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ORIGINAL ARTICLE

Analysis of shoulder motion with inertial sensors in Poland syndrome patients

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Abstract

Purpose Poland syndrome is a congenital malformation characterized by agenesis or hypoplasia of pectoralis muscles. There is a limited literature on how the anatomic anomalies of PS may impact the movement of the shoulder. This study analyzes the effects of absence of the pectoralis muscles on the shoulder kinematic.

Methods Clinical evaluation was performed analyzing range of motion (RoM), stability, cuff disease and internal rotation strength. In all patients, we used inertial sensors to analyze scapular motion in three degrees of freedom: medium-lateral rotation, posterior tilting and protraction-retraction. The same analysis was performed by dividing the patients into two groups by age to evaluate the presence of age-related alterations.

Results No differences in RoM between pathological and healthy side were observed. All patients were positive for posterior instability. No significant differences in strength in internal rotation were observed with average + 6,91% (s = 2,14) on the healthy side's strength. Kinematic analysis showed higher values of scapular medium-lateral rotation and anticipation of retraction of the pathological side during flexion and abduction. Reduced scapular tilt in under 18 years old was found.

Conclusions The absence of the pectoralis muscles seems not to affect the RoM. The increased scapular rotation on the medium-lateral axis is probably due to the absence of humeral insertion of the pectoralis major and the absence of the scapular insertion of the pectoralis minor. The increased retraction in abduction it can be explained by a hypercontraction of the scapular stabilizers. The reduced tilt in under 18 years old is influenced by the lack of adaptation by the muscle groups involved.

Keywords Poland syndrome · Pectoralis muscle · Shoulder kinematic · Scapular motion

Introduction

Poland syndrome (PS) is a rare congenital malformation characterized by agenesis or hypoplasia of pectoralis major muscle. The most common feature is the absence of the sternocostal head of the pectoralis major muscle with concomitant hypertrophy of the clavicular head and the absence of the pectoralis minor; other manifestations often associated are ipsilateral hand and upper limb anomalies, such as syndactyly, absence/hypoplasia of the breast and rib aplasia (Fig. 1) [1–4].

Although the syndrome usually occurs unilaterally, some cases of bilateralism have been observed [5–7]. The incidence of Poland syndrome has been reported to be approximately one in 30,000 live births [8], with a higher prevalence in males (M/F 3:1) and with the right side of the pectoralis being affected more frequently [3, 7]. The etiology underlying Poland syndrome is still unknown. A vascular disruption due to a hypoplasia of subclavian artery during the 6th week of gestation remains the most accepted pathogenic mechanism. The theory suggests that the interruption of the embryonic blood supply causes hypoplasia of the ipsilateral subclavian artery or one of its branches. This results in muscle absence and variable clinical manifestations in areas supplied by the artery [9]. Other risk factors that seem

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Fig. 1 Patient with Poland syndrome

to be involved are smoking or cocaine abuse in pregnancy [10–12]. The diagnosis of Poland syndrome is still mostly clinical. The unilateral agenesis or hypoplasia of the pectoralis major muscle is assessed by a clinical test, asking to the patient to push the palms of the hands against each other with the arms flexed anteriorly. Most patients live a normal life, carrying out daily activities and even practicing sports and exercises.

In most cases, reconstructive surgery is required by patients only for cosmetic reasons: In fact, during the pubertal development, the asymmetry of the thoracic region in males or of the mammary region in females become more evident and can cause discomfort, leading to a reduction in the quality of life [13–15].

Functional limitations of these patients are usually minimal, as demonstrated by the presence of Poland syndrome in athletes [16]. Actually, there is a limited literature on how the anatomic anomalies of PS may impact the movement of the shoulder both functionally and biomechanically.

The study of shoulder range of motion (RoM) using wireless inertial motion capture devices showed a high accuracy and reliability [17–19]. Currently, though, there are no studies on the impact of the Poland syndrome on the biomechanics and the ROM of the shoulder.

Aim of this study was to analyze the effects of the absence of the pectoralis major and minor muscles on the kinematic of glenohumeral and scapulothoracic joints in the life span, among a cohort of patients affected by Poland syndrome. The hypothesis was that even if the RoM of the shoulder do not differ, scapular motion on the pathological side significantly differ from healthy side. The research evaluated clinical aspect such as range of motion (RoM), joint instability, muscle compensation, rotator cuff alteration and the possible alteration of the scapular kinematics by motion sensors acquisitions.

Materials and methods

Patient population

The current study enrolled 24 Caucasian patients affected by Poland syndrome homolaterally, with a clinical and instrumental diagnosis of complete absence of pectoralis major and minor muscles. Fourteen patients (58.3%) were affected on their dominant side. Nine patients were females and 15 males, aged between 12 and 53 years old (average 27 years old). Four patients had an associated hand abnormality, syndactyly, that do not modify the RoM of the shoulder. Only eight patients (33.3%) underwent to surgical treatment for cosmetic defects. Almost all the patients were playing sports (91.6%), with three of them performing at a competitive level.

Clinical evaluation

Two orthopedic surgeons examined all the patients in order to assess: (1) RoM of the glenohumeral joint comparing both sides during anterior flexion and lateral abduction using goniometer; (2) stability of the affected shoulder versus contralateral healthy one, with Anterior Apprehension test [20], Posterior Apprehension test [21], Porcellini test [22]; (3) functional evaluation of rotator cuff with clinical test such as Jobe Test [23], Patte test [24], Lift-off test [25] and Belly-press test [26] and (4) comparison of the strength between the pathological side and the healthy side in internal rotation using the isokinetic dynamometer TechnoGym REV 9000 (TechnoGym SpA, Gambettola, Forlì-Cesena, Italy).

Kinematic evaluation with sensors

After the initial clinical assessment, all patients were evaluated with NCS Company (Carpi, Italy) ShowMotion® wireless inertial sensors to analyze the scapular motion patterns. These sensors measure, record and compare movements, providing a visual representation of the scapular motion in real time. The technology combines wireless motion tracking sensors and a dedicated biomechanical model to check the acquired movement patterns of the patients. Previous studies had already adopted this solution in the clinical setting to evaluate scapular-humeral rhythm with encouraging results [27]. Multi-inertial measurement units (MIMUs) (WISE, NCS Lab, Carpi, Italy) are used as wireless motion tracking sensors. Each inertial unit provides both raw data (accelerometer, magnetometer and gyroscope) and the orientation matrix, representing the local system of reference (SoR) orientation concerning a fixed SoR. The signals measured by the sensors, positioned in specific body parts of the patient, were used to obtain a real-time measurement of 3D kinematics through the use of a dedicated biomechanical protocol.

The embedded biomechanical protocol allows to analyze the 3D kinematic of the main joints of the upper limb (shoulder and elbow) using magnetic and inertial sensors. Moreover, the protocol was able to differentiate the contributions of the glenohumeral and scapulothoracic joints during humerus elevation.

Subjects were asked to move the shoulder through an active range of motion activities following a specific evaluation protocol (that includes flexions in the sagittal plane and abduction–adduction in the coronal plane). The first motion was anterior flexion with thumbs up, until patient's maximum available range without pain. The second motion was abduction in the scapular plane to patient's maximum available range without pain. A bilateral evaluation was conducted simultaneously.

Data from each MIMU were analyzed with a dedicated software. Five inertial sensors were placed: one sensor on the sternum, one sensor on each arm, one sensor on each scapula above the spine, 2 cm medial to the acromion. First, we calibrated the platforms according to the device instructions, then proceeded with the acquisition. We asked the patient to make five consecutively complete shoulder flexion and five consecutively shoulder abduction.

During the two sequences, the system recorded the scapular movements in his three degrees of freedom (DoF): medium-lateral rotation ("MELA"), tilting and protraction/retraction ("Pr/Re").

The relationship between humeral movement and scapular kinematic was represented in graphs generated by the technology 'software.

The analysis was divided in two steps: First, the scapular movements of the affected side and the healthy side were evaluated and compared with a normal curve; second, then, we divided the group of patients by age (<18 years/> 18 years) to evaluate the presence of age-related alterations.

Analysis of the graphs was performed by all the researchers, together with an NCS Company engineer.

All the procedures performed in this study involving human participants were in accordance with the institutional ethical standards and with the Helsinki Declaration.

Results

Clinical evaluation

There were no significant differences in RoM between the pathological side and the healthy side during the anterior flexion (Pathological side 168,04° s = 5,45; healthy side 166,25° s = 5,18) and lateral abduction (pathological side 164,87° s = 4,33; healthy side 163,83° s = 4,27) movements, among all patients. During the clinical evaluation, none of the patients were found to be positive to the anterior apprehension test. In the tests for posterior shoulder instability, four patients resulted positive for the posterior apprehension test and all patients were positive for the Porcellini test (Morey et al., 2016). Test for the rotator cuff was negative in all patients examined. The evaluation of the internal rotation strength with the use of the isokinetic dynamometer did not show significant differences between the pathological side versus the healthy side. Only one patient out of 24 had a reduced strength on the pathological side. The difference in strength between dominant and non-dominant limb is considered non-pathological when less than 10%. Higher than 10% differences in strength between the pathological and the healthy side were never observed in this study, which sample of patients averaged + 6,91% (s = 2,14) on the healthy side's strength. During the clinical evaluation, we observed a greater trophism of latissimus dorsi muscle on the pathological side compared to the healthy side in all patients.

Kinematic analysis

In the first step, three planes of scapula movement (y-axis) during humerus flexion and abduction were analyzed, comparing pathological sides (mean 1 and 2 standard deviation) to the average of non-pathological sides. Then three planes of scapula movement were analyzed comparing average of pathological sides and the average of non-pathological sides versus healthy normality band obtained from previous clinical studies with Showmotion (mean 1 and 2 standard deviations colored in green).

- *MELA scapula pathological side versus non-pathological side*: We observed that scapula of the pathological side showed a superior rotation on the mid-lateral axis than the healthy side. (Fig. 2).

- *MELA scapula pathological side/non-pathological side versus normality band*: We observed that scapula of the pathological side showed a superior rotation on the mid-lateral axis than the healthy side and normality band. (Fig. 3).

- *Tilt scapula pathological side versus non-pathological side:* We observed that scapula of the pathological side showed a slightly lower posterior tilt during abduction than the healthy side. (Fig. 4).

- *Tilt scapula pathological side/non-pathological side versus normality band*: We observed that scapula of the pathological side showed too anterior tilt during first 40 degrees of humerus elevation, after 40 degrees, it has a trend similar to the healthy group. (Fig. 5).

- *Pr/Re scapula pathological side versus non-pathological side:* We observed that scapula of the pathological side

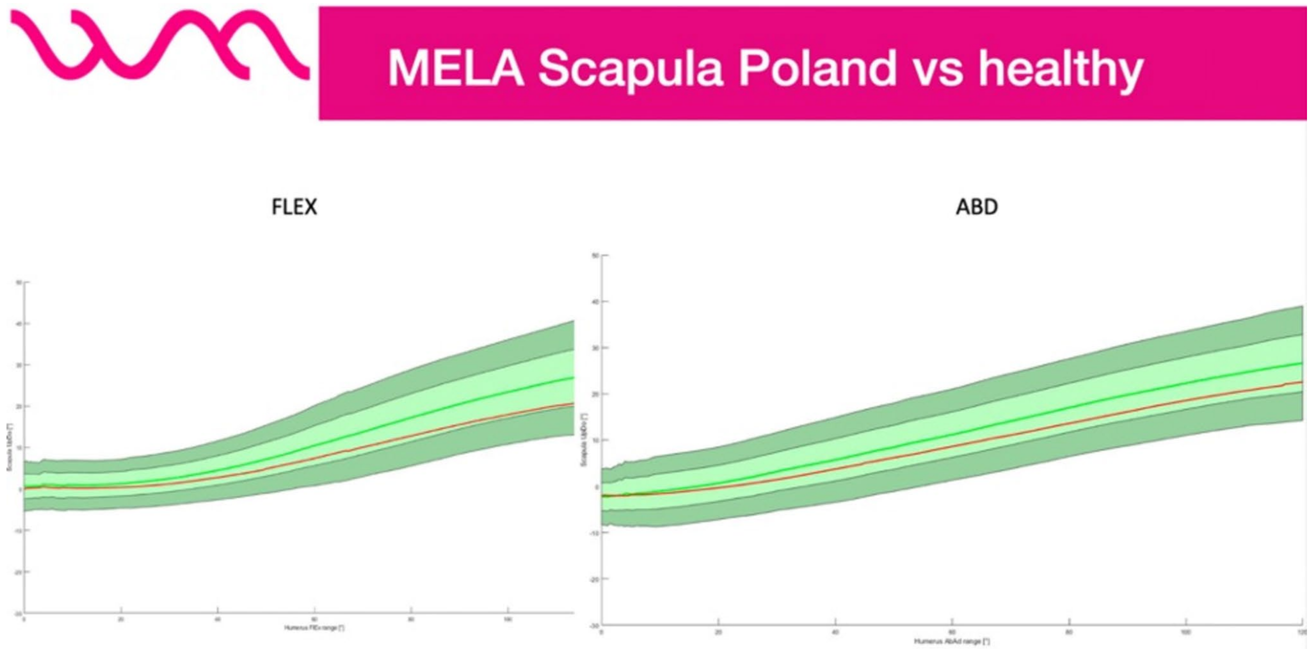

Fig. 2 The graph on the left represents the medium-lateral rotation of the scapula during sagittal elevation (x-axis = degree of humerus flexion range; y-axis = degree of scapula up-down); the graph on the right represents the medium-lateral rotation of the scapula during lateral abduction (x-axis = degree of humerus abduction range; y-axis = degree of scapula up-down). Scapula of the pathological side (green)/non-pathological side (red)

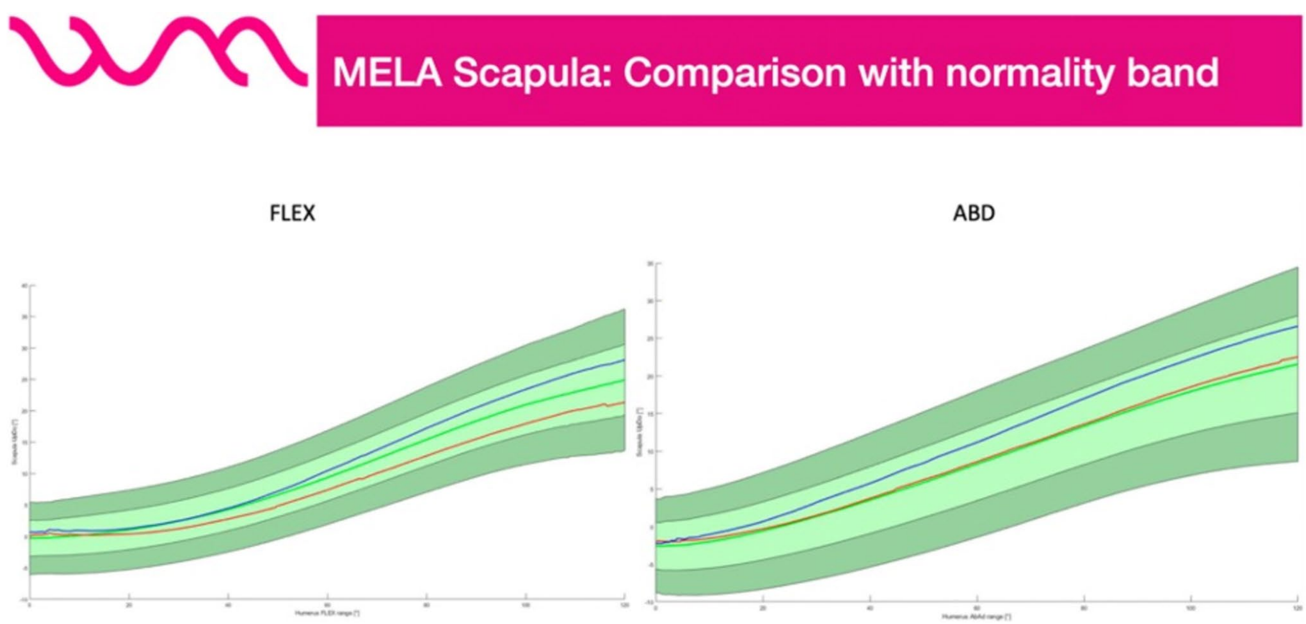

Fig. 3 The graph on the left represents the medium-lateral rotation of the scapula during sagittal elevation (x-axis = degree of humerus flexion range; y-axis = degree of scapula up-down); the graph on the right represents the medium-lateral rotation of the scapula during lateral abduction (x-axis = degree of humerus abduction range; y-axis = degree of scapula up-down). Pathological side (blue)/non-pathological side (red)/ normality band (green)

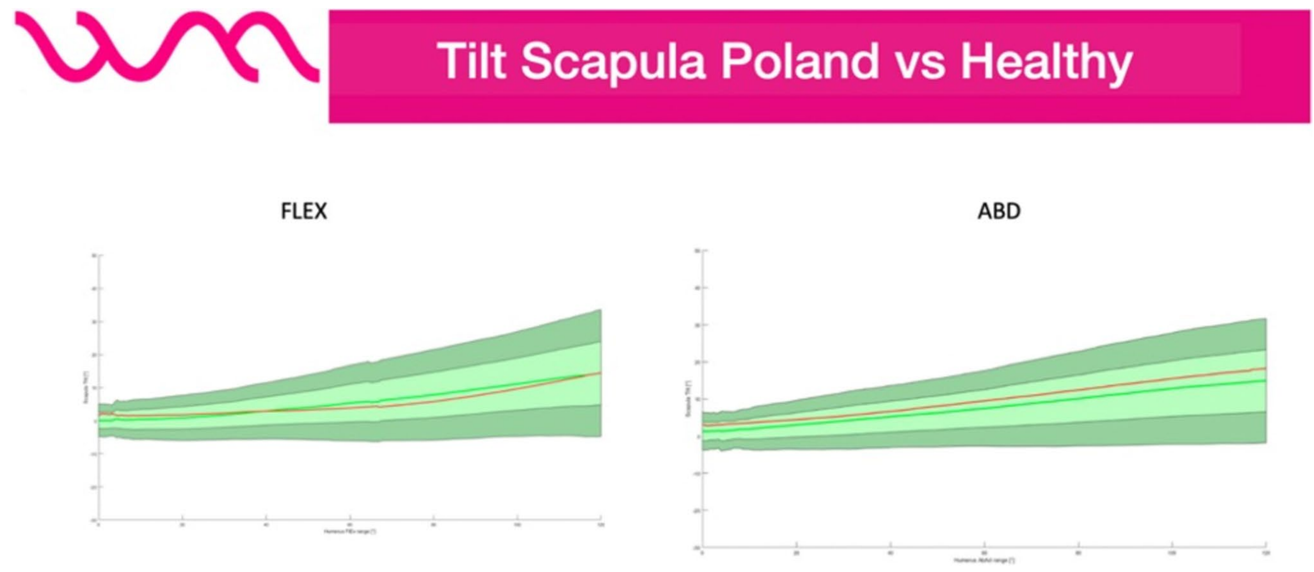

Fig. 4 The graph on the left represents the scapular tilt during sagittal elevation (x-axis = degree of humerus flexion range; y-axis = degree of scapular tilt); the graph on the right represents the scapular tilt during lateral abduction (x-axis = degree of humerus abduction range; y-axis = degree of scapular tilt). Pathological side (green)/non-pathological side (red)

(green) rotated more in external rotation than the healthy side (red). (Fig. 6).

- *Pr/Re scapula pathological side/non-pathological side versus normality band:* We observed that scapula of the pathological side (blue) rotated more in external rotation than the healthy side (red) and the normality band (green). (Fig. 7).

Tables 1 and 2 report average angles values and standard deviations for both flexion and abduction movements comparing pathological side to healthy side. The only statistically significant difference was found in medium-lateral rotation and protraction-retraction during abduction–adduction and anterior flexion. We observed higher values of medium-lateral rotation of the pathological side compared

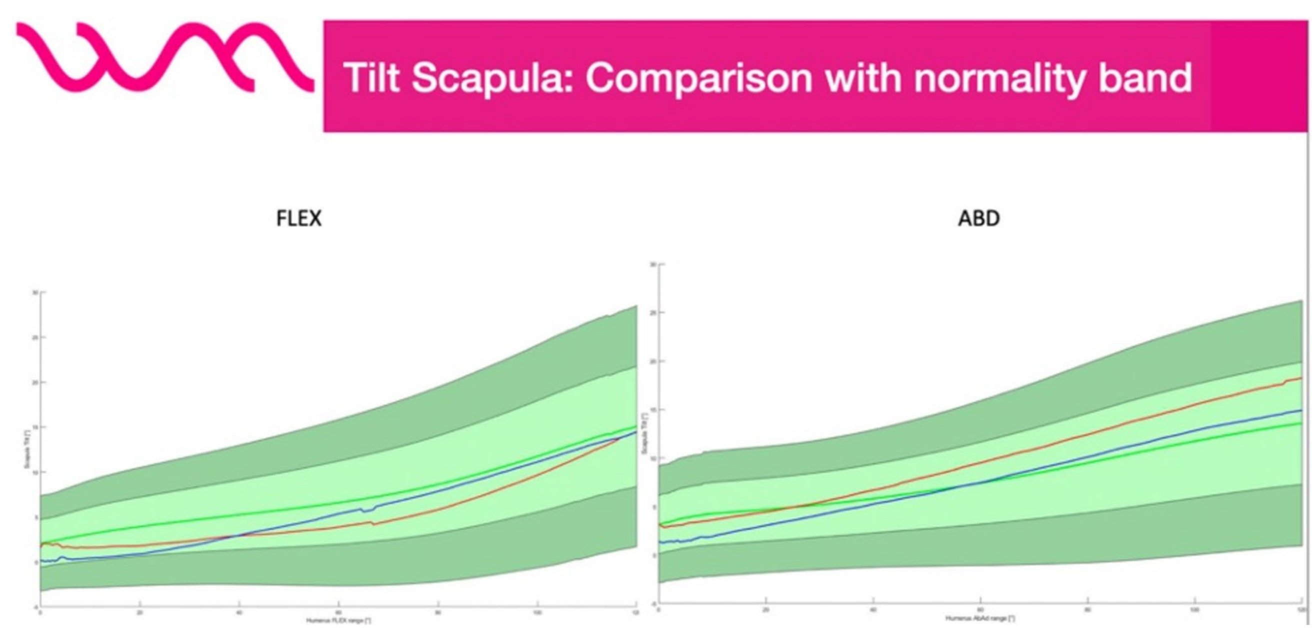

Fig. 5 The graph on the left represents the scapular tilt during sagittal elevation (x-axis = degree of humerus flexion range; y-axis = degree of scapular tilt); the graph on the right represents the scapular tilt during lateral abduction (x-axis = degree of humerus abduction range; y-axis = degree of scapular tilt). Pathological side (blue)/non-pathological side (red)/normality band (green)

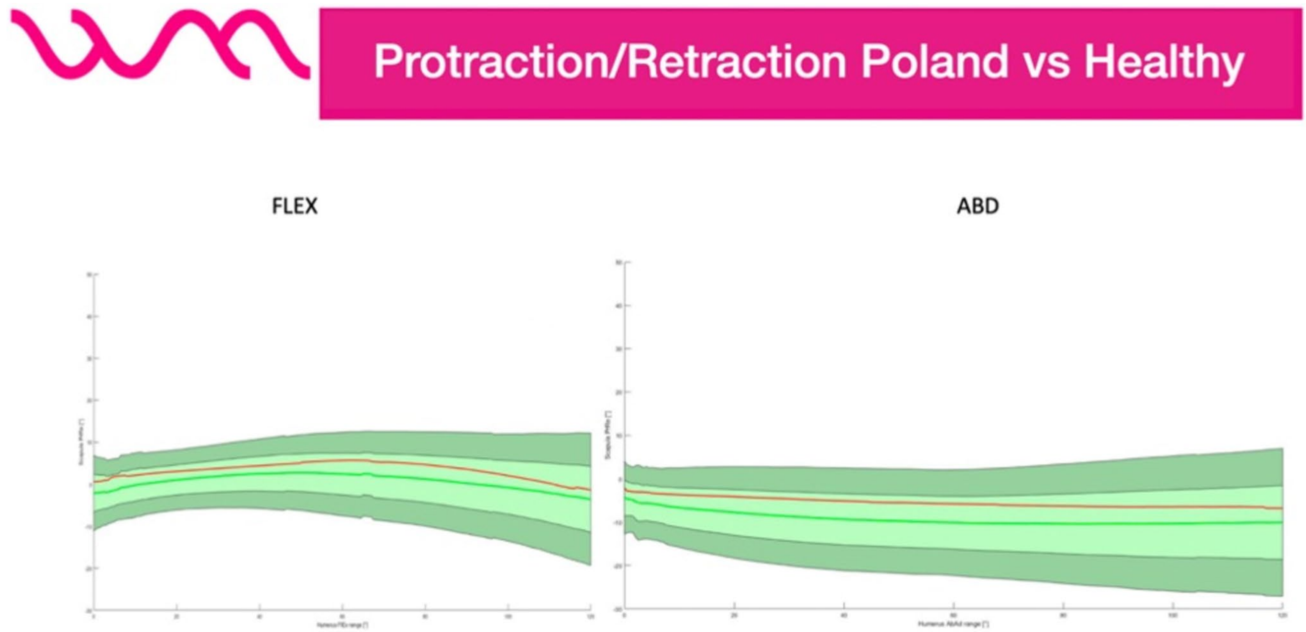

Fig. 6 The graph on the left represents the scapular protraction-retratction during sagittal elevation (x-axis = degree of humerus flexion range; y-axis = degree of scapular protraction and retraction); the graph on the right represents the scapular protraction-retraction during Lateral abduction (x-axis = degree of humerus abduction range; y-axis = degree of scapular protraction and retraction). Pathological side (green)/non-pathological side (red)

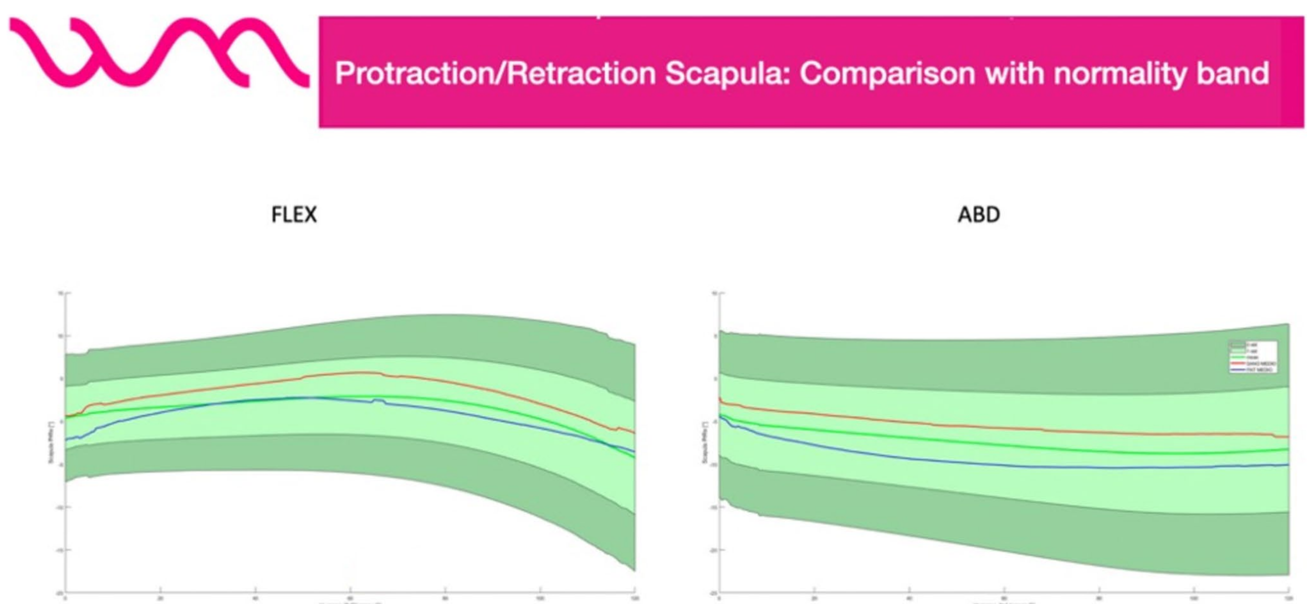

Fig. 7 The graph on the left represents the scapular protraction-retraction during sagittal elevation (x-axis = degree of humerus flexion range; y-axis = degree of scapular protraction and retraction); the graph on the right represents the scapular protraction-retraction during lateral abduction (x-axis = degree of humerus abduction range; y-axis = degree of scapular protraction and retraction). Pathological side (blue)/non-pathological side (red)/normality band (green)

to the healthy side appears to be a consistent trend throughout the range of motion (ROM). It is important to note that there is anticipation of retraction on the pathological side during both anterior flexion and abduction. As with posterior tilt, the average values are consistently lower throughout the abduction RoM, while they increase slightly during anterior flexion, but with no statistically significant difference.

Table 1 Representative values of the three scapular angles during abduction at four specific angulations (30°–60°–90° and 120°). The data are presented as mean ± standard deviation

Ab-Ad		0–30°		30–60°		60–90°		90–120°	
		Pathological	Healthy	Pathological	Healthy	Pathological	Healthy	Pathological	Healthy
Tilt	average	3,76	4,83	7,18	8,93	11,29	13,81	14,11	16,08
	SD	3,88	4,42	5,20	5,76	7,13	8,33	8,67	11,03
	p-value	0,225		0,086		0,073		0,249	
Up-Down	average	3,08	1,41	11,01	8,31	19,47	16,06	25,21	19,63
	SD	4,30	2,42	5,06	4,43	5,68	6,28	7,73	8,86
	p-value	0,017		0,001		0,002		0,001	
Pr-Re	average	-8,14	-3,79	-9,90	-5,25	-10,21	-6,26	-9,11	-5,22
	SD	5,67	6,88	6,07	7,90	7,37	8,34	7,61	7,21
	p-value	0,001		0,004		0,050		0,035	

Table 2 Representative values of the three scapular angles during flexion at four specific angulations (30°–60°–90° and 120°). The data are presented as mean ± standard deviation

Flex		0–30°		30–60°		60–90°		90–120°	
		Pathological	Healthy	Pathological	Healthy	Pathological	Healthy	Pathological	Healthy
Tilt	average	1,55	1,92	4,83	3,58	8,78	7,65	12,25	12,79
	SD	3,55	3,51	5,60	5,48	7,33	7,53	9,37	9,30
	p-value	0,582		0,253		0,417		0,750	
Up-Down	average	2,34	1,37	9,97	7,37	19,80	15,50	24,29	17,97
	SD	3,01	2,75	5,18	4,64	6,53	6,41	9,89	8,52
	p-value	0,123		0,019		0,006		0,001	
Pr-Re	average	1,94	3,88	2,51	5,56	0,50	3,76	-2,53	-1,08
	SD	3,29	4,23	4,66	5,45	5,82	7,07	7,33	8,07
	p-value	0,057		0,021		0,074		0,526	

In the end, we divided the sample of patients in two groups: under and over 18 years old (yo). We compared the average outcomes of pathological side in under 18 yo group and over -18 yo group versus previous normality bands. We analyzed separately the forward band (arm elevation) and backward band (arm lowering) trying to distinguish any defects.

We observed that the outcomes of over 18 group were similar to the normality bands (which contain adult people), but in under 18 group, the scapular tilt is less frequent during forward and backward than normality band (Figs. 8 and 9).

Discussion

Poland syndrome is a rare congenital malformation characterized by agenesis or hypoplasia of pectoralis major muscle. Functional limitations of these patients are usually minimal, as demonstrated by the presence of Poland syndrome in athletes [18].

The clinical analysis of the patients confirms that there are no substantial differences in ROM in flexion–extension and abduction–adduction between healthy and pathological side. Therefore, the absence of the pectoralis major and minor muscles seems not to affect the ROM of the shoulder. The constant presence of posterior shoulder instability found in the patients was likely due to the greater trophism of the posterior internal rotators like latissimus dorsi muscle observed during clinical evaluation which, not opposed by the action of the pectoralis major muscle, could favor a posterior translation of the humeral head. The rotator cuff was asymptomatic in the patients analyzed; therefore, the absence of the pectoral major and minor muscles does not cause functional deficits of the cuff. No differences in strength in internal rotation between the two sides were observed in most of the patients. This can probably be explained by the fact that there is sufficient compensation from the residual internal rotators, such as subscapularis and latissimus dorsi muscle.

The main aspects we found through kinematic analysis of the scapular movements of the healthy side and the

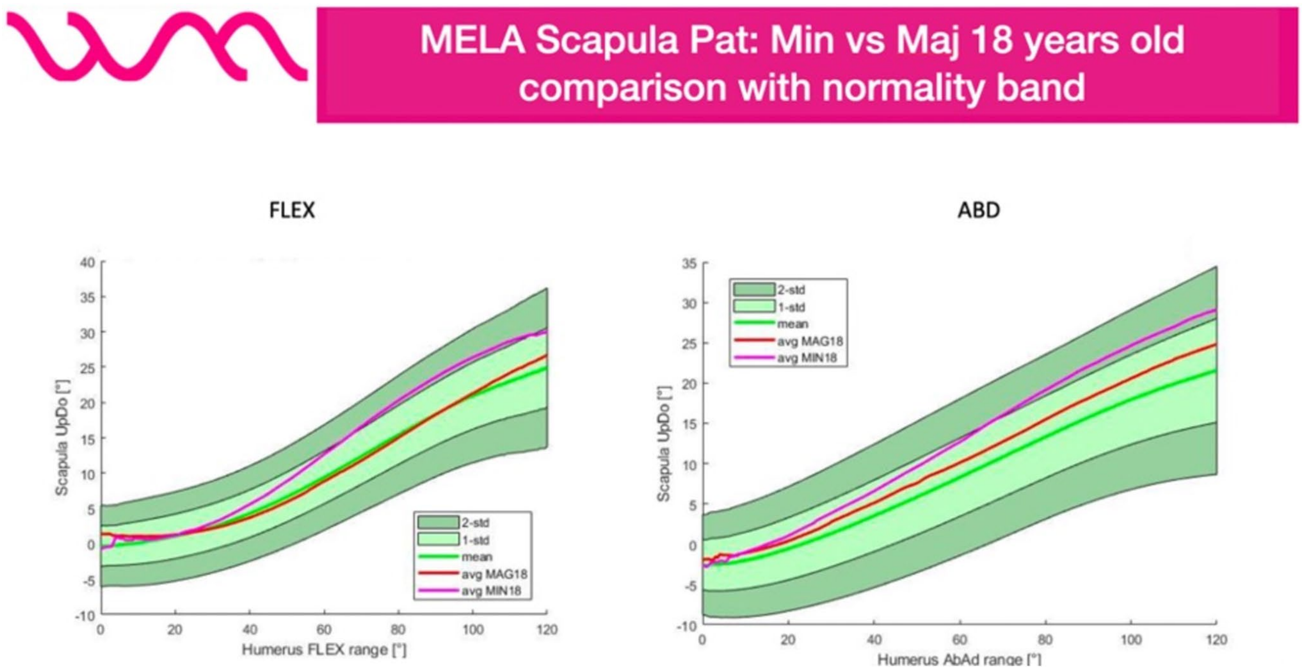

Fig. 8 The graph on the left represents the medium-lateral rotation of the scapula during sagittal elevation (x-axis = degree of humerus flexion range; y-axis = degree of scapula up-down); the graph on the right represents the medium-lateral rotation of the scapula during lateral abduction (x-axis = degree of humerus abduction range; y-axis = degree of scapula up-down). Pathological side over 18yo (red)/ pathological side under 18yo (pink)/normality band (green)

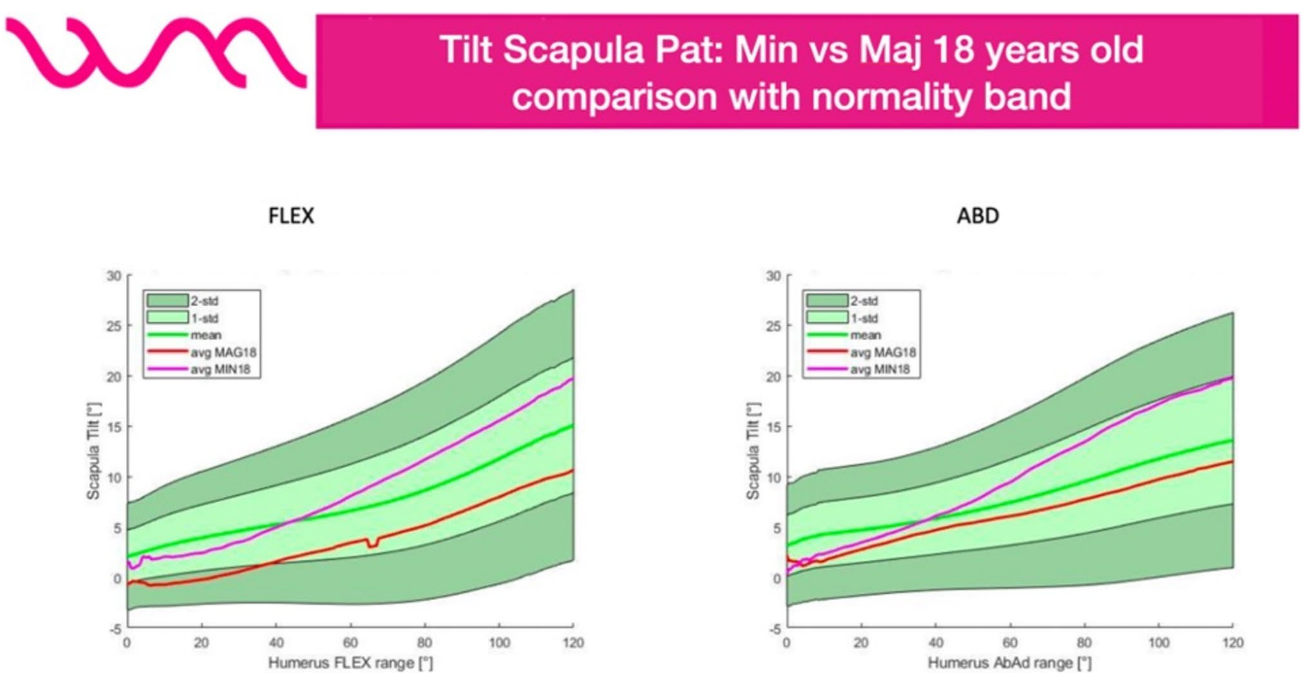

Fig. 9 The graph on the left represents the scapular tilt during sagittal elevation (x-axis = degree of humerus flexion range; y-axis = degree of scapular tilt); the graph on the right represents the scapular tilt during lateral abduction (x-axis = degree of humerus abduction range; y-axis = degree of scapular tilt). Pathological side over 18yo (red)/ pathological side under 18yo (pink)/normality band (green)

pathological side, it was noted a superior rotation on the medium-lateral axis in pathological side than the healthy side in both abduction and anterior flexion. We can hypothesize that it is due to the absence of humeral insertion of the pectoralis major and the absence of the scapular insertion of the pectoralis minor according to Borstad et al. [28]. The posterior tilt was lower in abduction on the pathological side, probably due to the absence of the pectoralis minor action on the scapula according to Umehara et al. [29]. The increased retraction in abduction and flexion it could be explained by a hypercontraction of the scapular stabilizers such as the trapezius and rhomboid muscles. Comparing the pathological side and the healthy side with a normal curve, the same trends were observed; therefore, patients do not make significant compensation with the healthy side compared to the general population. While a strong similarity between the average trends of the over 18 yo class and the bands of normality were found; the only evidences uncovered by the study were a reduced scapular Tilt both in flexion and abduction on the pathological side registered in under 18 yo group could be explained by the lack of adaptation by the muscle groups involved. We can hypothesize that the differences between the pathological side and the healthy side in under 18 yo is influenced by the lack of adaptation by the muscle groups involved. Indeed, adaption mechanism should be considered in order to explain why these differences are not shown in over 18 yo group respect to the normal band.

In conclusion, we observed altered scapular movement during anterior flexion and abduction in the patients with Poland syndrome. Based on the moderate effects on the patients RoM, we believe that the altered movement of the scapula is paraphysiological to compensate for the absence of the pectoralis major and minor muscles with other muscles. Further analysis could pinpoint the muscles involved in compensation to plan rehabilitation for patients with RoM deficits and for young patients in their developmental phase.

The limits of this study are: 1. the low number of patients analyzed; 2. not having used electromyographic analysis because surface electromyography analysis does not provide reliable results and because needle electromyography is difficult to perform due to lack of consent from patients; 3. the kinematic analysis with sensors did not allow to evaluate the movements of the scapula during rotations but only in anterior flexion and lateral abduction; 4. patients did not have an MRI examination to assess the presence of posterior glenoid labrum lesion or muscle hypertrophy. The point of strenght of the study is that is the first motion analyses study ever performed in absence of pectoralis major and minor muscles, in order to clarify their role in scapular motion.

Conclusion

Using clinical assessment and inertial motion sensors in the study of scapulohumeral kinematics of patients with PS, we observed that RoM is not affected by the absence of pectoralis muscles. Patients with this syndrome usually lead normal life and are able to engage in sports activities. We detected alterations in scapular kinematics in the pathological side that do not result in reduced force during shoulder movements, however, posterior instability seems to be constant. The presence of differences between the adult and young population suggests how compensatory mechanisms are established in these patients. Further analysis is needed to describe and study in detail the compensation mechanisms put in place by these patients.

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Declarations

Conflict of interest The authors declare no conflict of interest.

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