II CR/ABD and SPDYS phenotype	LBWC BSA
Case 2	ThAb	+	+ SUA	ULSA LLNSA						BSA Type V LBWC Type I CR phenotype	LBWC BSA
Cusí, Torrents, Vila, Antich, & Carrera, 1996 Case 1	Ab	+	+	LLNSA			+	+		BSA Type IV ABD and SPDYS phenotype	LBWC
Case 2	Ab	+	+	LLNSA			+	+		BSA Type IV ABD and SPDYS phenotype	LBWC
Case 3	Ab		ND	ULNSA		+	+			ABS	LBWC
Case 4	Ab		ND	LLNSA						ABS/FMR	LBWC
Case 5	Ab	+	+	LLNSA			+			BSA Type VIII ABD and SPDYS phenotype	LBWC
Case 6	Ab	+	+	LLNSA			+			BSA Type VIII ABD and SPDYS phenotype	LBWC
Case 7	Ab	+	+	LLNSA			+			BSA Type VIII ABD and SPDYS phenotype	LBWC
Case 8	Ab		ND	LLSA		+			+	ABS	LBWC
Das, Maharana, Subudhi, & Rao, 2013	Ab	+	+	LLSA LLNSA				+		BSA Type III LBWC Type II ABD phenotype	LBWC
Daskalakis & Nicolaides, 2002 Case 1	Ab	+	+ SUA	LLNSA			+	+		BSA Type IV ABD and SPDYS phenotype	BSA
Case 2	Ab	+	ND	LLNSA						BSA Type VIII ABD phenotype	BSA
Daskalakis, Pilalis, Papadopoulos, & Antsaklis, 2003	Ab	+	+					+		BSA Type IV ABD phenotype	BSA

(Continues)

TABLE 1 (Continued)

	BWA	SPA	UCA	Limb anomaly	AA	CRA and/or FAA	SPDYS	UA and/or GA	AB	Proposed classification	Authors' diagnosis
Deruelle et al., 2000 Case 1	ThAb	+	+	ULNSA LLNSA						BSA Type VI CR/ABD phenotype	LBWC
Case 2	Ab	+	ND	LLNSA	+		+	+		BSA Type IV ABD and SPDYS phenotype	LBWC
Case 3	Ab	+	+	LLNSA			+			BSA Type VIII ABD and SPDYS phenotype	LBWC
Case 4	Ab	+	+	LLNSA		+	+	+		BSA Type IV ABD and SPDYS phenotype	LBWC
Devi, Cicy, Thambi, & Poothiode, 2015	Ab	+	+	LLNSA		+	+		+	BSA Type VIII CR/ABD and SPDYS phenotype	LBWC ABSQ
D'Souza, Indrajit, & Menon, 2004	Ab	+	+ SUA	LLNSA	+			+		BSA Type IV ABD phenotype	LBWC
Durga & Renukadevi, 2016	Ab	+	+ SUA	LLSA LLNSA					+	BSA Type VII LBWC Type II ABD phenotype	ABS
Fukumasu et al., 1993	Ab	+	+ SUA	LLSA LLNSA	+				+	BSA Type III LBWC Type II ABD phenotype	LBWC
Gajzer et al., 2015 Case 1	Ab	+	+	LLNSA	+		+	+		BSA Type IV ABD and SPDYS phenotype	LBWC
Case 2	ThAb	+	+	ULSA LLNSA	+	+				BSA Type I LBWC Type I CR/ABD phenotype	LBWC
Case 3	Gs	+	N	LLNSA						Gs/FMR	LBWC
Case 4	Ab	+	+	LLNSA	+	+		+		BSA Type IV CR/ABD phenotype	LBWC
Gazolla et al., 2014 Case 1			+ SUA	LLNSA		+			+	ABS	LBWD
Case 2	Ab	+	+ SUA	LLNSA		+	+	+	+	BSA Type VIII ABD and SPDYS phenotype	LBWD

(Continues)

TABLE 1 (Continued)

	BWA	SPA	UCA	Limb anomaly	AA	CRA and/or FAA	SPDYS	UA and/or GA	AB	Proposed classification	Authors' diagnosis
Case 3	Ab	+	+	LLNSA					+	BSA Type VIII ABD phenotype	LBWD
Case 4	Ab	+	N	LLNSA					+	ABS	LBWD
Case 5	Ab	+	+ SUA	LLNSA						BSA Type VIII ABD phenotype	LBWD
Case 6			N	LLNSA		+			+	ABS	LBWD
Case 7	Ab	+	+	LLNSA			+			BSA Type VIII ABD and SPDYS phenotype	LBWD
Case 8	Ab	+	+ SUA	LLNSA			+		+	BSA Type VIII ABD and SPDYS phenotype	LBWD
Ginsberg, Cadkin, & Strom, 1997	Ab	+	+ SUA	LLNSA	+		+	+		BSA Type IV ABD and SPDYS phenotype	BSA
Grigore, 2014	Ab	+	+	ULNSA		+				BSA Type VIII CR phenotype	LBWC
Gulczyński, Świątkowska-Freund, Paluchowski, Hermann-Okoniewska, & Iżycka-Świeszewska, 2019 Case 1	ThAb	+	ND	LLSA	+	+		+		BSA Type I LBWC Type I CR/ABD phenotype	LBWC
Case 3	Ab	+	+	LLSA						BSA Type VII LBWC Type II ABD phenotype	LBWC
Case 4	Ab		+	LLNSA						BSA Type VIII ABD phenotype	LBWC
Case 5	Ab		+	ULNSS LLNSA		+				BSA Type VIII CR/ABD phenotype	LBWC
Case 6	Ab		+			+				BSA Type VIII CR phenotype	LBWC
Case 7	Ab	+	+	LLSA LLNSA						BSA Type VII LBWC Type II ABD phenotype	LBWC

(Continues)

TABLE 1 (Continued)

	BWA	SPA	UCA	Limb anomaly	AA	CRA and/or FAA	SPDYS	UA and/or GA	AB	Proposed classification	Authors' diagnosis
Case 8	Ab	+	+	LLNSA		+.	+			BSA Type VIII CR/ABD and SPDYS phenotype	LBWC
Case 9	Ab		+	LLNSA				+		BSA Type VIII ABD phenotype	LBWC
Case 10	Ab	+	+	LLNSA	+					BSA Type IV ABD phenotype	LBWC
Case 11	Ab	+	+	ULNSA LLNSA		+			+	BSA Type VIII CR/ABD phenotype	LBWC
Gupta, Venkatesan, Chandra, Rajeswari, & Devi, 2015	Ab	+	+ SUA	LLSA LLNSA					+	BSA Type VII LBWC Type II ABD phenotype	ABS
Haciveliogl u & Tarim, 2010 Case 1	Ab	+	ND	ULSA LLNSA		+				BSA Type VII LBWC Type II CR/ ABD phenotype	LBWC
Case 2	Ab	+	ND	LLSA						BSA Type V LBWC Type II ABD phenotype	LBWC
Case 3	Ab	+	ND	ULNSA		+				BSA Type VIII CR phenotype	LBWC
Halder, 2010 Case 1	ThAb	+	+	ULNSA LLNSA	+	+		+	+	BSA Type II CR/ABD phenotype	ABS LBWC
Case 3	ThAb		+			+			+	BSA Type VI CR phenotype	ABS LBWC
Case 4	ThAb		+			+			+	BSA Type VI CR phenotype	ABS LBWC
Hartwig, Vermeij-Keers, De Vries, Kagie, & Kragt, 1989 Case 1	ThAb	+	+	LLNSA		+			+	BSA Type VI CR/ABD phenotype	LBWC

(Continues)

TABLE 1 (Continued)

	BWA	SPA	UCA	Limb anomaly	AA	CRA and/or FAA	SPDYS	UA and/or GA	AB	Proposed classification	Authors' diagnosis
Case 2	Ab		+ SUA	LLSA	+			+		BSA Type III LBWC Type II ABD phenotype	LBWC
Case 3	ThAb	+	+	ULNSA						BSA Type VI CR phenotype	LBWC
Case 4	ThAb	+	+ SUA	LLNSA	+			+		BSA Type II ABD phenotype	LBWC
Higuchi et al., 2013	Ab	+	+	LLNSA						BSA Type VIII ABD phenotype	BSA
Hirokawa et al., 2003	Ab	+	+ SUA							BSA Type VIII ND phenotype	BSA
Hunter, Seaver, & Stevenson, 2011 Case 1	Ab	+	+ SUA	LLSA	+			+		BSA Type III LBWC Type II ABD phenotype	LBWD
Case 2	Ab	+	+	LLSA LLNSA	+			+		BSA Type III LBWC Type II ABD phenotype	LBWD
Case 3	Ab		+			+				BSA Type VIII CR phenotype	
Case 4	Ab		+	LLSA ULNSA	+	+		+	+	BSA Type III LBWC Type II CR/ ABD phenotype	LBWD
Iba et al., 2016 Case 1	Ab	+	+	LLNSA						BSA Type VIII ABD phenotype	BSA
Case 2	Ab	+	+	LLNSA						BSA Type VIII ABD phenotype	BSA
Jensen, Hägerstrand, Brun, & Löfgren, 1993	ThAb		+	ULSA		+			+	BSA Type V LBWC Type I CR phenotype	LBWC
Jun, Ahn, Lee, Chi, & Cha, 1991	Ab	+	+							BSA Type VIII ABD phenotype	BSA

(Continues)

TABLE 1 (Continued)

	BWA	SPA	UCA	Limb anomaly	AA	CRA and/or FAA	SPDYS	UA and/or GA	AB	Proposed classification	Authors' diagnosis
Kamudhamas & Manusook, 2001 Case 1	ThGs	+	N	ULNSA						ABS/FMR	LBWC
Case 2	Ab	+	ND	ULNSA		+				BSA Type VIII CR phenotype	LBWC
Kocherla, Kumari, & Kocherla, 2015	Ab	+	+	LLNSA		+	+			BSA Type VIII CR/ABD and SPDYS phenotype	BSC
Kruszka et al., 2015 Case 1	VACS		N	ULNSA LLNSA		+	+			Amyoplasia ABS/FMR	LBWC
Case 2			N	LLNSA	+				+	ABS	LBWC
Litwin, Merlob, & Grunebaum, 1988 Case 1	Ab	+	+	LLNSA	+	+		+		BSA Type IV CR/ABD phenotype	LBWC
Case 2	Ab		+	LLNSA	+			+		BSA Type IV ABD phenotype	LBWC
Liu, Yu, Chang, & Chang, 2003	Ab	+	+ SUA	ULNSA						BSA Type VIII CR phenotype	LBWC
Managoli, Chaturvedi, Vilhekar, & Gagane, 2003	Ab	+	+ SUA	LLNSA	+	+		+		BSA Type IV CR/ABD phenotype	LBWC
Mandrekar, Amoncar, Banaulikar, Sawant, & Pinto, 2014	Ab	+	+	LLNSA	+		+	+		BSA Type IV ABD and SPDYS phenotype	OEIX BSA/LBWC
Maruyama et al., 2015	Ab	+	+ SUA		+					BSA Type IV ABD phenotype	BSA
Mathai et al., 2009	Ab		+ SUA	LLNSA	+					BSA Type IV ABD phenotype	BSA
Moerman et al., 1992 Case 7	Ab	+	+ SUA	LLNSA				+		BSA Type IV ABD phenotype	LBWC
Case 8	Ab	+	+	ULNSA LLNSA		+			+	BSA Type VIII CR phenotype	LBWC
Case 9	ThAb	+	+ SUA	ULNSA LLNSA		+			+	BSA Type VI CR/ABD phenotype	LBWC

(Continues)

TABLE 1 (Continued)

	BWA	SPA	UCA	Limb anomaly	AA	CRA and/or FAA	SPDYS	UA and/or GA	AB	Proposed classification	Authors' diagnosis
Case 10	Ab	+	+	ULNSA LLNSA		+			+	BSA Type VIII CR/ABD phenotype	LBWC
Case 11	Th		+			+			+	ABS	LBWC
Case 12			ND	LLNSA		+			+	ABS	LBWC
Case 13			N	ULNSA LLNSA		+			+	ABS	LBWC
Case 14	ThAb	+	+ SUA	ULNSA LLNSA		+			+	BSA Type VI CR/ABD phenotype	LBWC
Case 15	ThAb	+	+	ULNSA LLNSA		+			+	BSA Type VI CR/ABD phenotype	LBWC
Case 16	ThAb	+	+	ULNSA		+			+	BSA Type VI CR phenotype	LBWC
Case 17	Ab	+	+ SUA	LLNSA						BSA Type VIII CR/ABD phenotype	LBWC
Case 18	Ab	+	+	ULNSA LLNSA						BSA Type VIII CR/ABD phenotype	LBWC
Negishi et al., 1998 Case 1	Ab	+	+ SUA	LLNSA	+			+		BSA Type IV ABD phenotype	LBWC
Case 2	ThAb	+	+ SUA	LLSA LLNSA			+			BSA Type V LBWC Type I ABD and SPDYS phenotype	LBWC
Case 3	Ab	+	+ SUA	LLNSA	+		+			BSA Type IV CR/ABD and SPDYS phenotype	LBWC
Case 4	Ab	+	+ SUA	LLNSA			+			BSA Type VIII ABD and SPDYS phenotype	LBWC
Case 5	ThAb	+	SUA	ULNSA LLSA	+	+			+	BSA Type I LBWC Type I CR/ABD phenotype	LBWC

(Continues)

TABLE 1 (Continued)

	BWA	SPA	UCA	Limb anomaly	AA	CRA and/or FAA	SPDYS	UA and/or GA	AB	Proposed classification	Authors' diagnosis
Case 6	Ab	+	+ SUA	LLNSA		+	+			BSA Type VIII CR/ABD and SPDYS phenotype	LBWC
Case 7	Ab	+	+	LLNSA						BSA Type VIII ABD phenotype	LBWC
Case 8	Ab	+	+ SUA	LLNSA						BSA Type VIII CR/ABD phenotype	LBWC
Okido et al., 2017	ThAb	+	+	ULSA						BSA Type V LBWC Type I CR phenotype	LBWC
Palacios & Rodriguez, 1990	Ab	+	+ SUA	LLNSA	+		+	+		BSA Type IV ABD and SPDYS phenotype	LBWC
Patten et al., 1986 Case 1	Ab	+	ND	LLNSA				+		BSA Type IV ABD phenotype	LBWC
Case 2	ThAb	+	+	ULSA		+				BSA Type V LBWC Type I CR phenotype	LBWC
Case 3	Ab	+	+ SUA	LLNSA			+			BSA Type VIII ABD and SPDYS phenotype	LBWC
Case 4	Ab	+	+	ULSA LLNSA		+				BSA Type VII LBWC Type II CR/ABD phenotype	LBWC
Case 5	ThAb	+	+	LLNSA			+	+		BSA Type II ABD and SPDYS phenotype	LBWC
Case 6	ThAb	+	+ SUA	ULNSA LLNSA		+			+	BSA Type VI CR/ABD phenotype	LBWC
Case 7	ThAb	+	ND	ULNSA		+	+			BSA Type VI CR and SPDYS phenotype	LBWC
Case 8	Ab	+	+			+				BSA Type VIII CR phenotype	LBWC

(Continues)

TABLE 1 (Continued)

	BWA	SPA	UCA	Limb anomaly	AA	CRA and/or FAA	SPDYS	UA and/or GA	AB	Proposed classification	Authors' diagnosis
Case 9	ThAb	+	ND	LLSA			+		+	BSA Type V LBWC Type I CR/ABD and SPDYS phenotype	LBWC
Case 10	ThAb		ND	ULNSA						BSA Type VI CR phenotype	LBWC
Case 11	ThAb	+	ND	LLNSA					+	BSA Type VI CR phenotype	LBWC
Case 12	ThAb	+	+ SUA	LLNSA NSDLL						BSA Type VI ABD phenotype	LBWC
Case 13	ThAb	+	ND	LLNSA			+			BSA Type VI ABD and SPDYS phenotype	LBWC
Paul, Zosmer, Jurkovic, & Nicolaides, 2001	Ab	+	ND	LLNSA						BSA Type VIII ABD phenotype	BSA
Plakkal, John, Jacob, Chithira, & Sampath, 2008	ThAb	+	+	ULSA						BSA Type V LBWC Type I CR phenotype	LBWC
Prasun, Behera, & Pradhan, 2008	Ab	+	+	LLNSA	+			+		BSA Type IV ABD phenotype	LBWC
Pumberger, Schaller, & Bernaschek, 2001 Case 1	Ab	+	ND	LLSA	+	+		+		BSA Type III LBWC Type II CR/ABD phenotype	LBWC
Case 2	Ab	+	ND	LLSA	+	+		+	+	BSA Type III LBWC Type II CR/ABD phenotype	LBWC
Case 3	Ab	+	+ SUA	LLNSA				+		BSA Type IV ABD phenotype	LBWC
Case 4	ThAb	+	+	LLNSA				+		BSA Type II ABD phenotype	LBWC
Quijano, Rey, Echeverry, & Axt-Fliedner, 2014	Ab	+	+	LLSA LLNSA		+				BSA Type VII LBWC Type II CR/ABD phenotype	BSA

(Continues)

TABLE 1 (Continued)

	BWA	SPA	UCA	Limb anomaly	AA	CRA and/or FAA	SPDYS	UA and/or GA	AB	Proposed classification	Authors' diagnosis
Rachad, Chaara, Bouguern, & Melhouf, 2012	Ab	+	+	ULSA LLNSA		+				BSA Type VII LBWC Type II CR/ABD phenotype	LBWC
Routhu et al., 2016 Case 1			ND	ULSA ULNSA					+	ABS	ABS
Case 2			ND	LLSA					+	ABS	ABS
Case 3	Ab	+	N	LLNSA					+	ABS/FMR	BSA ABS
Case 4	Ab	+	N	LLNSA						ABS/FMR	BSA ABS
Case 5	ThAb	+	+			+				BSA Type VI CR phenotype	BSA ABS
Case 6	Ab	+	N			+	+		+	ABS/FMR	BSA ABS
Case 7	Ab		+	LLNSA						BSA Type VIII ABD phenotype	BSA ABS
Case 8	ThAb	+	+	LLNSA						BSA Type VI ABD phenotype	BSA ABS
Case 9	ThAb		ND							BWCA	BSA ABS
Case 10	Ab	+	N	LLNSA						ABS/FMR	BSA ABS
Russo, D'Armiento, Angrisani, & Vecchione, 1993 Case 1	Ab	+	+	LLNSA	+		+	+		BSA Type IV ABD and SPDYS phenotype	LBWC
Case 2	Ab	+	ND	LLNSA	+		+	+		BSA Type IV ABD and SPDYS phenotype	LBWC
Case 3	Ab	+	+	LLNSA	+		+	+		BSA Type IV ABD and SPDYS phenotype	LBWC
Case 4	Ab	+	+	LLNSA	+			+		BSA Type IV ABD phenotype	LBWC
Case 5	Ab	+	+	LLNSA	+		+	+		BSA Type IV ABD and SPDYS phenotype	LBWC

(Continues)

TABLE 1 (Continued)

	BWA	SPA	UCA	Limb anomaly	AA	CRA and/or FAA	SPDYS	UA and/or GA	AB	Proposed classification	Authors' diagnosis
Case 6	Ab	+	+	LLNSA	+		+	+		BSA Type IV ABD and SPDYS phenotype	LBWC
Case 7	Ab	+	ND	LLNSA	+		+	+		BSA Type IV ABD and SPDYS phenotype	LBWC
Case 8	Ab	+	ND	LLSA	+			+		BSA Type III LBWC Type I ABD phenotype	LBWC
Sahinoglu et al., 2007 Case 1	Ab	+	+	LLSA LLNSA				+		BSA Type III LBWC Type II ABD phenotype	Type 3 LBWC
Case 2	Ab	+	+	LLNSA				+		BSA Type IV ABD phenotype	Type 3 LBWC
Case 3	Ab	+	+ SUA	NS						BSA Type VIII ABD phenotype	Type 3 LBWC
Case 4	ThAb	+	+ SUA	ULSA LLNSA					+	BSA Type V LBWC Type I CR/ABD phenotype	Type 2 LBWC
Case 5		+		ULSA LLNSA	+	+		+		ABS/FMR	Type 1 LBWC
Case 6	ThAb	+	+ SUA	ULSA						BSA Type V LBWC Type I CR phenotype	Type 2 LBWC
Saritha et al., 2013	Ab	+	+	ULSA ULNSA				+		BSA Type IV LBWC Type II CR/ABD phenotype	LBWC
Smrcek et al., 2003 Case 1	ThAb	+	+							BSA Type VI ND phenotype	BSA
Case 2	ThAb		+ SUA							BSA Type VI ND phenotype	
Case 3	ThAb	+	+	ULSA LLNSA						BSA Type V LBWC Type I CR/ABD phenotype	BSA

(Continues)

TABLE 1 (Continued)

	BWA	SPA	UCA	Limb anomaly	AA	CRA and/or FAA	SPDYS	UA and/or GA	AB	Proposed classification	Authors' diagnosis
Case 4	Ab	+	+ SUA							BSA Type VIII ND phenotype	
Case 5	Ab	+	+ SUA	LLNSA			+			BSA Type VIII ABD and SPDYS phenotype	BSA
Case 6	ThAb	+	+	LLNSA						BSA Type VI ABD phenotype	BSA
Case 7	Ab	+	+ SUA	LLNSA			+			BSA Type VIII ABD and SPDYS phenotype	BSA
Case 8	Ab	+	+ SUA	LLNSA				+		BSA Type IV ABD phenotype	BSA
Case 9	Ab	+	+							BSA Type VIII ND phenotype	BSA
Case 10	ThAb	+	+	LLNSA						BSA Type VI ABD phenotype	BSA
Case 11	Ab	+	+							BSA Type VIII CR phenotype	BSA
Stein et al., 2009	ThAb		+	LLNSA		+				BSA Type VI CR/ABD phenotype	Cantrell/LBWC
Tang, Oechler, Hinke, Segura, & Franciosi, 1991	Ab	+	+ SUA	ULNSA LLNSA	+		+	+		BSA Type IV ABD and SPDYS phenotype	LBWC Sirenomelia
Vujovic et al., 2017	Th			ULNSA						ABS/FMR	LBWC
Wu, Yang, & Yuan, 2000	Ab	+		LLNSA		+		+		ABS/FMR	LBWC
Zeidler, Oudesluijs, Schoonderwaldt, & Van Bever, 2014	ThAb		+ SUA	LLNSA		+			+	BSA Type VI CR/ABD phenotype	LBWS BSA AB

Abbreviations: AA, Anal atresia; Ab, Abdominoschisis; AB, Amniotic band; ABD, Abdominal; ABS, amniotic band syndrome; BSA, Body Stalk Anomaly; BSC, Body stalk complex; BWA, body wall anomaly; BWCA, Body wall closing anomaly; CR, cranial; CRA, cranial anomaly; FAA, Facial anomaly; FMR, Fetal movement restriction; GA, Genital anomaly; Gs, Gastroschisis; LBWC, Limb Body Wall Complex; LBWD, Limb Body Wall Defect; LBWS, limb body wall sequence; LLNSA, Lower limb nonstructural anomaly; LLSA, Lower limb structural anomaly; N, Normal; ND, not defined; SPDYS, Spinal dysraphism; UA, Urinary anomaly; UCA, Umbilical cord anomaly; ULNSA, Upper limb nonstructural anomaly; ULSA, Upper limb structural anomaly; VACS, Ventral anomaly covered with skin; SPA, spine anomaly; SPDYS, Spinal dysraphism; SUA, Single umbilical artery; Th, thoracoschisis; ThAb, thoracoabdominoschisis.

classification distinguished the type of body wall (abdominoschisis and thoracoabdominoschisis) and limb anomalies. Based on the studies by Rittler et al. (2019), they defined limb structural anomalies on the basis of embryological failures, and considered nonstructural anomalies when they were thought to be caused by amniotic bands actions, and/or fetal movement restrictions (FMR), including arthrogryposis. Amelia and/or pelvic limbs phocomelia were evaluated as structural anomalies, and thoracic limbs phocomelia, arthrogryposis, ankyloses, and/or anomalous rotation as nonstructural limb anomalies (Martín-Alguacil & Avedillo, 2020a). They established four types of presentation for BSA: (a) BSA Type I: fetus with spinal and umbilical cord (UC)anomalies, thoracoabdominoschisis, anal atresia and/or other internal organs structural anomalies, and structural limb anomalies; (b) BSA Type II: fetus with spinal and UC anomalies, thoracoabdominoschisis, anal atresia and/or other internal organs structural anomalies, and nonstructural limb anomalies; (c) BSA Type III: fetus with spinal and UC anomalies, abdominoschisis, anal atresia and/or other internal organs structural anomalies, and structural limb anomalies; and (d) BSA Type IV: fetus with spinal and UC anomalies, abdominoschisis, anal atresia and/or other internal organs structural anomalies, and nonstructural limb anomalies. Additionally they differentiated two types of LBWC; Type I: characterized by thoracoabdominoschisis and structural limb anomalies, and Type II: characterized by abdominoschisis and structural limb anomalies, which corresponded to BSA type I and type III. The aim of this study is to apply this anatomical- and embryological-based criteria to classify CBWA cases found in the medical literature.

2 | MATERIALS AND METHODS

This is a morphological study designed to establish a precise criteria to differentiate and classify CBWA in an accurate manner. PubMed was searched using the following keywords: body stalk anomalies (158 articles), limb body wall complex (177 articles), body stalk anomalies and limb body wall complex (19 articles), body stalk anomalies and amniotic band syndrome(nine articles), and limb body wall complex and amniotic band syndrome (14 articles). Only articles containing clinical cases were selected and reviewed. A clear description of fetal anomalies found was necessary to be included in the study. Thus, articles without a complete postmortem clinical data were discarded. After revision, 76 articles were sort out, and from those 69 were selected, from which 218 cases were studied and classified.

A fetus was considered BSA when presented the following clinical features: a body wall anomaly, skeletal abnormalities, and the UC anomalous, absent or rudimentary. And it was considered LBWC when presented body wall and structural limb anomalies with or without craniofacial abnormalities. This study distinguished the type of body wall (abdominoschisis and thoracoabdominoschisis) and limb anomalies. Amelia and/or pelvic limbs phocomelia were considered as structural anomalies, and thoracic limbs phocomelia, arthrogryposis, ankyloses, and/or anomalous rotation as nonstructural limb anomalies.

3 | RESULTS

A summary of all the studied cases with the original diagnosis and the proposed classification is presented on Table 1. Cases with normal UC were not considered BSA, whereas not defined UC cases were considered for the BSA study. For this study eight BSA types were defined and presented on Table 2. BSA Type I: fetus with thoracoabdominoschisis, spinal defect, anomalous UC, structural limb defect, anal atresia or urinary and/or genital defect. BSA Type II: fetus with thoracoabdominoschisis, spinal defect, anomalous UC, nonstructural limb defect or no limb defect, anal atresia or urinary and/or genital defect. BSA Type III: fetus with abdominoschisis, spinal defect, anomalous UC, structural limb defect, anal atresia or urinary and/or genital defect. BSA Type IV: fetus with abdominoschisis, spinal defect, anomalous UC, nonstructural limb defect or no limb defect, anal atresia or urinary and/or genital defect. BSA Type V: fetus with thoracoabdominoschisis, spinal defect, anomalous UC, and structural limb defect. BSA Type VI: fetus with thoracoabdominoschisis, spinal defect, anomalous UC, and nonstructural limb defect. BSA Type VII: fetus with abdominoschisis, spinal defect, anomalous UC, and structural limb defect. BSA Type VIII: fetus with abdominoschisis, spinal defect, anomalous UC, and nonstructural limb or no limb defect. One hundred and eighty nine cases were classified as BSA, 21 as ABS with or without FMR, four as ABS/LBWC, two as OEIS, one as GS/FMR, and one as BWCA (Table 2). In addition, 47 BSA cases presenting structural limb anomalies were also classified as LBWC (Table 3). BSA with thoracoabdominoschisis (Type I, and Type V) were considered LBWC Type I, and BSA with abdominoschisis (Type III and Type VII) were considered LBWC Type II (Martín-Alguacil & Avedillo, 2020a). Sixteen cases were classified as LBWC Type I, and 31 LBWC Type II. Some cases without spinal anomaly but presenting some other skeletal anomaly were classified as BSA and identified on Table 2.

4 | DISCUSSION

In the original articles, LBWC was diagnosed in 148 cases (two of them with sirenomelia), BSA in 42 cases, OEIS in five cases, ABS in three cases, LBWC/BSA in three cases,

TABLE 2 CBWD cases classification

BSA	Clinical features		# BSA
Type I	Thoracoabdominoschisis, spinal defect, anomalous umbilical cord, structural limb defect, anal atresia or urinary and/or genital defect.	Chen, 2001; Chen et al., 2007 Case 1; Gajzer et al., 2015 Case 2; Gulczyński et al., 2019 Case 1; Negishi et al., 1998 Case 5	5/189 2.64%
Type II	Thoracoabdominoschisis, spinal defect, anomalous umbilical cord, nonstructural limb defect or no limb defect, anal atresia or urinary and/or genital defect.	Bugge, 2012 Cases: 1[a], 6, 16; Chen et al., 2007 Case 6[a]; Crespo et al., 2012 Case 1; Halder, 2010 Case 1; Hartwig et al., 1989 Case 4; Patten et al., 1986 Case 5; Pumberger et al., 2001 Case 4	9/189 4.76%
Type III	Abdominoschisis, spinal defect, anomalous umbilical cord, structural limb defect, anal atresia or urinary and/or genital defect.	Arici et al., 2004;Bugge, 2012 Cases: 2, 3[a], 9, 10; Chen et al., 2007 Cases: 4, 7; Chen et al., 2009; Coleman et al., 2018 Case 2; Craven et al., 1997 Case 1; Das et al., 2013; Fukumasu et al., 1993; Hartwig et al., 1989 Case 2[a]; Hunter et al., 2011 Cases: 1, 2, 4[a]; Pumberger et al., 2001 Cases: 1, 2; Russo et al., 1993 Case 8; Sahinoglu et al., 2007 Case 1	20/189 10.58%
Type IV	Abdominoschisis, spinal defect, anomalous umbilical cord, nonstructural limb defect or no limb defect, anal atresia or urinary and/or genital defect.	Baruah & Choudhur, 2013; Bugge, 2012 Cases: 4[a], 5, 7, 8, 11, 12, 14, 15; Chen et al., 2007 Cases: 2, 3, 8; Chikkannaiah et al., 2013; Coleman et al., 2018 Cases: 3, 4; Colpaert et al., 2000 Cases: 2, 3, 4; Craven et al., 1997 Cases: 2–5; Cusí et al.,1996 Cases: 1, 2; Daskalakis & Nicolaides, 2002 Case 1; Daskalakis et al., 2003; Deruelle et al., 2000 Cases: 2, 4; D'Souza et al., 2004; Gajzer et al., 2015 Cases: 1, 4; Ginsberg et al., 1997; Gulczyński et al., 2019 Case 10; Litwin et al., 1988 Cases: 1, 2; Managoli et al., 2003; Mandrekar et al., 2014; Maruyama et al., 2015; Mathai et al., 2009[a]; Moerman et al., 1992 Case 7; Negishi et al., 1998 Cases: 1, 3; Palacios & Rodriguez, 1990; Patten et al., 1986 Case 1; Prasun et al., 2008; Pumberger et al., 2001 Case 3; Russo et al., 1993 Cases: 1–7; Sahinoglu et al., 2007 Case 2; Saritha et al., 2013; Smrcek et al., 2003 Case 8; Tang et al., 1991	57/189 30.15%
Type V	Thoracoabdominoschisis, spinal defect, anomalous umbilical cord, and structural limb defect.	Crespo et al., 2012 Case 2; Hacivelioglu & Tarim, 2010 Case 2; Jensen et al., 1993[a]; Negishi et al., 1998 Case 2; Okido et al., 2017; Patten et al., 1986 Cases: 2, 9; Plakkal et al., 2008; Sahinoglu et al., 2007 Cases: 4, 6; Smrcek et al., 2003 Case 3	11/189 5.82%
Type VI	Thoracoabdominoschisis, spinal defect, anomalous umbilical cord, and nonstructural limb defect.	Colpaert et al., 2000 Case 1; Deruelle et al., 2000 Case 1; Halder, 2010 Cases: 3[a], 4[a]; Hartwig et al., 1989 Cases: 1, 3; Moerman et al., 1992 Cases: 9, 14–16; Patten et al., 1986 Cases: 6, 7, 10–13; Routhu et al., 2016 Cases: 5, 8; Smrcek et al., 2003 Cases: 1, 2, 6, 10; Stein et al., 2009;Zeidler et al., 2014	24/189 12.69%
Type VII	Abdominoschisis, spinal defect, anomalous umbilical cord, and structural limb defect.	Bohîlţea et al., 2017; Chen et al., 2007 Cases: 5, 9; Durga & Renukadevi, 2016; Gulczyński et al., 2019 Cases: 3, 7; Gupta et al., 2015; Hacivelioglu & Tarim, 2010 Case 1; Patten et al., 1986 Case 4; Quijano et al., 2014; Rachad et al., 2012	11/189 5.82%
Type VIII	Abdominoschisis, spinal defect, anomalous umbilical cord, and nonstructural limb or no limb defect.	Bhat et al., 2016 Cases: 1–3; Bugge, 2012 Case 13; Chen et al., 2018; Coleman et al., 2018 Cases: 6; Cusí et al.,1996 Cases: 5, 6, 7; Daskalakis & Nicolaides, 2002 Case 2; Deruelle et al., 2000 Case 3; Devi et al., 2015; Gazolla et al., 2014 Cases: 2, 3, 5, 7, 8; Grigore, 2014; Gulczyński et al., 2019 Cases: 4–6, 8, 9, 11; Hacivelioglu & Tarim, 2010 Case 3; Higuchi et al., 2013; Hirokawa et al., 2003; Hunter et al., 2011 Case 3[a]; Iba et al., 2016 Cases: 1, 2; Jun et al., 1991; Kamudhamas & Manusook, 2001 Case 2; Kocherla et al., 2015; Liu et al., 2003; Moerman et al., 1992 Cases: 8, 10, 17, 18; Negishi et al., 1998 Cases: 4, 6, 7, 8; Patten et al., 1986 Cases: 3, 8; Paul et al., 2001; Routhu et al., 2016 Case 7; Sahinoglu et al., 2007 Case 3; Smrcek et al., 2003 Cases: 4, 5, 7, 9, 11	52/189 27.51%

Abbreviation: BSA, Body stalk anomalies.

[a]No spinal defect.

TABLE 3 Phenotypes of the studied cases

BSA Phenotypes	Limb anomaly		CASES
CRANIAL	SLA LBWC	Bohîlţea et al., 2017; Crespo et al., 2012 Case 2; Jensen et al., 1993; Okido et al., 2017; Patten et al., 1986 Case 2; Plakkal et al., 2008; Sahinoglu et al., 2007 Case 6	7/189 3.7%
	NSLA	Bugge, 2012 Case 13; Colpaert et al., 2000 Case 1; Grigore, 2014; Gulczyński et al., 2019 Case 6; Hacivelioglu & Tarim, 2010 Case 3;Halder, 2010 Cases: 3, 4; Hartwig et al., 1989 Case 3; Hunter et al., 2011 Case 3; Kamudhamas & Manusook, 2001 Case 2; Liu et al., 2003; Moerman et al., 1992 Cases: 8, 16; Patten et al., 1986 Cases: 7, 8, 10, 11; Routhu et al., 2016 Case 5; Smrcek et al., 2003 Case 11	19/189 10.05%
ABDOMINAL	SLA LBWC	Arici et al., 2004; Bugge, 2012 Cases: 2, 3; Chen et al., 2007 Cases: 1, 4, 5, 7, 9; Chen et al., 2009; Coleman et al., 2018 Case 2; Craven et al., 1997 Case 1; Das et al., 2013; Durga & Renukadevi, 2016; Fukumasu et al., 1993; Gulczyński et al., 2019 Cases: 2, 7; Gupta et al., 2015; Hacivelioglu & Tarim, 2010 Case 2; Hartwig et al., 1989 Case 2; Hunter et al., 2011 Cases: 1, 2; Negishi et al., 1998 Case 2; Russo et al., 1993 Case 8; Sahinoglu et al., 2007 Case 1	24/189 12.69%
	NSLA	Baruah & Choudhur, 2013; Bhat et al., 2016 Cases: 1; Bugge, 2012 Cases: 1, 4, 5, 6, 8, 11, 12, 15, 16; Chen et al., 2007 Cases: 2, 3, 6, 8; Chen et al., 2018; Coleman et al., 2018 Cases: 3–6; Colpaert et al., 2000 Cases: 2–4; Craven et al., 1997 Cases: 2–5; Cusí et al.,1996 Cases: 1, 2, 5, 6, 7; Daskalakis & Nicolaides, 2002 Cases: 1, 2; Daskalakis et al., 2003; Deruelle et al., 2000 Cases: 2–4; D'Souza et al., 2004; Gajzer et al., 2015 Case 1; Gazolla et al., 2014 Cases: 2, 3, 5, 7, 8; Ginsberg et al., 1997; Gulczyński et al., 2019 Cases: 4, 9, 10; Hartwig et al., 1989 Case 4; Higuchi et al., 2013; Iba et al., 2016 Cases: 1, 2; Jun et al., 1991; Litwin et al., 1988 Case 2; Mandrekar et al., 2014; Maruyama et al., 2015; Mathai et al., 2009; Moerman et al., 1992 Case 7; Negishi et al., 1998 Cases: 1, 3, 4, 7; Palacios & Rodriguez, 1990; Patten et al., 1986 Cases: 1, 3, 5, 12, 13; Paul et al., 2001; Prasun et al., 2008; Pumberger et al., 2001 Cases: 3[a], 4; Routhu et al., 2016 Cases: 7, 8; Russo et al., 1993 Cases: 1–7; Sahinoglu et al., 2007 Cases: 2, 3; Smrcek et al., 2003 Cases: 5–8, 10; Tang et al., 1991[a]	90/189 47.61%
OVERLAPPED CR/AB	SLA LBWC	Bugge, 2012 Cases: 9, 10; Chen, 2001; Gajzer et al., 2015 Case 2; Gulczyński et al., 2019 Case 1; Hacivelioglu & Tarim, 2010 Case 1; Hunter et al., 2011 Case 4; Managoli et al., 2003; Negishi et al., 1998 Case 5; Patten et al., 1986 Case 4; Pumberger et al., 2001 Cases: 1, 2; Quijano et al., 2014; Rachad et al., 2012; Sahinoglu et al., 2007 Case 4	15/189 7.93%
	NSLA	Bugge, 2012 Cases: 7, 14; Chikkannaiah et al., 2013; Crespo et al., 2012 Case 1; Deruelle et al., 2000 Case 1; Devi et al., 2015; Gajzer et al., 2015 Case 4; Gulczyński et al., 2019 Cases: 5, 8, 11; Halder, 2010 Case 1; Hartwig et al., 1989 Case 1; Kocherla et al., 2015; Litwin et al., 1988 Case 1; Moerman et al., 1992 Cases: 9, 10, 14, 15, 17, 18; Negishi et al., 1998 Cases: 6, 8; Patten et al., 1986 Case 6; Saritha et al., 2013; Smrcek et al., 2003 Case 3; Stein et al., 2009; Zeidler et al., 2014	27/189 14.28%
NO DEFINED		Hirokawa et al., 2003; Smrcek et al., 2003 Cases: 1, 2, 4, 9	5/189 2.64%
SPDYS	SLA LBWC	Coleman et al., 2018 Case 2; Negishi et al., 1998 Case 2; Patten et al., 1986, Case 9	3/189 1.58%
	NSLA	Bhat et al., 2016 Cases: 1–3; Bugge, 2012 Cases: 6, 14; Coleman et al., 2018 Cases: 3, 4, 6; Colpaert et al., 2000 Case 2; Craven et al., 1997 Cases: 2–5; Cusí et al.,1996 Cases: 1, 2, 5–7; Daskalakis & Nicolaides, 2002 Case 1; Deruelle et al., 2000 Case 2–4; Devi et al., 2015; Gajzer et al., 2015 Case 1; Gazolla et al., 2014 Cases: 2, 7, 8; Ginsberg et al., 1997; Gulczyński et al., 2019 Case 8; Kocherla et al., 2015; Mandrekar et al., 2014; Negishi et al., 1998 Cases: 3, 4, 6; Palacios & Rodriguez, 1990; Patten et al., 1986 Cases: 3, 5, 7, 13; Russo et al., 1993 Cases: 1–3, 5–7; Smrcek et al., 2003 Cases: 5, 7; Tang et al., 1991	48/189 25.39%

Note: ABDOMINAL: fetus with body wall anomaly, lower limb anomaly, anal atresia or urinary and/or genital anomaly. CRANIAL: fetus with body wall anomaly, upper limb anomaly and/or craniofacial anomaly, and/or diaphragm anomaly or other thoracic anomalies (heart or lungs anomalies). OVERLAPPED CR/AB: fetus with body wall anomaly, craniofacial anomaly, anal atresia and/or urinary and/or genital anomaly. SPDYS: fetus that additionally presented some kind of spinal dysraphism.

Abbreviations: BSA, body stalk anomalies; CR/AB, cranial abdominal; LBWC, limb body wall complex; NSLA, nonstructural limb anomaly or no limb anomaly; SLA, structural limb anomaly; SPDYS, spinal dysraphism.

[a]Sirenomelia.

OEIS/LBWC in one case, LBWC/ABS in one case, BSA/ABS in eight cases, LBWC/Cantrell in one case, LBWC/BSA/AB in one case, OEIS BSA/LBWC in one case, and for three cases no specific diagnosis was given. In addition, some cases which did not show body wall anomaly were diagnosed as LBWC but not as BSA (Gazolla et al., 2014 case 1; Kruszka et al., 2015 case 2; Moerman, Fryns, Vandenberghe, & Lauweryns, 1992 cases: 11, 12, and 13; Sahinoglu et al., 2007 case 5) and some others as ABS (Routhu, Thakkallapelli, Mohan, & Ahmed, 2016 cases 1 and 2). ABS may include body wall anomalies, cranial anomalies, and limb amputation. The presence of the characteristic anomalies is enough for ABS diagnosis, even if bands are not present (Martín-Alguacil & Avedillo, 2020c). The presence of amniotic bands was described in 47 of the studied cases, from which 17 cases showed structural limb anomalies, 29 nonstructural limb anomalies, and one case with no limb anomalies (Table 1). Three different phenotypes were observed in the studied cases (cranial, abdominal, and cranial/abdominal phenotypes) and presented on Table 3. There were several cases: Hirokawa et al. (2003), and Smrcek et al. (2003) cases 1, 2, 4, and 9 that did not show specific cranial or abdominal anomalies and were not considered on Table 3. The higher incidence of SPDYS in the BSA classified cases (26.98% 51/189), made this author to consider an additional group called the SPDYS phenotype. Myelomeningocele was the most frequent anomaly among SPDYS described 80.3% (41/51). Most cases presenting SPDYS did not show structural limb anomalies, except for three cases that were classified as LBWC/SPDYS (Table 3). Additionally, 78.43% (40/51) of the SPDYS cases presented abdominal phenotype, 1.96% (1/51) cranial phenotype, 15.68% (8/51) CR/AB overlapped phenotype, and 2SPDYS cases did not show specific phenotype. The phenotype in the ABS classified cases was 11 abdominal, five cranial, three CR/AB overlapped phenotype, one CR/SPDYS, and two ABS cases did not show specific phenotype. All cases classified in this study as ABS were considered as LBWC in their original report (Gajzer et al., case 3; Kamudhamas & Manusook, 2001 case 1; Kruszka et al., 2015 cases: 1 and 2; Moerman et al., 1992 cases: 11–13; Sahinoglu et al., 2007 case 5; Vujovic et al., 2017), or as LBWD (Gazolla et al., 2014 cases 1 and 6), except for cases 1, 2, and 9 which were reported as ABS by Routhu et al. (2016).Case 3 reported as LBWC by Gajzer, Hirzel, Saigal, Rojas, and Rodriguez (2015) showed gastroschisis, normal UC, spinal anomaly, and clubfoot, thus it was not classified as BSA or LBWC. Gastroschisis is a full-thickness abdominal wall occurring lateral to the UC, and, in most cases, is an isolated anomaly, but sometimes may occur in combination with other congenital anomalies as arthrogryposisin the hindlimbs (Martín-Alguacil & Avedillo, 2020b). Stein, Haller, Hawighorst, and EmonsGöttingen (2009) reported a case with thoracoabdominoschisis, no spinal anomaly, normal UC, and nonstructural limb and craniofacial anomalies. This case was diagnosed in the original article as a complex malformation sharing features of LBWC and Cantrell Syndrome. Considering the studies of our group in the pig (Martín-Alguacil & Avedillo, 2020b, 2020d), the case is herein classified as BSA VI (Table 2).

The proposed classification is based on anatomical features and on presumptive etiology of the limb anomalies as unifying criterion for a precise diagnosis and classification of CBWA when limbs are affected as in BSA, LBWC, and/or ABS.

CONFLICT OF INTEREST

The author have no potential conflict of interests.

AUTHOR CONTRIBUTIONS

Nieves Martín-Alguacil is the only author and contributor to this manuscript. She is responsible for the conception and design, analysis, and interpretation of data. And also responsible for drafting and revising the manuscript for intellectual content. She gave the final approval of the version to be published. And agreed to be accountable for all aspects of the work.

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