http://dx.doi.org/10.2106/JBJS.19.01056

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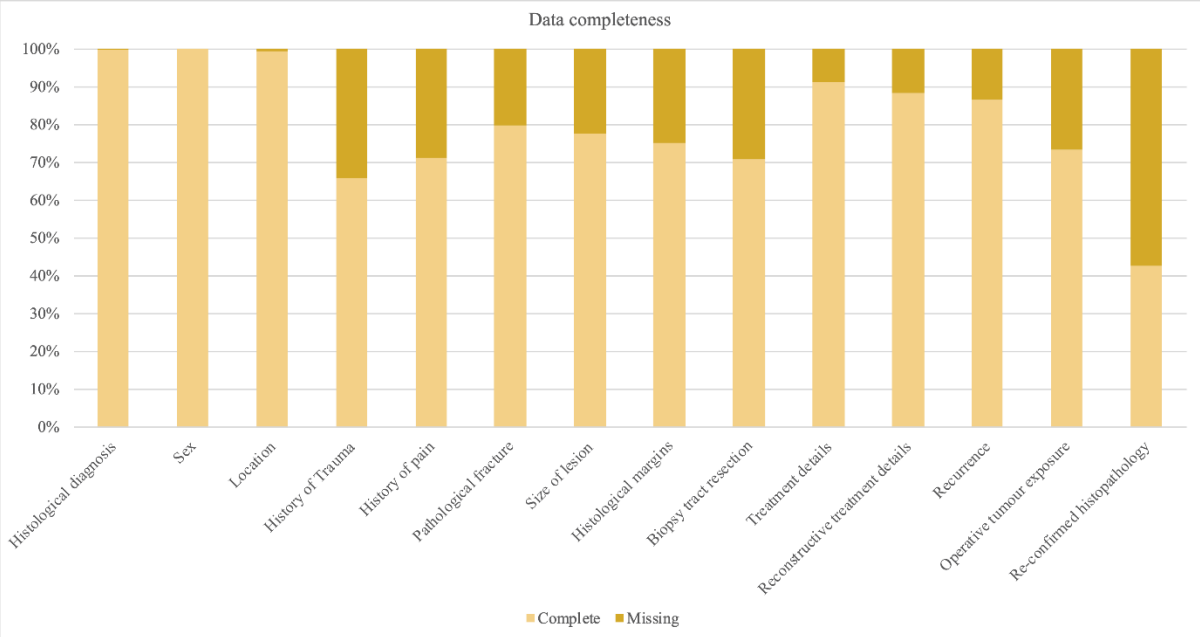
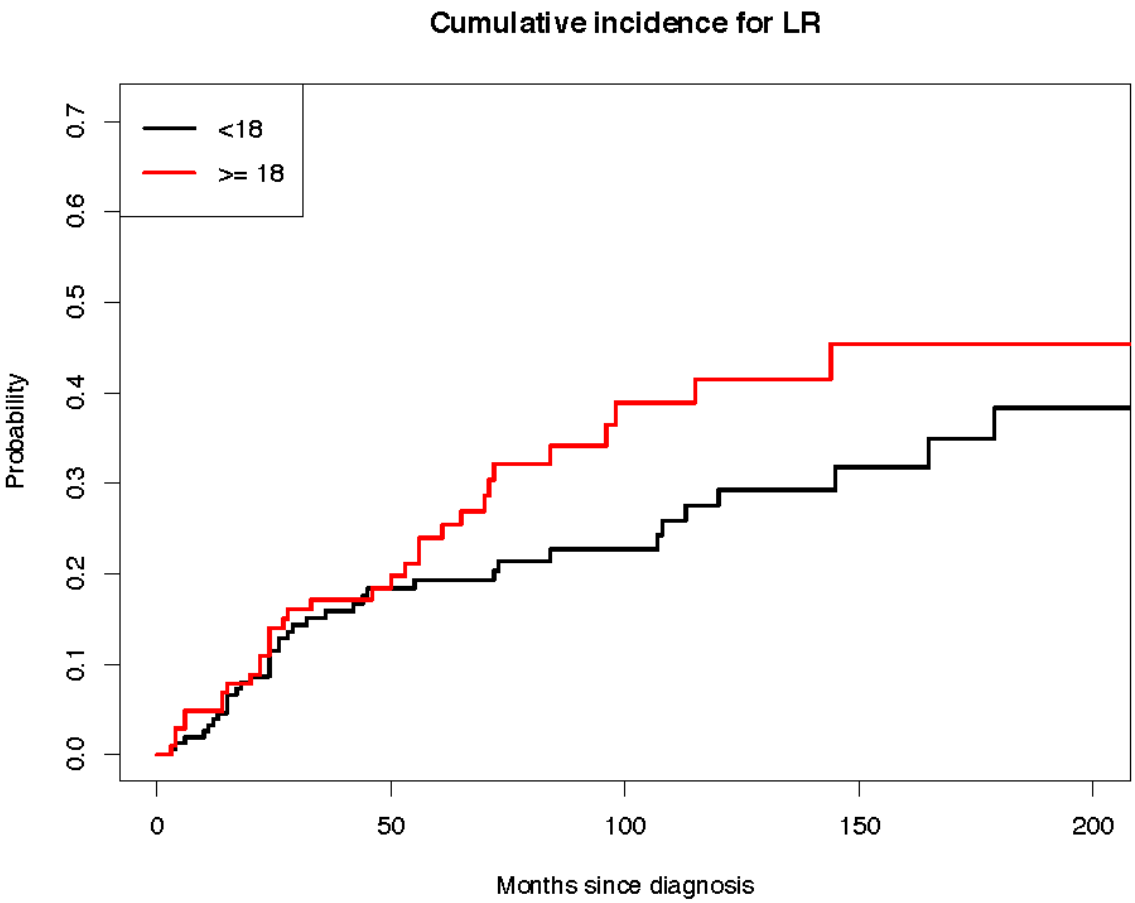
APPENDIX

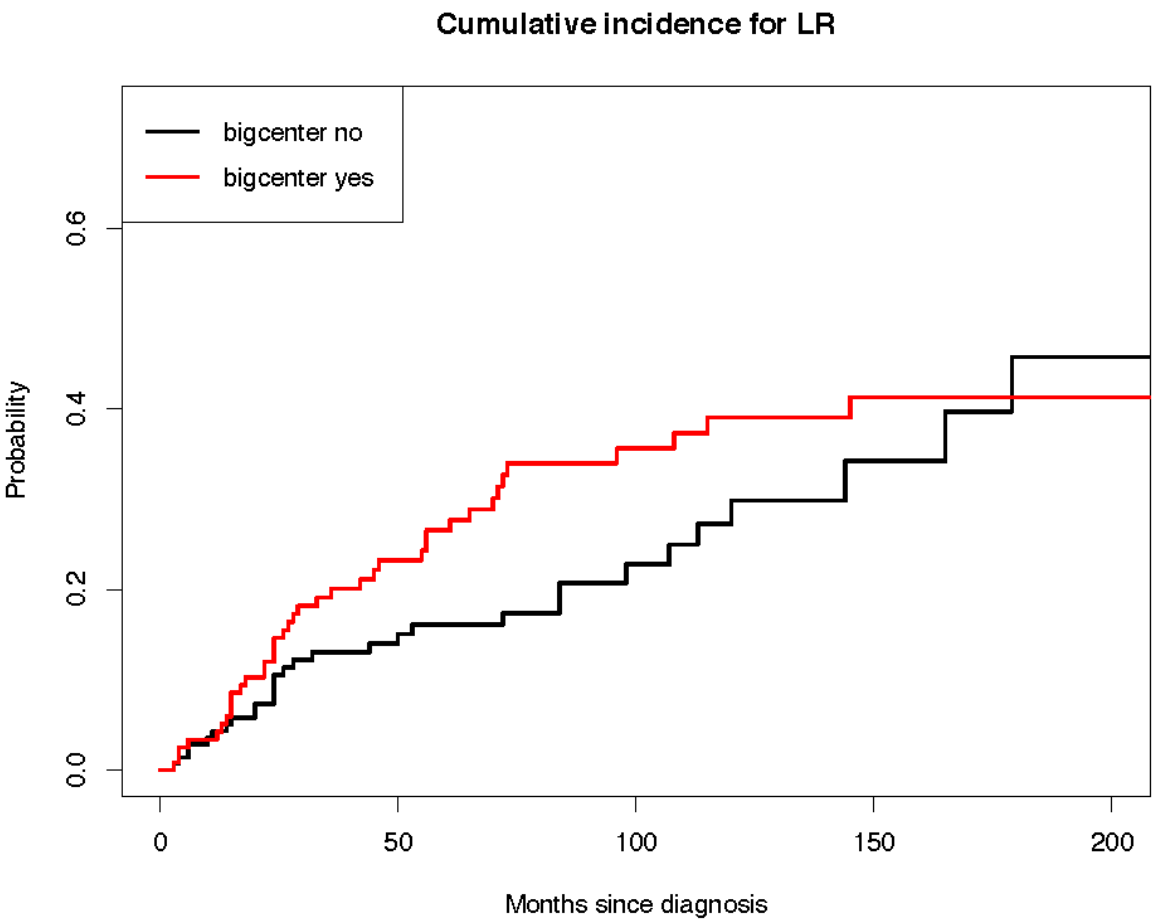


Appendix 1 Proportion of data missing per variable.

http://dx.doi.org/10.2106/JBJS.19.01056

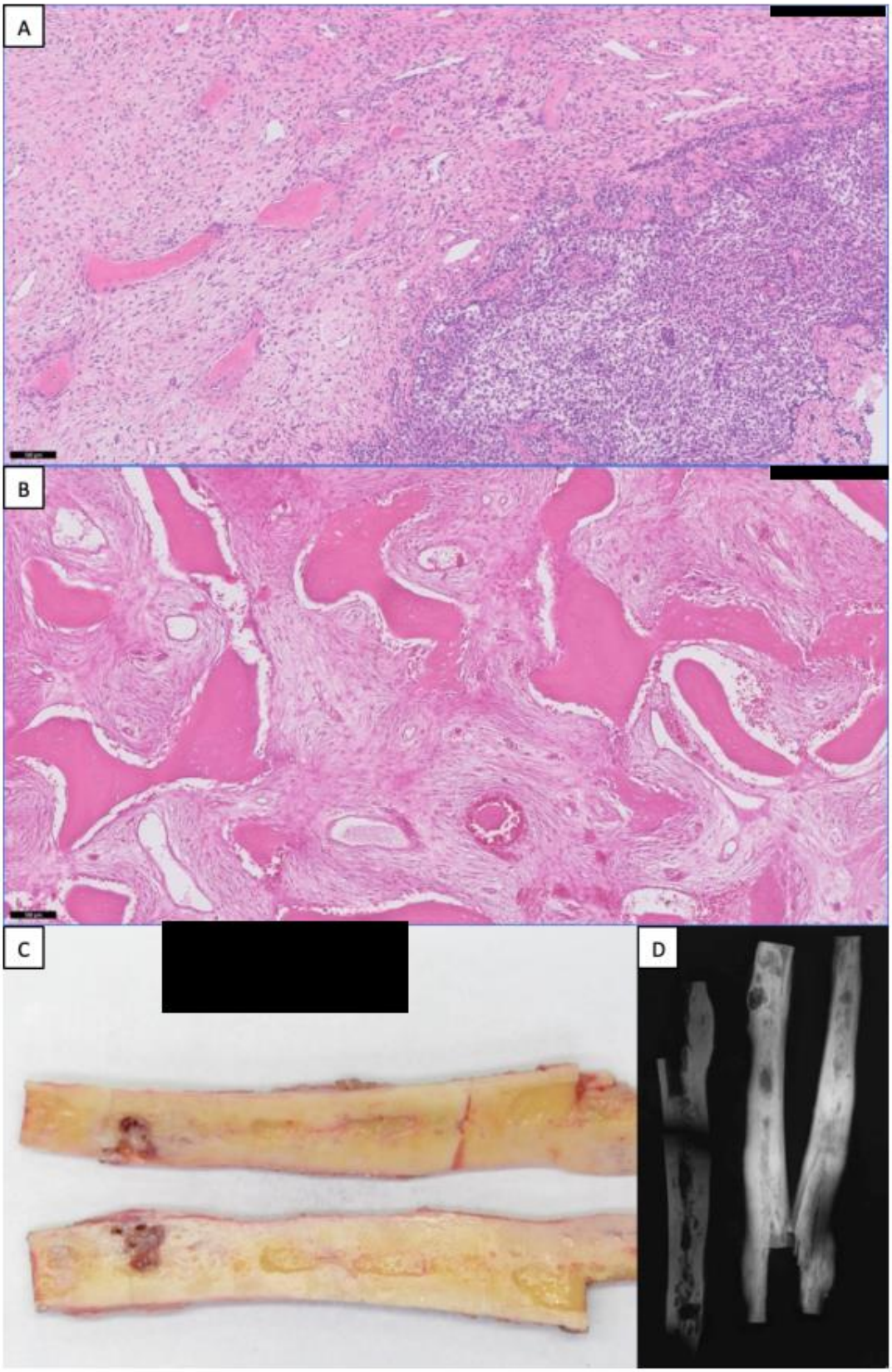


Appendix 2 Unadjusted cumulative incidence stratified per age group of the patients in OFD-AD and AD.



Appendix 3 Unadjusted cumulative incidence for LR for cases treated at centers with more or less than 20 submissions (arbitrary distinction).

http://dx.doi.org/10.2106/JBJS.19.01056



http://dx.doi.org/10.2106/JBJS.19.01056

Appendix 4. H&E stain of AD located within an area of OFD-AD obtained in 2016 (A). Histology obtained in 1987 demonstrating OFD-AD (B). Specimen resected in 2016 from which both slides A&B were obtained (C). Radiograph of resection specimen (D). Black scale bar equals 100μm.

Table E1 Univariable analysis of multiple factors for LR.

Variable	Available numbers for analysis	HR	0.95 CI	P value
Sex, female vs male	258	0.568	0.356-0.907	0.018*
Size, > 5cm vs <= 5cm	225	0.989	0.572-1.708	0.968
Pathological fracture, yes vs no	228	1.947	1.129-3.359	0.017*
Uncontaminated resection margins (R0), yes vs no	233	0.205	0.121-0.347	<0.001*
Under 18 years of age, yes vs no	258	1.428	0.900-2.263	0.130
Classic adamantinoma vs OFD-AD	258	1.254	0.781-2.012	0.349
Intralesional resection (R2)$^{\Sigma}$, yes vs no	216	4.179	2.381-7.333	<0.001*
High volume centre$^{\Omega}$, yes vs no	258	1.413	0.885-2.256	0.148
Operative resection margin narrow (R0)$^{\Psi}$, yes vs no	184	0.399	0.212-0.751	0.004*
Operative resection margin wide (R0)^{Z}, yes vs no	184	0.147	0.079-0.272	<0.001*

$^{\Sigma}$Intralesional is defined as surgery where en-bloc resection was not attempted. This includes curettage and surgeries where macroscopic tumour was left (R2).

$^{\Omega}$Centres that have submitted more than 20 (arbitrary) cases.

$^{\Psi}$Where the histopathological margins were described as R0 but narrow (arbitrary)

^{Z} Where the histopathological margins were described as R0 but wide (arbitrary)

Table E2. published datasets of adamantinomas of the long bones.

Author	Year	Histological diagnosis (*n*)	Mean age (years)	Mean tumor size (cm)	Mean follow-up (months)	Local Recurrence % (*n*)	Metastatic Disease % (*n*)	Fatal Disease % (*n*)	Remarks	Reference
Zumarraga et al[18] (Sao Paulo, Brazil)	2018	7 classic AD	28.5 (17-49)	9.16 (4.1-12.8)	180 (36-324)	0%	Classic AD 28.6% (2)	Classic AD 0% (0)		(15)
Houdek et al[19] (Rochester, USA)	2018	46 classic AD	24 (7-79)	7 (1-17)	192 (36-504)	7.5-15%	Classic AD 26.1% (12)	Classic AD 21.7 % (10)		(16)
Scholfield et al[7] (Birmingham, United Kingdom)	2017	21 classic AD 10 OFD-AD	Classic AD 38 (14-86) OFD-AD 13.4 (6-28)	Not reported	Classic AD 139 (4-396) OFD-AD 118 (36-315)	Classic AD 28.5% (6) OFD-AD 30% (3)	Classic AD 42.8% (9)	Classic AD 33.3 % (7)	Classic AD included 4 cases of Ewing's like Adamantinoma.	(7)
Puchner et al[16] (Vienna, Austria)	2016	10 classic AD 1 OFD-AD	Classic AD 28 (5-62) OFD-AD 26	Not reported	Classic AD 232 (48-564) OFD-AD 60	Classic AD 40% (4) OFD-AD 0% (0)	Classic AD 20% (2)	Classic AD 10% (1)		(17)
Szendroi et al[17](Budapest, Hungary)	2009	11 Classic AD	Classic AD 29 (4-80)	14.3 (2-16)	Classic AD 152 (36-480)	Classic AD 36% (4)	Classic AD 18% (2)	Classic AD 9% (1)		(18)
Gleason et al[20] (Boston, USA)	2008	3 Classic AD 5 OFD-AD	Classic AD 16.3 (13-18) OFD-AD 13.6 (9-17)	Not Reported	Classic AD 156 (108-216) OFD-AD 94 (30-144)	Classic AD 0% (0) OFD-AD 0% (0)	Classic AD 33.3% (1)	Classic AD 33.3 % (1)		(19)

Desai et al[21] (Mumbai, India)	2006	12 Classic AD	Classic AD 30 (18-65)	5 (maximum 11)		Classic AD 25% (3)	Classic AD 8.3% (1)	Classic AD 0% (0)	1 patient lost to follow-up after diagnosis	(20)
Van Rijn et al[22] (Amsterdam, The Netherlands)	2006	6 Classic AD	Classic AD 7.7 (3-14)	Not Reported	Classic AD 72 (19-144)	Classic AD 0% (0)	Classic AD 0% (0)	Classic AD 0% (0)	Only reported on children. Included in this paper	(21)
Qureshi et al[10] (USA, Canada, Italy)	2000	70 Classic AD	Classic AD 31 (7-86)	Not Reported	Classic AD 84 (14-188)	Classic AD 18.6% at 10 years	Classic AD 10% (7)	Classic AD 12.8 % at 10 years	23 contributing centers	(11)
Kuruvilla et al[23] (New York, USA)	1998	5 OFD-AD	OFD-AD 8.3 (4.5-14)	Not Reported	OFD-AD 42-180	OFD-AD 60% (3)	OFD-AD 0% (0)	OFD-AD 0% (0)	No evidence of progression to classic AD, all cases treated y curettage	(22)
Jundt et al[24] (Swtizerland, Germany)	1995	23 Classic AD	Classic AD 25.4 (5-67)	Not Reported	Classic AD 72 (6-156) (19/23 cases)	Classic AD 21.7% (5)	Classic AD 13% (3)	Classic AD 13% (3)		(23)
Hazelbag et al[14] (Leiden, The Netherlands)	1994	25 classic AD 7 OFD-AD	Classic AD 28.7 (4-70) OFD-AD 22 (5-64)	Not Reported	122 (11-350)	Classic AD 24% (6) OFD-AD 42.8% (3)	Classic AD 32% (8)	Classic AD 28% (7)	OFD-Ad seen as a subtype of classic AD. Included in this paper	(13)
Czerniak et al[25] (New York, USA)	1989	17 Classic AD 8 OFD-AD	Classic AD 40 (15-65)	1-12 (combined)	Not Reported	Not Reported	Not Reported	Not Reported		(24)

			OFD-AD 11 (3-17)							
Keeney et al[1] (Rochester, USA)	1989	85 adamantinomas	25.9 (3-72)	Not Reported	108 (1-564)	31% (26)	15% (13)	13% (11)	Not clear if distinction was made between Classic AD and OFD-AD	(1)
Moon and Mori[3] (Japan, USA)	1986	195 adamantinoma	32.9 (4-74)	Not Reported	Not Reported	Not Reported	Not Reported	18% (36)	Meta-analysis, 180 cases from literature. Not clear if distinction was made between Classic AD and OFD-AD	(3)
Campanacci et al.[26] (bologna, Italy)	1981	9 adamantinomas	26.6 (8-57)	Not Reported	76 (12-264)	44% (4)	0% (0)	0% (0)	Not clear if distinction was made between Classic AD and OFD-AD	(25)
Weiss et al[27] (Baltimore, USA)	1977	9 adamantinomas	38 (15-65)	Not Reported	(0-120)	11% (1)	22% (2)	11% (1)	Not clear if distinction was made between Classic AD and OFD-AD	(26)
Huvos et al[28] (New York, USA)	1975	14 adamantinomas	40 (13-67)	Not Reported	144 (0-408)	71% (10)	14% (2)	14% (2)	10/14 patients treated with curettage initially.	(27)

									Not clear if distinction was made between Classic AD and OFD-AD	
Baker et al[29] (Rochester, USA)	1954	27 adamantinomas	31 (12-57)	Not Reported	134 (12-276) For only 8 cases	55.6% (15)	29.6% (8)	22.2% (6)	24 cases from literature. Not clear if distinction was made between Classic AD and OFD-AD	(28)
Total Classic AD OFD-AD Adamantinomas*		251 36 339	7.7-38 8.3-26 25.9-40			0-40% 0-60% 11-71%	0-42.8% 0% 0-29.6%	0-33.3% 0% 0-22.2%		